

Volume One

Self-Esteem, Coping and
Attribution in
Chronic Fatigue Syndrome

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CONTENTS

	Page Number
Abstract	8
Acknowledgements	10
List of abbreviations	11
CHAPTER ONE:	
INTRODUCTION 1: EPIDEMIOLOGY AND AETIOLOGY	12
1.1 Overview	12
1.2 Definitions	13
1.3 Epidemiology	16
1.4 Prognosis	19
1.5 The role of infection in CFS	20
1.6 The role of the immune system in CFS	21
1.7 The role of muscular function in CFS	22
1.8 Neuropsychology	22
1.9 Neuroimaging	24
1.10 Neuroendocrinology	25
1.11 The role of psychiatric disorders in CFS	26
CHAPTER TWO:	
INTRODUCTION 2: PSYCHOLOGICAL PROCESSES	34
2.1 Overview	34
2.2 Psychological mechanisms in CFS	34
2.3 Cognitive characteristics of CFS	35
2.4 Cognitive Behaviour Therapy for CFS	45

2.5 The role of defence mechanisms in CFS	51
2.6 Investigating core beliefs	56
2.7 Summary	63
2.8 Hypotheses	65
CHAPTER THREE: METHOD	66
3.1 Overview	66
3.2 Participants	66
3.3 Ethical considerations	69
3.4 Procedure	69
3.5 Measures	70
CHAPTER FOUR: RESULTS	82
4.1 Overview	82
4.2 Demographic information	83
4.3 Symptoms of Chronic Fatigue Syndrome	83
4.4 Testing the hypotheses	86
4.5 Further investigations	106
CHAPTER FIVE: DISCUSSION	111
5.1 Overview	111
5.2 Hypothesis one	112
5.3 Hypothesis two	114
5.4 Hypothesis three	119
5.5 Hypothesis four	123

5.6 Further investigations	124
5.7 Limitations of the present study	130
5.8 Implications for future research	134
5.9 Implications for clinical practice	136
5.10 Conclusions	139
CHAPTER SIX: REFERENCES	141
APPENDICES	163
1. The Green College, Oxford Definitions and Recommendations	163
2. The Pragmatic Inference Test	164
3. Information sheet for participants with CFS	170
4. Information sheet for healthy volunteers	171
5. Information sheet for participants with Diabetes	172
6. Consent forms for healthy volunteers	173
7. Consent form for participants with CFS or Diabetes	174
8. Letter to doctors working with patients with CFS	175
9. Letter to doctors working with patients with Diabetes	177
10. Letters to potential participants with CFS	179
11. Letters to potential participants with Diabetes	180
12. Letters of approval from Local Research Ethics Committees	181

LIST OF TABLES

	Page Number
1.1 Summary of research case definitions for Chronic Fatigue Syndrome	15
1.2 Epidemiology of Chronic Fatigue Syndrome	18
2.1 Outcome studies of CBT for CFS	50
2.2 The proportion of defensive high anxious participants in illness populations	55
3.1 Inclusion criteria for participants with CFS	68
3.2 Inclusion criteria for participants with Diabetes	68
3.3 Inclusion criteria for healthy participants	68
4.1 Demographic information	84
4.2 Symptoms reported by CFS participants	85
4.3 Illness attributions of participants with CFS: ‘Which one of the following best applies?’	85
4.4 Prevalence of depression and anxiety	89
4.5 Pearson correlations of length of illness, fatigue, anxiety and depression	90
4.6 Participants with CFS’ reports of guilt during a Major Depressive Episode:	90
4.7 Total ratings for positive and negative events on the Attribution Style Questionnaire (Parallel Form)	93
4.8 Number of positive and negative words endorsed on the Self-Referent Inferential Recall Test	95
4.9 Number of positive and negative words recalled on the Self-Referent Inferential Recall Test	98
4.10 Time to Colour-Name (Seconds) on the Emotional Stroop Test	99

4.11 Number of internalising responses on the Pragmatic Inference Test	102
4.12 Manifest Anxiety Scale and Marlowe-Crowne Social Desirability Scale scores	104
4.13 Coping styles	105
4.14 Summary of participants with CFS responses to ‘What do you think is the cause of your fatigue?’	107

LIST OF FIGURES

	Page Number
2.1 A complex specific model of chronic fatigue syndrome	49
4.1 Illness attribution and self-esteem amongst participants with CFS	87
4.2 Responses to the Attribution Style Questionnaire (Parallel Form)	92
4.3 Self-esteem and depression amongst participants with CFS, Diabetes and a healthy comparison group	94
4.4 Time to colour-name positive and negative words (seconds) on the Emotional Stroop Test	100
4.5 Coping styles	105
4.6 Internal attributions for positive and negative events on the Attribution Style Questionnaire (Parallel Form) and illness attributions of participants with CFS	108
4.7 Coping styles of CFS participants who have and have not experienced a Major Depressive Episode (MDE)	110
4.8 Discrepancies between internalising attributions on the Attribution Style Questionnaire (Parallel Form) and the Pragmatic Inference Test for CFS participants who have and have not experienced a Major Depressive Episode (MDE)	110

ABSTRACT

Chronic Fatigue Syndrome describes a disorder characterised by a principal complaint of fatigue accompanied by substantial functional impairment. The aetiology of CFS has been investigated from various perspectives, in particular with regards the presence of infectious diseases, immune system dysfunction, muscular dysfunction, neuropsychology and neurophysiology. Few consistent findings have, however, been reported. The high prevalence of psychiatric disorder amongst people with CFS, particularly of depression, has led investigators to examine the relationship between these factors. A cognitive behavioral model of CFS has been proposed (Sharpe, 1997) although evidence to support the various claims of the model is sparse, in particular relating to the hypothesis that people with CFS have low underlying levels of self-esteem which are protected by rigid defence mechanisms.

The present study investigates the hypothesis that CFS occurs in people who have low levels of self-esteem and an underlying depressogenic attribution style when they are confronted with a life stressor which is incompatible with their defence mechanisms, in particular a defensive coping style. A comparison group of people with a different chronic illness is included in the present study as well as a comparison group of healthy people.

Evidence was found to suggest that participants with CFS had lower underlying levels of self-esteem than was accounted for by depression and overtly expressed self-esteem. Physical illness attributions did not appear to be a likely candidate for a defensive mechanism, however a greater proportion of participants with CFS than comparison groups held a Defensive High Anxious coping style. Those who had never experienced a Major Depressive Episode were more likely to

hold a Repressive Coping style. These findings are consistent with the suggestion that individuals with CFS attempt to defend against low self-esteem but those who had experienced depression had been unable to do so completely.

Implications for clinical practice and future research are discussed. In particular, the findings highlight important similarities between neuroendocrinological findings in the study of defensive coping and CFS and emphasise the importance of a multi-disciplinary approach to understanding and treatment of CFS.

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LIST OF ABBREVIATIONS

ASQ (pf)	Attribution Style Questionnaire- Parallel Form
ASQ	Attribution Style Questionnaire
B-MAS	Bendig short form of the Manifest Anxiety Scale
CDC	Centers for Disease Control
CFS	Chronic Fatigue Syndrome
EST	Emotional Stroop Test
MAS	Manifest Anxiety Scale
MCSD	Marlowe-Crowne Social Desirability Scale
MDE	Major Depressive Episode
ODC	Oxford Diagnostic Criteria for Chronic Fatigue Syndrome
PIT	Pragmatic Inference Test
SADS	Schedule for Affective Disorder and Schizophrenia
SRIRT	Self-Referent Inferential Recall Test

CHAPTER ONE

INTRODUCTION 1:

EPIDEMIOLOGY AND AETIOLOGY

1.1 OVERVIEW

Although medical reports have described clinical syndromes of fatigue for at least two centuries, over the last decade considerable interest has developed around medical conditions characterised predominantly by fatigue for which there is no clear explanation (White, 1990). Various names have been used for these syndromes including Myalgic Encephalopathy (ME), Royal Free Disease, Fibromyalgia, Neurocirculatory Asthenia, Post-Viral Fatigue Syndrome (PVFS) and Chronic Fatigue Syndrome (CFS). From hereon the term Chronic Fatigue Syndrome will be used as it is a descriptive term which makes no further assumptions about the nature of the illness, which is most appropriate at the present time when there is little consensus regarding the aetiology of the syndrome.

This chapter begins with an overview of the current research definitions of CFS and the difficulties associated with making a diagnosis of CFS. How these difficulties impact upon studies of prevalence and prognosis will then be discussed. The following sections evaluate various hypothesised mechanisms that may be associated with CFS. Chapter Two describes theories relating to the role of psychological mechanisms in CFS. Based on this discussion, the hypotheses of the present study will be outlined.

1.2 DEFINITIONS

As there are currently no specific laboratory test abnormalities associated with CFS, the diagnosis is based on clinical presentation and the exclusion of other disorders. Operational case definitions for CFS have been proposed by three research groups: the Centers for Disease Control in the United States of America (Holmes, Kaplan, Gantz, Komaroff, Shonberger *et al*, 1988), an Australian group (Lloyd, Wakefield, Boughton and Dwyer, 1988) and the Green College, Oxford group from the United Kingdom (Sharpe, Archard, Banatvala, Borysiewicz, Clare *et al*, 1991). The UK Patient Organisation (1993) has also proposed a definition for ME/PVFS (Dowsett, Goudsmit, Macintyre, Shepherd. *et al*, 1994). These are summarised in Table 1.1. The main similarities and differences between these will be briefly reviewed and the difficulties associated with providing a comprehensive case definition of CFS are discussed.

The definitions all have in common the presence of a principal complaint of fatigue that has been present for at least six months and is accompanied by substantial functional impairment. The definitions differ most with regards the emphasis that is put on somatic symptoms and signs and the exclusion of psychiatric problems. The presence of a greater number of somatic symptoms has been found to increase the probability of the presence of psychiatric disorder (Katon and Russo, 1992) and lower estimates of the prevalence of CFS (e.g. Gunn, Connell and Randall, 1993). This appears, however, to leave a large number of people with unexplained chronic fatigue undiagnosed. On the other hand, if the definition lacks specificity it becomes impossible to distinguish between CFS and other disorders. Section 1.1 discusses difficulties in distinguishing between diagnoses of CFS and depression. It is likely that the lack of diagnostic specificity arises in part because CFS lies at the

extreme end along a continuum of fatigue states, rather than being a distinct syndrome (Wessely, Hotopf and Sharpe, 1998). Preliminary evidence for this view comes from population studies of fatigue in which general practice patients report a continuous distribution of fatigue symptoms (David, Pelosi, McDonald, Stephens, Ledger *et al*, 1990). These difficulties must be borne in mind when considering studies of the epidemiology of CFS.

None of the symptoms included in the definitions of CFS are specific to it and few symptoms are able to discriminate CFS from other disorders (Komaroff, Faglioli, Geiger, Doolittle, Lee *et al*, 1996). The Australian definition is the only one, to date, to include a laboratory marker in the diagnosis of CFS. Although this may increase the specificity of the diagnosis these criteria have been criticised for being premature due to the lack of replicability of a specific pattern of putative immune impairment and unknown functional significance of immune abnormalities (Demitrack and Abbey, 1996).

For the purposes of the present study the Green College, Oxford, criteria will be adopted as this definition has been used most widely within the British population, since being developed in 1991. These criteria also do not make any assumptions about the psychiatric status of people with CFS, which is imperative for this study which will be considering the relationship between CFS and psychiatric disorder. These criteria are given in full in Appendix One.

Table 1.1
Summary of research case definitions for Chronic Fatigue Syndrome

	CDC	CDC-revised	Australian	U.K.	London
	Holmes <i>et al</i> (1988)	Fukuda <i>et al</i> (1994)	Lloyd <i>et al</i> (1988)	Sharpe <i>et al</i> (1991)	Dowsett. <i>et al.</i> (1994)
Minimum duration (months)	6	6	6	6	6
Functional impairment	50% decrease in activity	Substantial	Substantial	Disabling	Fluctuates
Cognitive/ neuropsychiatric symptoms	May be present	May be present	Required	Mental fatigue required	Required
Other symptoms	6 or 8 required	4 required	Not specified	Not specified	Not required, although common
New onset	Required	Required	Not required	Required	Not required
Medical exclusions	Extensive list of known physical causes	Clinically important	Known physical causes	Known physical causes	Not specified
Psychiatric exclusions	Endogenous depression, hysterical personality disorder, anxiety neurosis, schizophrenia	Melancholic depression, substance abuse, bipolar disorders, psychosis, eating disorder	Psychosis, bipolar, substance abuse, eating disorder	Psychosis, bipolar, eating disorder, organic brain disease	Not specified

Adapted from Wessely, Hotopf and Sharpe (1998)

1.3 EPIDEMIOLOGY

The difficulty in defining CFS has complicated the epidemiological study of CFS. Furthermore, results have varied widely due to the different populations studied and the use of a variety of methodologies (see Table 1.2).

Table 1.2 includes the results of three studies that have used GPs as key informants of the prevalence of CFS. Clearly, the results vary widely. Although this may reflect differences in the prevalence rate of CFS in different parts of the world, there are limitations in the key informant method. In particular the studies rely on people who are suffering from CFS actually presenting to their GP, the GP recognising a case as CFS and the GP remembering all the affected patients at the time of completing the survey.

An alternative to using GPs as key informants is to target practitioners who are likely to receive referrals of patients with CFS. Two studies that used this method are described in Table 1.2. This method however retains many of the difficulties in surveying General Practitioners as the patient must have first been referred to that clinic, most probably by his or her GP. Furthermore patients who present to specialist services have been found to have different characteristics to primary care patients, including a higher proportion being categorised as Social Class 1 and a greater tendency to make physical attributions for illness (Euba, Chalder, Deale and Wessely, 1996).

More systematic population surveys are now beginning to be carried out, however there are still various limitations involved. Rates of refusal to participate in time consuming assessments tend to be high (Buchwald, Umali, Umali, Kith, Pearlman *et al*, 1995; Bates, Schmitt, Buchwald, Ware, Lee *et al*, 1993). Prevalence figures are affected by whether or not it is assumed that non-participants have the

same rate of CFS as participants. One useful aspect of some of these studies, however, is the investigation of multiple case definitions within a single survey. This enables more direct comparison between different studies. Wessely, Chalder, Hirsch, Wallace and Wright (1997), for example, carried out a prospective study of primary care patients in England. The point prevalence of CFS varied between 0.8% and 2.6% depending on the case definition used. If comorbid psychiatric disorders were excluded the rate fell to 0.5%.

The majority of studies report an excess of females among patients with CFS, which is maintained after adjustment for comorbid psychological disorder (Wessely *et al*, 1997). This excess is more modest in community studies in which estimates of the relative risk for women vary between 1.3 and 1.7 (David, Pelosi, McDonald, Stephens, Ledger *et al*, 1990; Cathebras, Robbins, Kirmayer and Hayton, 1992; Fuhrer and Wessely, 1995).

In summary, the prevalence rates of CFS found vary widely, from less than one per 100,000 in a Japanese hospital based study (Minowa and Jiamo, 1996) to 2,600 per 100,000 in a British community study (Wessely *et al*, 1997). This variation appears to relate to the definition adopted, methodology used and population surveyed. Generally, it appears that prevalence figures have increased in more recent studies, reflecting the application of more community sampling techniques and perhaps greater acceptance of the label 'CFS' by physicians and patients. With these difficulties in defining CFS and, hence, selecting participants for studies in mind, the following section reviews current literature regarding the prognosis of CFS.

Table 1.2
Epidemiology of Chronic Fatigue Syndrome

Study	Location	Case definition	Method	Point prevalence	Comments
Lloyd <i>et al</i> (1990)	Australia	Australian	GP key informants	'at least' 37.1 per 100,000	Low (25%) response rate from GPs.
Ho Yen and McNamara (1991)	Scotland	Similar to UK criteria but 3 months minimum duration (Ho-Yen 1990)	GP key informant	130 per 100,000 (range 30-270 per 100,000).	91% response rate by GPs. Variable recognition of CFS by GPs
Bazelmans <i>et al</i> (1997)	Netherlands	Minimum duration 1 year	GP key informant	112 per 100,000	60% response rate by GPs.
Minowa and Jiamo (1996)	Japan	Japanese version of CDC	Key informants from various clinical departments (internal medicine, paediatrics, psychiatry, neurology)	Period prevalence 0.85 per 100,000 during one year	52.5% response rate from hospital departments.
Gunn, Connell and Randall (1993)	USA	CDC	Key informants from various clinical departments (internal medicine, infectious diseases, rheumatology, neurology)	2 yr prevalence 2-7.3 per 100,000 pro-rated 4.6-11.3 per 100,000	Psychiatry not surveyed.
Price <i>et al</i> (1992)	USA	CDC (approx.)	Reviewed Epidemiology Catchment Area database	7.4 per 100,000	Used restrictive 1988 criteria. Based on one identified case.
Bates <i>et al</i> (1993)	USA	CDC (1988) UK Australian	Survey of Ambulatory Care Clinic	CDC 300 UK 400 Aus 1000 per 100,000	69% refused/unavailable for assessment.
Lawrie and Pelosi (1995)	Scotland	UK	Survey of GP registered patients	560 per 100,000	Based on 4 identified cases.
Buchwald <i>et al</i> (1995)	USA	CDC (1988) with 1992 guidelines	Random sample from health maintenance organisation	75-267 per 100,000 depending on assumptions about non-participants	Based on 3 identified cases. 63% refused/unavailable for assessment.
Wessely <i>et al</i> (1997)	UK	CDC (1988) CDC (1994) ODC Australian	Prospective primary care survey	CDC (88) 1200 CDC (94) 2600 Exclude psychiatric comorbidity 500 ODC 2200 Aus 1400 per 100,000	Only reviewed 18-45 year olds

1.4 PROGNOSIS

Various studies have confirmed that CFS is, indeed, a chronic condition, although studies differ with regards the prognosis of CFS depending on the selection criteria adopted. Formal follow-up studies from specialist clinics have tended to be fairly pessimistic about the prognosis of CFS. Two studies carried out in the USA found that although half of patients referred had significantly improved one year after assessment, only 6% of patients no longer experienced symptoms (Hellinger, Smith, Van Scoy, Soitzer, Forgacs *et al*, 1988; Gold, Bowden, Sixbey, Riggs, Katon *et al*, 1990). In Britain, a systematic follow-up of patients with CFS seen in an infectious diseases clinic found that two-thirds of patients had improved to some degree three years after initial assessment and only 13% had 'fully recovered' (Sharpe, Hawton, Seagroatt and Pasvol, 1992). The prognosis for children has tended to be better. From a review of outcome studies Joyce, Hotopf and Wessely (1997) reported that studies report definite improvement or full recovery for between 54 and 94% of children with CFS.

The poor prognosis associated with CFS for patients seen in specialist clinics may not be surprising given the inevitable selection bias in those that have been referred on for specialist treatment. The majority of these patients are likely to have been ill for many years before being seen at the clinic. Follow-up studies in primary care settings have tended to report better prognosis, however reports remain fairly pessimistic. Joyce *et al* (1997) merged comparable studies of outcome of CFS patients in primary care and reported that only 32% were improved or recovered one year later.

Among patients with CFS, only a small number (<5%) develop new diagnoses in the years following a diagnosis of CFS, suggesting that CFS does not

increase the risk of developing other conditions or act as a prodrome to another illness (Joyce *et al*, 1997). There also does not appear to be an effect on morbidity, except possibly for death by suicide (Wessely, Hotopf and Sharpe, 1998). Various studies have attempted to assess factors associated with outcome, with varying degrees of consistency. Factors which may be associated with poor prognosis include older age at onset, comorbid psychiatric disorder, attribution of illness to physical factors, more severe illness, avoidance of alcohol, belonging to a self-help group and changing or leaving employment (Joyce *et al*, 1997). The role of various psychological factors in prognosis is encouraging with regards the potential application of psychological interventions with people with CFS. This point will be returned to in Section 2.3.

The search for physical markers of CFS to date from studies of infection, immunity, muscular, neuropsychological, neuroanatomical and neuroendocrinological function will now be reviewed.

1.5 THE ROLE OF INFECTION IN CFS

The large number of patients with CFS who report the onset of their CFS being preceded by viral infection (e.g. Wessely and Powell, 1989) has encouraged the study of infectious agents in CFS. Claims have been made for the presence of various viral agents including Epstein Barr Virus (EBV), Human Herpes Virus-6 (HHV-6) and enteroviruses. To date, however, there has been no evidence that a single virus infects all patients with CFS, and fatigue syndromes identical to CFS have been described following infection by many different viral agents (Lloyd *et al*, 1990). Although in the case of some viruses there may be a distinct post-infectious fatigue syndrome, for example, following glandular fever (White, Thomas, Amess,

Grover, Kangro *et al*, 1995), in many cases there does not appear to be a clear link between viral infection and CFS. Wessely, Chalder, Hirsch, Pawlikowska, Wallace *et al* (1995) carried out a prospective study of over 1000 primary care patients with common clinical viral infections and found no association between infection and the later development of CFS. In conclusion, although fatigue syndromes do seem to develop after certain infections it seems most likely that CFS develops as a non-specific response to a wide range of infectious agents (Wessely, 1998).

1.6 THE ROLE OF THE IMMUNE SYSTEM IN CFS

The possibility that chronic fatigue syndrome may be precipitated by viral infection suggests that there may be a role of the immune system in CFS. Various studies have provided evidence of some laboratory abnormalities, in particular regarding subsets of T-cells and natural killer cells (Klimas, Salvato, Morgan and Fletcher, 1990), however these findings have not been confirmed by all studies (Peakman, Deale, Field, Mahalingam, and Wessely, 1997). The findings are also relatively nonspecific to CFS (Buchwald and Komaroff, 1991), are unrelated to the main clinical features of CFS and outcome (Peakman *et al*, 1997) and are confounded by other factors including inactivity, psychiatric morbidity, sleep disturbance, drug therapy, stress and demoralisation (Strober, 1994). Furthermore, studies vary in sample sizes used, selection criteria and comparison groups and use a variety of laboratory techniques. In conclusion, recent, detailed reviews have asserted that it is currently impossible to determine the significance of the observed changes in immunological status in CFS (Wessely, Hotopf and Sharpe, 1998).

1.7 THE ROLE OF MUSCULAR FUNCTION IN CFS

One of the primary clinical features of CFS is muscle pain and a variety of minor biochemical and structural abnormalities of muscle have been found (summarised by Lane, Barrett, Woodrow, Moss, Fletcher *et al*, 1998). However the studies are limited by the lack of appropriate comparison groups and are confounded by factors including inactivity and deconditioning. Lloyd, Gandevia, Brockman, Hales and Wakefield (1994) found that dynamic muscle function, muscle strength, endurance and fatiguability are normal after inactivity has been controlled for. It is possible that a subgroup of patients (ten per cent) with CFS may have primary neuromuscular disturbance (Wessely and Powell, 1989). These cases are likely to be those in which fatigue is exercise related and not experienced at rest. Other patients experience muscle pain at rest and immediately after waking. This cannot be accounted for by any known mechanism of muscle function (Edwards, Newham and Peters, 1991). Furthermore, a large number of patients with CFS, also experience mental fatigue and neuropsychological symptoms, such as poor concentration and short-term memory, which cannot be explained by a muscle defect (Wessely, Hotopf and Sharpe, 1998).

1.8 NEUROPSYCHOLOGY

Patients with CFS frequently report cognitive disturbance. Common complaints include forgetfulness, distractibility or decreased concentration, and impaired reasoning ability (Tiersky, Johnson, Lange, Natelson and DeLuca, 1997). Such complaints are more frequent and severe than in other clinical populations, including multiple sclerosis and affective disorders (DeLuca, Johnson, Beldowicz and Natelson, 1995).

Studies of general intellectual functioning have tended to find group means of average to above-average levels (Tiersky *et al*, 1997). With more specific tests of cognitive functioning, the results are more inconsistent. This is likely to result from selection biases, small sample sizes, different criteria for matching control groups, a variety of tests being used and different case criteria for CFS being adopted. From a detailed review of the literature, Tiersky and colleagues (1997) conclude that:

1. In general CFS patients perform within normal limits on simple tests of attention/concentration, although more difficulties are experienced with increased task complexity suggesting difficulties relating to slowed processing speed and/or efficiency.
2. Most studies of learning and memory do not find deficits in recall of information from long-term storage, although there is some evidence for decreased acquisition. There is some evidence to suggest patients with CFS may have difficulty learning and recalling semantically complex material.
3. Studies of higher order skills, including planning, set-shifting, verbal fluency and concept formation, have generally reported unimpaired performance by people with CFS.

The most consistent difficulty found appears to relate to effortful tasks. The reasons why this might be so are difficult to ascertain as these tasks can be impaired by anything that reduces attentional capacity, such as age, arousal, mood and ill health (Hasher and Zacks, 1979). Sleep disturbance (Smith, Pollock, Thomas, Llewelyn, and Borysiewicz, 1996) and low activity levels (Vercoulen *et al*, 1997) are particularly prevalent in CFS and may influence performance on objective measures. DeLuca, Johnson, Ellis and Natelson (1997) included a direct comparison between patients with CFS and no history of psychiatric disorder and CFS patients with a

concurrent Axis 1 diagnosis. Those CFS patients with no history of psychiatric disorder still exhibited significant neuropsychological impairment. They concluded that, although depression may influence neuropsychological performance it can not entirely account for the difficulties found.

In summary, formal test results appear to support objective cognitive difficulties among patients with CFS, in particular relating to speed or efficiency of processing. These objective difficulties are not, however, consistent with the broad range and severity of subjective cognitive complaints reported by patients with CFS. Test performance may be influenced by a number of potential confounding variables, and it is essential for future studies to undertake to reduce the influence of these variables, in particular by using clinical control groups and screening for conditions that may impact on cognitive functioning.

1.9 NEUROIMAGING

Recently studies of CFS have been conducted using structural and functional brain imaging techniques. Tiersky *et al* (1997) conclude that magnetic resonance imaging (MRI) has generally shown abnormalities in the cerebral white matter of patients with CFS. Similar abnormalities have, however, been found in controls and participants with depressive disorder (Dupont, Jernigan, Heindel, Butters, Shafer *et al*, 1995). Functional imaging studies have found no consistent pattern of abnormal cerebral blood flow that is characteristic of CFS (Ichise, Salit, Abbey, Chung, Gray *et al*, 1992, Goldstein, Mena, Jouanne and Lesser, 1995). A number of studies have failed to find differences between CFS patients and depressed controls (e.g. Goldstein *et al*, 1995), although, as in CFS, there is great heterogeneity amongst psychiatric controls.

1.10 NEUROENDOCRINOLOGY

Neuroendocrinology research has identified a potential candidate for an abnormal physiological response to stress amongst patients with CFS related to reductions in hypothalamic-pituitary-adrenal (HPA) axis functioning and neurotransmission. More specifically, low levels of cortisol and increased serotonin neurotransmitter function have been reported in CFS by an increasing number of authors (Demitrack, Dale, Straus, Laue, Listwak *et al*, 1991; Bakheit, Behan, Dinan, Gray and O'Keane, 1992; Cleare, Bearn, Allain, McGregor, Wessely *et al*, 1995). These findings are the opposite of those found in depressed participants, i.e. increased levels of cortisol and decreased serotonin neurotransmitter function (Cleare *et al*, 1995). It is unclear, however, how these findings account for patients who experience both CFS and depression. Yatham, Morehouse, Chisholm, Haase, MacDonald *et al* (1995) compared CFS patients, 10/11 of whom had a history of psychiatric disorder, with healthy controls and found no differences in cortisol release. Again, a number of confounding variables confuse interpretation of the literature, including selection biases and the effect of weight loss and sleep disturbance (Wessely, Hotopf and Sharpe, 1998).

The association between CFS and affective disorder, particularly depression, is far from clear from studies of the neuropsychology, neuroimaging and neuroendocrinology of CFS. Although, taken as a whole, the neuropsychology and neuroendocrinology reports appear to support separate underlying processes in CFS and affective disorder, the neuroimaging studies have identified a number of similarities. As discussed by Wessely, Hotopf and Sharpe (1998), although it is important to distinguish the differences between CFS and depression, the equally

important similarities may be left unexplained. Section 1.11.1 will discuss the complex relationship between CFS and depression in greater detail.

1.11 THE ROLE OF PSYCHIATRIC DISORDERS IN CFS

A number of studies have suggested that there is a significant relationship between CFS and psychiatric disorder. The nature of this relationship, however, remains unclear. Katon and Walker (1993) reviewed studies from community, primary care and tertiary care settings and concluded that, no matter which population is studied, there is an increased prevalence of past and current psychiatric disorder amongst people with CFS. A recent prospective study compared the rates of psychiatric disorder amongst people who went on to develop chronic fatigue or CFS with healthy controls in a primary care setting (Wessely, Chalder, Hirsch, Wallace and Wright, 1996). 75% of those people who developed CFS were suffering from psychiatric disorder, compared to 60% of those with chronic fatigue (that did not fulfil research criteria for CFS) and 19% of healthy controls. The most prevalent psychiatric disorder was depression. 81% of people who developed CFS were probable cases of depression, compared to 55% of the people with chronic fatigue and 44% of the control group, having removed the presence of fatigue from the case criteria of depression. Those who were fatigued were also more likely to have visited their GP for emotional reasons or had been prescribed psychotropic medication in the past. The high rate of comorbidity of depression and CFS has been found in community, primary care and specialist settings (Wessely, Hotopf and Sharpe, 1998). Other studies have also suggested that there may be increased rates of other psychiatric disorders in CFS, in particular anxiety and somatisation disorder (Wessely, Hotopf and Sharpe, 1998). The following section addresses the nature of the relationship between depression and CFS. Less research has been carried out in

to the relationship between anxiety disorders and CFS, this will be reviewed in Section 1.11.2.

1.11.1 CFS and Depression

The vast majority of studies which have investigated the comorbidity of depression and CFS have identified relatively high rates of current and lifetime depression amongst people with CFS (Abbey, 1996). As mentioned earlier, however, it is important that studies take in to account the increased risk of psychiatric disorder associated with the presence of medical illness. 6-31% of patients attending outpatient medical clinics have been reported to be depressed (Glass, Allan, Uhlenhuth, Kimball and Borinstein, 1978; Cleary, Goldberg, Kessler, and Nycz, 1982). Taerk, Toner, Salit, Garfinkel and Ozersky (1987) carried out standardised psychiatric interviews with patients with CFS, neuromuscular disease and healthy volunteers. The CFS group continued to have an increased rate of current and lifetime depression, even when compared to the other chronically ill group who shared a number of symptoms. Similarly, Wessely and Powell (1989) found that 43% of CFS patients had a previous history of depression compared to 30% of patients with neuromuscular disease and 64% of depressed controls, and 72% of the CFS patients were currently depressed compared to 36% of the neuromuscular disease group. These figures represent the prevalence when fatigue is excluded from the diagnostic criteria for depression. Furthermore, when physical symptoms alone were taken in to account 47% of the CFS patients were indistinguishable from the depressed group. Abbey and Garfinkel (1991) have proposed four possible models of the relationship between CFS and depression, which are discussed in turn.

1.11.1.(a) Depression is primary to CFS

According to this model CFS is seen as an atypical presentation of depression, or alternatively, depression may be seen to complicate recovery from physical illness. There is clearly an overlap between the symptoms of depression and CFS in particular reduction of energy and decreased levels of activity. Katon, Kleinman and Rosen (1982) have suggested that people may report somatic symptoms rather than cognitive and affective symptoms of depression because of an inability to describe emotional states, or due to the presence of protective psychological defence structures or due to socio-cultural pressures. If CFS were, however, equivalent to depression it would be necessary for the various findings associated with CFS to be accounted for by the aetiology of depression.

Unfortunately, as discussed earlier, there remains much inconsistency in the CFS research with regards the role of infection, the immune system, muscular function and neuropsychology, neurophysiology and neuroendocrinology.

Could current knowledge about depression, however, explain the possible roles of these factors if CFS were primarily a depressive illness? With regards infection and immunity, anecdotal reports and clinical research have suggested that episodes of major depression are associated with an increased risk of illness associated with immune system competence (Dorian and Garfinkel, 1987). Schleifer, Keller, Bond, Cohen, and Stein (1989) concluded that although altered immunity does not appear to be a specific correlate of major depressive disorder it may occur in some subtypes of depression. Further research is necessary to determine for which subtypes of depression this may be the case. With regards muscular function and depression, currently little research has been carried out (Abbey and Garfinkel, 1991). Although imaging studies have failed to find consistent differences between

patients with CFS and depressed controls (e.g. Goldstein *et al*, 1995), neuropsychology studies suggest that although depression may influence performance to some extent, it can not entirely account for the difficulties found (deLuca *et al*, 1997) (see Section 1.8). Finally, although the HPA axis has been implicated in both depression and CFS, it appears to have an opposite relationship with the two disorders, and it's role remains unclear (Wessely, Hotopf and Sharpe, 1998).

In summary, although certain features of depression and CFS suggest the presence of a close relationship between the two disorders, the evidence to date does not provide consistent evidence that CFS is an atypical presentation of depression.

A classic study by Imboden, Canter and Cluff (1961) demonstrated that the presence of psychiatric illness could complicate recovery from illness. In their prospective study those people who recovered more slowly from influenza had been identified as having a propensity to become depressed prior to contracting the influenza virus. That people with CFS may be vulnerable to becoming depressed has been suggested by anecdotal and retrospective reports that suggest that patients with CFS are achievement-orientated and goal-driven (e.g. Salit, 1985; Ware, 1993). These characteristics might be expected to be associated with a vulnerability to depression as they tend to be a means of stabilising self-esteem and experiencing pleasure (Beck, 1983). Powell, Dolan and Wessely (1989) suggest that a tendency to make physical attributions for symptoms in CFS may represent a defence against low self-esteem. Formal investigations have yet to identify particular personality styles in people with CFS (e.g. Johnson, DeLuca and Natelson, 1996; Wood and Wessely, 1999).

1.11.1 (b) Depression is secondary to CFS

Abbey and Garfinkel (1991) suggest two ways in which depression may arise as a result of CFS, first because of an organic mental syndrome and second as a reaction to the presence of CFS. An organic mental syndrome is a mood disturbance caused by a specific organic factor. Many toxic and metabolic factors appear to lead to organic mental syndromes, including, medications, endocrine disorders, structural disease of the central nervous system and viral illnesses (Abbey and Garfinkel, 1991). Organic mental syndromes are also often associated with a mild degree of cognitive disturbance (APA, 1987). How CFS would cause an organic mental syndrome is currently unclear. It has been suggested that it may relate to the presence of circulating interferon and other cytokines induced by viral infection as the therapeutic use of interferon has been reported to induce a clinical syndrome of extreme fatigue, depression and cognitive symptoms (Jones and Miller, 1987). The validity of extrapolating from this data, however, is not certain (Abbey and Garfinkel, 1991).

Depression has been found to be a common psychological response to major illness, for example, amongst patients with cancer, strokes and heart disease, particularly if the illness has a chronic course (Ray, 1991). Furthermore, the impact of CFS is likely to reduce levels of activity, exercise and certainty about the future, which are all likely to influence a person's mood and opportunities to gain rewards in their life. Ware (1992) has described the distressing effects of the treatment that people with CFS often encounter from other people due to the unclear nature of their illness. 90% of the people with chronic fatigue studied reported having experienced delegitimation, in particular, trivialisation of symptoms by others or being labelled as 'psychosomatic', which often led to feelings of self-doubt, humiliation and shame.

As mentioned earlier, however, even when compared to other chronically ill groups there is an increased prevalence of depression amongst patients with CFS and depression often predates the onset of CFS. This is also true when CFS patients are compared to a group with another poorly understood chronic condition, chronic pain (Blakely, Howard, Sosich, Murdoch, Menkes, *et al*, 1991). This suggests that it is unlikely that the high rate of depression in CFS is simply a response to the presence of chronic illness.

1.11.1 (c) Depression is concurrent or coincident with CFS

According to this model depression and CFS may be distinct outcomes of a particular cause. For example, a viral cause of CFS may also lead to depression. A number of viruses are neurotropic and may act on the central nervous system producing subtle biochemical or receptor lesions in the system which may be responsible for affect regulation (Abbey and Garfinkel, 1991). At this stage this is purely speculative.

1.11.1 (d) Depression is an artefact of CFS

According to this model, due to the overlap of symptoms in the definitions of depression and CFS, symptoms of CFS may be inappropriately attributed to depression. A number of studies have, however, now eliminated the overlapping criteria when diagnosing depression amongst patients with CFS and have continued to find an increased prevalence of depression in CFS (e.g. Kruesi, Dale, and Straus, 1989; Wessely and Powell, 1989; Wood, Bentall, Gopfert and Edwards, 1991).

1.11.2 Anxiety and CFS

A lot less attention has been paid in the literature to the association between CFS and anxiety disorders. Over the last ten years, incidence of anxiety disorders in CFS of between 13 and 20 per cent have been described (Manu, Mathews and Lane, 1991; Farmer, Jones, Hillier, Llewelyn, Borysiewicz *et al*, 1995). Fischler, Cluydts, De Gucht, Kaufman and DeMeirleir (1997) found the prevalence of Generalised Anxiety Disorder amongst CFS participants was eight times that of medical controls. The same considerations apply to the possible relationship between anxiety and CFS as to depression and CFS. In particular whether anxiety disorders are primary or secondary to CFS, or are coincident with or an artefact of CFS. Generalised Anxiety Disorder and CFS share a number of diagnostic criteria, including easy fatigue, difficulty concentrating and muscle tension. People with CFS also often report avoidance behaviour as a means of reducing physical symptoms (Deale, Chalder and Wessely, 1998). At this stage, the relationships between CFS and anxiety disorders warrants further investigation.

A number of difficulties are apparent when attempting to draw conclusions about the relationship between psychiatric disorders and CFS. In particular, no study has found that *all* CFS patients suffer from psychiatric disorder. The difficulties may be compounded as CFS, depression and anxiety are heterogeneous in terms of severity, degree of disability and duration. Furthermore, depression and anxiety are multifaceted, involving cognitive, emotional and somatic components. Although their group found little difference between the physical symptoms of CFS and depression, Powell, Dolan and Wessely (1989) reported that the cognitive

components of depression, such as guilt and low self-esteem, were not found in CFS.

Johnson, DeLuca and Natelson (1996) reported similar findings.

Ray (1991) concludes that any working model of CFS should recognise that

- (i) CFS may be a nonspecific response, with multiple somatic and psychological causes.
- (ii) The relationship between the causes may be interactive, rather than additive.
- (iii) Variables may have a reciprocal influence on each other, leading to vicious cycles developing and further reducing the level of functioning.
- (iv) CFS may be a heterogeneous condition.

With this in mind, this study will address the hypothesis that people with CFS may have a vulnerability to psychiatric illness. This might prolong recovery from illness, and perhaps leads to the development of vicious cycles in which the presence of psychiatric disorder contributes to the maintenance of the illness. It has been suggested that this vulnerability may relate to cognitive mechanisms that are in place to defend against low self-esteem. The following chapter examines the current literature on the potential psychological mechanisms that may predispose a person to developing CFS.

CHAPTER TWO

INTRODUCTION 2:

PSYCHOLOGICAL PROCESSES

2.1 OVERVIEW

A cognitive-behavioural model of CFS, derived primarily from clinical and anecdotal reports, will be described. Although this model has successfully driven treatment for CFS, the evidence for many of its assertions is scarce. This chapter describes the existing evidence for this model and the hypotheses of the present study.

2.2 PSYCHOLOGICAL MECHANISMS IN CFS

Figure 2.1 reproduces the cognitive behavioural model of CFS proposed by Sharpe (1997) which describes potential predisposing factors for CFS. The model suggests that a core belief reflecting low self esteem such as 'I am inadequate' leads to the development of rigidly held beliefs, such as 'I must always perform perfectly', 'I must never show weakness' and a lifestyle characterised by hard work, putting on a brave face and not asking for help. A triggering event such as stress or an acute illness, which would lead to symptoms such as fatigue, muscle pain and difficulty in functioning, would then be interpreted as signifying physical illness outside of personal control. This would be associated with feelings of distress and frustration, behaviour characterised by extreme rest with episodic bursts of activity and subsequent physiological effects of inactivity and emotional distress. The model proposes that a vicious cycle develops in which thoughts, mood, behaviour and physiology perpetuate symptoms, which then perpetuate the developing pattern of thoughts, mood, behaviour and physiology. Evidence relating to the cognitive aspects

of the model comes from two main sources: first, direct empirical investigation of specific aspects of the model. Second, indirect evidence based on the efficacy of therapy guided by the model. These will be discussed in turn. The following section addresses studies that have looked directly at cognitive factors in relation to CFS.

2.3 COGNITIVE CHARACTERISTICS OF CFS

In order to evaluate the existing evidence for the cognitive components of Sharpe's (1997) model of CFS, the four hypotheses relating to cognitive characteristics will be discussed in turn: (i) Core beliefs of inadequacy; (ii) Beliefs associated with perfectionism; (iii) Lifestyles characterised by hard work and striving for achievement; and (iv) Thoughts associated with a physical basis for the illness. Whilst the first three items are hypothesised to have a predisposing role in CFS, thoughts associated with a physical basis for the illness are hypothesised to have a maintaining role in the illness.

2.3.1 Core beliefs of inadequacy

Rather than suggesting that people with CFS hold core beliefs of inadequacy and low self-esteem, studies which have directly addressed this hypothesis have tended to find quite the opposite. Even when people with post-infectious syndromes and CFS fulfil diagnostic criteria for depression they tend to report few feelings of guilt and preserved self esteem (Imboden, Canter and Cluff, 1959; Johnson, DeLuca and Natelson, 1996; Powell, Dolan and Wessely, 1990). In their description of common themes that arise when carrying out therapy with people with CFS, Surawy, Hackmann, Hawton and Sharpe (1995) note a relative lack of expressed distress by participants. Similarly, a number of studies report participants' self-descriptions as

'not the sort of person to become depressed' (Surawy *et al*, 1995; Chalder, Power and Wessely, 1996; Powell, Dolan and Wessely, 1990; Katz and Andiman, 1988).

Two studies have found reduced levels of self-esteem amongst people with CFS who were also depressed (Johnson, Paananen, Rahinantti, and Hannonen, 1997; Moss-Morris and Petrie, 1996). Moss-Morris and Petrie (1996) concluded, however, that on the whole people with CFS have good self-esteem unless they are depressed. Similarly, Johnson *et al* (1997) found no significant differences in levels of self-esteem between CFS participants and healthy controls and controls with rheumatoid arthritis. They did report, however, a non-significant tendency for depressed CFS participants to have lower basic self-esteem, but enhanced earned self-esteem (a sense of self-worth that is earned by competence). The evidence to date, therefore is inconsistent regarding levels of self-esteem amongst people with CFS who are also depressed, but appears to consistently report that self-esteem is intact amongst people with CFS who are not depressed.

Two studies have looked at factors associated with self-esteem. Findley, Kerns, Weinberg and Rosenberg, (1998) investigated the concept of self-efficacy amongst people with CFS. Numerous studies have identified a strong link between self-efficacy, an individual's belief that he or she is able to execute behaviours necessary to obtain particular outcomes, and sense of self-worth or self-esteem (e.g. Bandura, 1977). Findley *et al* (1998) reported that a significant, but small, proportion of the variance in symptoms, disability and distress was accounted for by self-efficacy. Higher levels of self-efficacy were independently related to less intense somatic symptoms, psychiatric symptoms, total constellation of symptoms and disability and distress. If the usual association between self-efficacy and self-esteem applies amongst people with CFS we might anticipate a similar relationship between

self-esteem and symptomatology, although, the direction of causality would remain unclear.

Various studies have suggested that an individual's self-esteem is established to a large extent by experiences of relationships during childhood (e.g. Cotton, 1983). Ware (1993) reported that approximately half of the CFS participants that she interviewed reported significant difficulties during childhood, with 44% reporting chronic tension or fighting within the family. These high figures were reported despite these topics not being directly brought up by the interviewer, suggesting that they may well reflect an underestimation of the prevalence of difficult childhood experiences amongst people with CFS, although the lack of a control group prevents firm conclusions. A recent study by Fischer and Chalder (in press) found that adult participants with CFS reported that their mothers had been highly protective but less caring than mothers of the medical comparison group.

At this stage, there is more evidence to suggest that self-esteem is likely to be intact than there is to suggest that patients have core beliefs of inadequacy. Moss-Morris (1997) draws attention, however, to the limitations of research to date which has restricted itself to the investigation of conscious cognitive processes, and, hence, is vulnerable to response bias. This issue will be returned to in Section 2.6.

2.3.2. Beliefs associated with perfectionism

Perfectionism has been defined as 'the practice of demanding of oneself or others a higher quality of performance than is required by the situation' (Hollender, 1965). With regards CFS, reports of perfectionism have been anecdotal. Ware and Kleinman (1992), for example, noted from their clinical work that there appeared to be a tendency for exacting standards for personal performance amongst their patients. Similarly, Surawy *et al* (1995) noted that perfectionism was common

amongst their patient population. Ware (1993) found that many of her sample of people with CFS described themselves as ‘perfectionists’ or ‘Type A personalities’. Empirical work, however, has failed to find an increased rate of perfectionism amongst participants with CFS to date (e.g. Wood and Wessely, 1999).

The construct of Type A personality shares a number of features with perfectionism, for example, setting oneself high standards and an association with having demanding or critical parents (Flett, Hewitt, Blankstein, Dynin, 1994). Although Lewis, Cooper, and Bennett (1994) found that participants with CFS rated themselves as better listeners than participants with Irritable Bowel Syndrome (IBS) and healthy controls, which they suggest may reflect high personal standards in personal relationships, they found no evidence for the global Type A behaviour construct amongst participants with CFS. Blakely, Howard, Sosich, Murdoch, Menkes *et al* (1991) assessed a wide range of psychological characteristics within a sample of patients with CFS and concluded that there was no unique set of psychological characteristics associated with CFS.

In summary, although anecdotal reports have been made of a tendency for perfectionism amongst people with CFS, this has not been supported by empirical investigation to date. A possible basis for the discrepancies between clinicians’ reports based on therapeutic interviews and participants’ responses to formal questionnaires are discussed in Section 2.6.

2.3.3. Lifestyles characterised by hard work and striving for achievement

The study of the pre-morbid lifestyles of people with CFS has generally been based on retrospective interviews with patients. This makes it difficult to draw any firm conclusions as the nature of the illness may alter people's perceptions of their lifestyle prior to becoming ill. Furthermore much of the evidence cited in this area is anecdotal. A number of authors have noted that common themes in therapy with patients with CFS are overextended and overcommitted pre-morbid lifestyles (Duff, 1993), and a desire for accomplishment and success (Ware and Kleinman, 1992). Ware (1993) reported patients' descriptions of themselves prior to becoming ill with CFS with particular reference to how active they were. Well used terms included 'busy', 'involved in life', 'working up to eighty hours a week', 'involved in a million things at once', 'combining jobs, voluntary work, child care, social lives', 'workaholics'. Lewis *et al* (1994) found that patients with CFS did tend to report a greater number of outside interests than patients with IBS and healthy controls, and both CFS and IBS patients reported themselves as being more hard-driving prior to their illness than the healthy controls rated themselves currently. That both patient groups shared this tendency may suggest that the impact of a chronic illness may affect how people tended to rate themselves pre-morbidly.

Abbey (1993) states that, from patients' anecdotes, it was clear that an active lifestyle was an important part of their self-image and a more important maintaining factor for self-esteem than in other patient populations. As mentioned earlier, Johnson and colleague's (1997) study suggests that although this may be the case for a subgroup of people with CFS who become depressed it was not the case for those who were not also depressed. Although there are a great deal of anecdotal references to a pre-morbid lifestyle oriented towards achievement, prospective studies are

necessary to overcome the inherent biases in the studies that have been carried out to date.

2.3.4. Thoughts associated with a physical basis for the illness.

A number of studies have reported a tendency for patients with CFS to hold primarily physical attributions about the cause and maintenance of their illness, both from clinical reports (e.g. Surawy *et al*, 1995) and empirical investigation. Powell, Dolan and Wessely (1990), for example, found that 80% of patients with CFS believed that their illness was physical. In fact, Wessely and Powell (1989) were unable to distinguish between a group of patients with CFS and a group of patients with depression based on symptoms. The groups were distinguishable, however, by their attributions for their illness with 18 out of 21 CFS patients reporting a physical cause compared to 4 out of 26 depressed patients. Moss-Morris and Petrie (1996) obtained similar results. Other findings, however, have not been wholly consistent. Clements, Sharpe, Simkin, Borrill and Hawton (1997) reported that although 97% of their sample rated physical or disease factors as important, only 45% of these stated that physical factors were the sole cause. 56% reported that social or lifestyle factors were important, although explanations tended to refer to the physical vulnerability that arose following stress, rather than a psychogenic cause. Ray, Jefferies and Weir (1995) found that the majority of patients reported that both 'physical' and 'non-physical' factors had caused CFS, the wording in this study was careful to avoid the use of the term 'psychological' which may have increased the frequency that people endorsed a 'non-physical' cause.

The reported tendency for patients to make physical attributions about their illness may be influenced by selection bias. Euba *et al* (1996) compared patients with CFS who attended primary versus tertiary care clinics, the tertiary care attenders had

higher socio-economic status and tended to make a greater number of physical attributions about their illness. McDonald *et al* (1993) found that primary care attenders were less likely to make wholly physical attributions for their illness than hospital attenders. Community surveys have also been less likely to identify excess physical attributions for illness by CFS patients (Mann, McDonald, Cope, Pelosi and David, 1994; Lawrie and Pelosi, 1995).

Perhaps one reason why this area has received so much attention is that the notion that the physical illness is the sole problem is often at variance with the clinicians' experiences (Abbey, 1993), i.e. that there is often a presence of other psychosocial factors or negative life events. Furthermore, patients may continue to have as much conviction in an organic disease process as, for example, patients with Multiple Sclerosis, despite the skepticism that patients encounter and the uncertainty from research (Trigwell, Hatcher, Johnson Stanley and House, 1995).

Despite the inconsistencies in the findings, the presence of physical attributions for CFS has continued to be regarded as an important factor due to a possible relationship with the prognosis of the illness. Clearly, whether it is correct to make a physical attribution for the illness cannot be judged, but it may be possible to judge whether it is *useful* for the patient to hold a physical attribution in terms of their prognosis. The results with regards prognosis have, however, been mixed. Some studies have found an association between holding a physical attribution about the illness and poor prognosis (Sharpe, Hawton, Seagroatt and Pasvol, 1992) and poor outcome from Cognitive Behaviour Therapy (Butler, Chalder, Ron and Wessely, 1991). Community studies have found no relationship (Cope, David, and Mann 1994; Wessely, Chalder, Hirsch, Pawlikowska, Wallace *et al*, 1995). Again, this may relate to some extent to selection biases. Euba *et al* (1996) found that in their primary

care group the presence of physical attributions was not associated with more physical symptoms, less psychological distress or worse outcome. Alternatively, the inconsistencies may relate to the lack of attention that has been paid to the different functions of explanations, such as labelling or description, moral evaluation, causal attribution and self-presentation (Brewin and Antaki, 1987). The impact of holding a particular attribution is likely to depend on the function of the attribution held.

In accordance with a 'labelling' view of illness attributions, Deale, Chalder and Wessely (1998) have suggested that it is not physical attributions per se that predict a poor outcome from CBT, but avoidance of exercise. It is understandable that this behaviour would arise if fatigue were labelled as a sign of damaged joints or muscles combined with a fear of making this worse. This is likely to relate to the tendency reported by Petrie, Moss-Morris and Weinman (1995) for people with CFS to hold 'catastrophising' beliefs about the illness, for example regarding the effects of pushing oneself, and for the presence of these beliefs to be associated with greater fatigue and disability. Similarly, Moss-Morris, Petrie and Weinman (1996) found that in CFS, cognitive distortions relating to somatic experiences were associated with pessimistic illness beliefs, a belief in serious consequences of the illness and a chronic course of the illness. Furthermore, the interaction between a belief in serious consequences of the illness and somatic cognitive distortions was predictive of the extent of disability six months later.

Why is it that some people appear to hold on to strong physical attributions for the cause and maintenance of the illness so firmly in the face of the skepticism that they endure from others and the lack of research evidence to support the view? The ambiguous status of the illness may be one factor which produces social and personal pressure to offer a plausible explanation for the incapacity experienced,

both to oneself and to others (Frank, 1946). In the search for a plausible explanation, viruses may appear to be a likely candidate as they are common and likely to coincide with the onset of illness, indeed this may be a correct attribution for the initial onset of the illness.

A possible reason why this attribution may be held on to so vehemently in some cases draws together hypotheses relating to the preceding sections and to a morally evaluative function of illness attribution. If high standards and achievement determine a person's sense of self-worth, self-esteem will be threatened in situations when they are unable to meet these standards. Such a situation would occur when a person becomes incapacitated by fatigue. It has been suggested that holding a physical disease attribution for that fatigue may protect the psychologically vulnerable from a threat to their self-esteem (Wessely, Hotopf and Sharpe, 1998). Surawy *et al* (1995) comment on the tendency for their patients to view depression as a weakness for which the person is at fault or blameworthy. Further empirical investigation must take in to account the influence of selection biases and the strong possibility that there may be subgroups of patients with CFS for whom these factors play a stronger part than for others.

It is also unclear whether attributions about the cause and maintenance of CFS relate to a more general attributional style. This might be expected if illness attributions reflect moral evaluations or self-presentation strategies. Three studies have attempted to investigate the attributional style of people with CFS using a more global measure, the Attributional Style Questionnaire (ASQ) (Peterson, Semmel, von Baeyer, Abramson, Metalsky *et al*, 1982). Chalder, Power and Wessely (1996) used an adaptation of the ASQ with participants in a community sample. The study did not, however, involve a non-CFS comparison group and the adapted version of the

questionnaire primarily addressed attributions about illness. Howlett and Lindegger (1996) carried out a pilot study to compare a group with CFS to a depressed group and a chronically physically ill group on the ASQ and measures of depression and illness behaviour. The CFS group were more similar to the depressed group on measures of depression and illness behaviour, but more similar to the chronically physically ill group on their responses to the ASQ. This study was limited by small sample sizes, the recruitment of three groups from different sources (i.e. primary and tertiary care) and length of illness was not taken in to account. Nevertheless this is an interesting result as a consistent pattern has been found between depression and attributional style, with depressed patients tending to make internal, global and stable attributions for negative events (Abramson, Seligman and Teasdale, 1978). Despite giving a 'depressed' pattern of results on measures of depression and illness behaviour, the CFS group did not report a depressive attributional style. Chubb, Jones, Hillier, Moyle, Sadler *et al* (1999), however, found contrasting results with larger samples of participants. For attributions regarding negative events, participants with CFS who were concurrently depressed tended to respond in a similar manner to depressed participants, whereas the responses of participants who were not depressed resembled those of the healthy comparison group.

The present study addresses the hypothesis that the maintenance of a non-depressive attributional style and reports of unimpaired self-esteem amongst people with CFS may reflect defence mechanisms aimed at preserving self-esteem. First, outcome studies from therapy trials based on the cognitive-behavioural model of CFS will be reviewed in order to ascertain whether these studies shed light on the applicability of the cognitive aspects of the model to CFS.

2.4. COGNITIVE BEHAVIOUR THERAPY FOR CFS

Cognitive Behaviour Therapy (CBT) for CFS aims to change the thoughts and beliefs that are thought to maintain disability and symptoms associated with CFS. Table 2.1 summarises the results of a number of trials that have been carried out to assess the outcome of CBT for CFS. On the whole, the results are favourable and participants who undergo CBT have been found to achieve better outcomes in terms of physical functioning, fatigue and mood compared to those who do not participate in any intervention, those who have standard medical care and those that participate in relaxation training.

Two studies stand out due to the absence of successful outcomes following CBT. Freidberg and Krupp (1994) found that CBT only had an impact on measures of depression, and did not affect participants' levels of disability and fatigue. In this study, however, the CBT delivered differed from other studies in that it was based on a model of accepting disability, so did not involve behavioural activation or cognitive re-attribution strategies. Lloyd *et al* (1993) carried out a double-blind randomised controlled trial, essentially comparing CBT with no psychological treatment and an immunologic therapy with a placebo. In this study no substantial differences were found between the groups. The striking difference between this study and others is the relatively short length of CBT treatment. This treatment involved only six sessions, compared to between 12 and 16 sessions in other trials, allowing little time for work on relapse-prevention. Furthermore, the nature of the behavioural interventions carried out in the study is unclear and no attempts appeared to be made to alter participants' thoughts and beliefs. Finally, all participants were concurrently being administered a regular injection of immunologic treatment or placebo. This would be likely to compromise the rationale for carrying out CBT, which is based on

a model that might allow for the presence of immunologic factors in the onset of illness, but not in the continuation of symptoms.

Due to the methodological difficulties involved in these two studies, the results do not preclude us from concluding that CBT appears to be an effective treatment for many people with CFS. The fact that the two studies which did not find successful results did not appear to address cognitive aspects of CFS may further support the cognitive hypotheses of the CBT model of CFS. However, the behavioural tasks carried out in these two studies are also unclear. Can we, at this stage then, conclude that the cognitive components of CBT for CFS add anything over and above the behavioural components of the treatment?

One study that might suggest that it is the behavioural components of CBT that are of primary importance is that of Fulcher and White (1997). In their study, 66 participants meeting Oxford Diagnostic Criteria for CFS were randomly allocated to a graded exercise programme or a flexibility programme. Both treatment programmes consisted of 12 weeks of weekly supervised treatment plus 5 sessions of home-exercise per week, controlling for therapists' time. At the end of treatment 16 of the 29 completers in the graded exercise group rated themselves as 'very much or much improved' on a measure of global impression of well being, compared to 8 of the 30 completers in the flexibility group. One person in each group rated themselves as worse after treatment. Many of the participants in the flexibility group then went on to complete the graded exercise programme, and 12 out of 23 rated themselves as better after treatment. At a one year follow-up 35/47 participants in the exercise group who were assessed were rated as better, and physiological improvements were generally maintained or exceeded, compared to 2 out of 5 participants who only took part in the flexibility programme. Significantly better outcomes were also found for

the graded exercise group in terms of physical fatigue, global ratings of health and better physical functioning on the SF-36. There was little difference in pre- and post-treatment anxiety and depression scores for either group. At one year follow-up 74% of completers of a graded exercise programme rated themselves as better, which is comparable to the results of CBT trials which have generally found that about 70% of participants demonstrate a significant improvement. The graded exercise programme described by Fulcher and White did not make any attempt to address thoughts or beliefs, but used basic principles of exercise prescription adapted for participants' current capacity. As objective measures of physical fitness were not linked to outcome, Wessely, Hotopf and Sharpe (1999) suggest that the benefits were linked to confidence, predictability, and overcoming avoidance, supporting the view that disability is more related to behavioural avoidance and confidence than physical fitness alone. Wessely and colleagues suggest that exercise treatment is likely to address the fears of patients, and may, hence, work in a similar manner to CBT that involves the use of behavioural experiments to test out patients' fears.

In summary, the evidence to date suggests that both CBT and graded exercise programmes may be effective in the treatment of CFS. How these interventions work, however, is not yet clear although it does not appear to be a matter of simply increasing participants' physical fitness but may relate to challenging the fears which they hold relating to their illness. This suggestion supports Sharpe's (1997) model with regards the tendency for people with CFS to hold beliefs such as 'I must be physically ill', 'I'm making myself worse' and 'There's nothing I can do'. As these thoughts are about the illness, rather than about the individuals themselves, it does not, however, go anyway to support the earlier aspects of the model relating to predisposing factors. The following sections discuss the difficulties inherent in

attempting to investigate predisposing factors related to self-esteem, particularly if defence mechanisms are in place to enable individuals to cope better with their underlying difficulties.

Figure 2.1

'A complex specific model of chronic fatigue syndrome', Sharpe (1997)

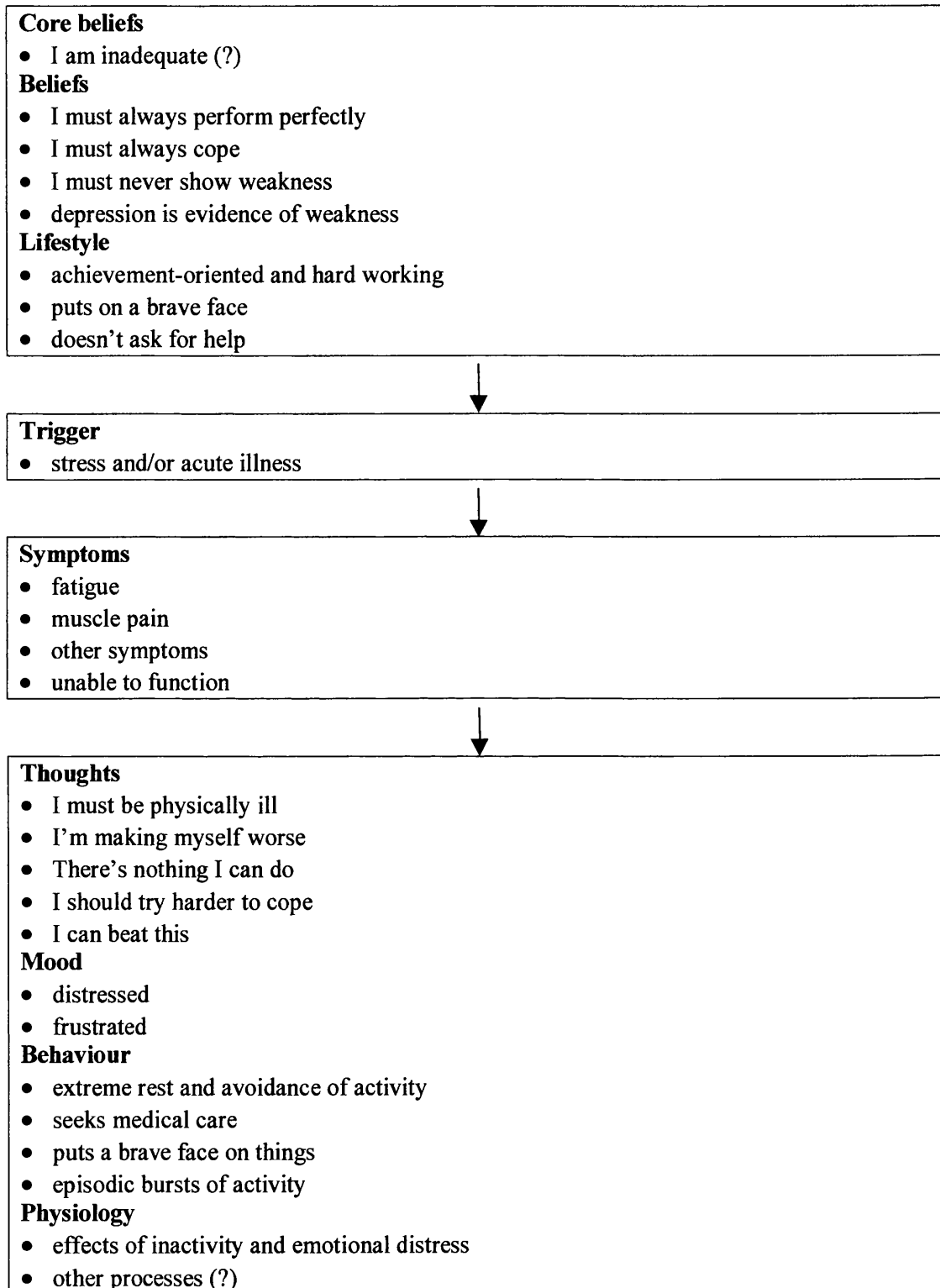


Table 2.1
Outcome studies of CBT for CFS

	Design	n	Outcome	Treatment	Notes
Butler <i>et al</i> (1991)	No control	50	70% completers 'better'/ 'much better'	2-20 hours CBT mean time= 7.5 hours	High refusal rate (36%)
Bonner <i>et al</i> (1994)	4 year follow-up of Butler <i>et al</i> (1991)	29	20/23 completers remained well; 2/16 refusers/ incompleters spontaneously recovered	As above	
Lloyd, <i>et al</i> (1993)	Double blind RCT CBT vs. no treatment; Immunologic therapy vs. placebo	90	All conditions: no specific improvements; CBT + immunologic therapy: small improvements on self-report, not sustained at 7 month follow-up	6 x 30-60 minutes CBT bi-weekly	Short treatment; Behavioural interventions unclear; No attempt to alter beliefs; CBT rationale compromised by concurrent immunologic therapy?
Freidberg and Krupp (1994)	Non-randomised trial CBT vs. refusers	64	Improved on depression measure only; No change re. disability or fatigue	6 sessions CBT	CBT based on accepting disability; No behavioural activation or cognitive re-attribution
Sharpe <i>et al</i> (1996)	CBT vs. standard medical care	60	CBT: 60% subjective improvement, 73% improved on Karnovsky scale at 12 month follow-up Standard medical care: 23% subjective improvement, 27% improved on Karnovsky scale	16 x 1 hour weekly sessions	CBT comprised of individual formulation, explanation and education; behavioural experiments, collaborative re-evaluation of thoughts and beliefs; problem solving
Chalder, Butler and Wessely (1996)	Uncontrolled	6 severe cases	5/6 improved on work, social functioning, mood and symptoms	Inpatient treatment	CBT similar to Sharpe <i>et al</i> (1996) but more behavioural emphasis
Deale <i>et al</i> (1997)	RCT: CBT vs. relaxation	60	CBT: 70% substantial improvements on physical functioning Relaxation: 19%	13 weekly/ fortnightly sessions mean time= 15 hours	CBT similar to Chalder <i>et al</i> (1996)

2.5. THE ROLE OF DEFENCE MECHANISMS IN CFS

Although it has not been directly addressed by the literature to date, similarities are apparent between Sharpe's (1997) model of CFS and the literature on defensive coping styles. Weinberger, Schwartz and Davidson (1979) proposed a four-fold classification of individuals based on their coping styles: repressors (low anxiety - high defensiveness), low anxious (low anxiety – low defensiveness), high anxious (high anxiety – low defensiveness) and defensive high anxious (high anxiety – high defensiveness). Repressive coping has received a great deal of attention in the research literature, and a number of characteristics of this group have been identified. Defensive-high anxious individuals, on the other hand, have received minimal research attention to date, quite possibly because of the tendency for fewer individuals to fit the criteria for inclusion in this group.

Weinberger (1990) argued that repressors are individuals for whom maintenance of a low level of negative affect is central to their self- concept. Evidence which supports this view includes repressors' high levels of physiological reactivity in potentially stressful situations while reporting low levels of distress (Asendorpf and Scherer, 1983), limited availability of negative emotional memories (Myers, Brewin and Power, 1992), use of an avoidant processing style for negative affective material (Myers and McKenna, 1996) and the employment of strategies in maintaining favourable images of their self-concept when this is threatened (Baumeister and Cairns, 1992). Weinberger and colleagues (1979) give examples of repressors' self-descriptions, which they describe as reflecting a preoccupation with mastering negative emotion, rigorously controlling behaviour and giving value to a rational, non-emotional approach to life. Weinberger and colleagues suggest that the repressive coping style leads individuals to avoid seeking help for personal problems

but may make them prone to physical ill health (Blackburn, 1965) and mental ill health (Shedler, Mayman and Manis, 1993). The repressive coping literature has clear overlaps with the cognitive behavioural model of CFS. In both underlying low levels of self-esteem are hypothesised. The method of coping with this, in both cases, is hypothesised to be the development of beliefs which value low levels of negative affect, for example, Sharpe (1997) describes the belief 'depression is evidence of weakness' amongst people with CFS. Such beliefs, as hypothesized in both cases, lead to a lifestyle characterised by not asking for help and putting on a brave face.

One crucial difference between repressive copers and people with CFS is that high levels of anxiety have been found amongst people with CFS (e.g. Wessely *et al*, 1996). Although earlier studies of psychiatric morbidity made little mention of anxiety disorders, more recent studies have suggested they have an increased role (see Section 1.11.2). The similarities between the repressive coping literature and the literature on CFS raise the question that these similarities may reflect high levels of defensiveness amongst people with CFS.

A lot less research has been carried out to date specifically looking at defensive high anxiety. This may reflect the low prevalence of this coping style, even amongst illness populations who tend to report higher levels of anxiety than healthy populations (see Table 2.2). Studies that included defensive high anxious participants as a control group suggest that they have some characteristics in common with repressive copers and others with genuine high anxious participants. In particular, defensive high anxious participants tend to respond in the same manner as genuine high anxious participants on self-report, physiological and behavioural measures of anxiety (e.g. Eysenck and Derakshan, 1997).

A number of studies have suggested that a defensive high anxious style may impinge as much as, or even more than, the repressive coping style on physical health. A hypothesised mechanism is via the hypothalamic-pituitary-adrenal axis, which may also have a role in CFS (see Section 1.10). Jamner, Schwartz and Leigh (1988) found that defensive high anxious attenders of a Behavioural Medicine Clinic had the lowest monocyte counts (reflecting lower immunocompetence), followed by repressive copers. Jamner, Schwartz and Leigh (1988) suggest that defensive-high anxious participants may represent 'failed repressors', whose coping mechanisms have become ineffective. They propose that the defensive style leads to an increase in endogenous opiate levels, which is associated with decreased immunocompetence. The co-occurrence of anxiety may lead to a greater reduction in monocytes through its effects on corticosteroids and/ or catecholamines. They suggest that this may also explain the reported interaction of greater social desirability with heightened distress as a predictor of worse medical outcome in malignant melanoma (Temoshok, 1987). At this stage, however, this remains speculative. As Bonanno *et al* (1991) describe 'defensive high anxiety somehow changes the effects of anxiety in ways that are currently little understood'.

For people who maintain a defensive position (repressive copers or defensive high anxious), traditional measures of psychological functioning, such as self-esteem questionnaires and attributional style questionnaires may be particularly prone to response bias. Rigid defence mechanisms are likely to distort self-reports that rely on conscious cognitive processes. The present study attempts to avoid this difficulty by using indirect tests of self-esteem and attributional style. Indirect tests of psychological functioning have not been applied to repressive copers. Myers (in press), however, described a comparison of participants' responses to interviews with

their responses to structured questionnaires and found conflicting reports about paternal relationships. From a semi-structured interview, repressors held a negative view of their fathers, reporting more paternal antipathy, indifference and being less emotionally or physically close to their fathers compared to non-repressors (Myers and Brewin, 1994). When a questionnaire measure of global childhood experience was used repressors reported more positive childhood experiences (Myers, Brewin and Winter, 1999). A similar pattern in CFS might account for the common discrepancies described in the above review of psychological processes in people with CFS between clinicians' anecdotal reports and the results of empirical, self-report based, investigations.

The present study investigates the prevalence of a defensive-high anxious coping style amongst participants with CFS. It is suggested that this style may be a mechanism that has developed in an attempt to protect vulnerable self-esteem. In the following section, literature from the study of psychosis will be drawn upon to identify methodology that may access participant's underlying or core beliefs about themselves, without the influence of conscious cognitive processes that lead to response biases.

Table 2.2

The proportion of defensive high anxious participants in illness populations

(studies using median splits to assign to coping style)

Authors	Population	Criteria for defensive high anxious	% of population (n)
Jensen (1987)	Overall	MA \geq 8 SD \geq 17	R 49% (42) D 17% (15) H 22% (19) L 12% (10)
	Advanced breast cancer		R 64%(16) D 8% (2) H 16% (4) L 12% (3)
	Recurrence free breast cancer		R 44% (12) D 19% (5) H 26% (7) L 11% (3)
	Minor surgery		R 41% (14) D 23.5% (8) H 23.5% (8) L 12% (4)
Jamner <i>et al</i> (1988)	Behavioural Medicine Clinic patients (stress-related complaints)	MAS \geq 9 MC \geq 17	R 27% (79) D 24% (69) H 42% (124) LA 7% (20)

R= Repressive Copers

D= Defensive High Anxious

H= High Anxious

L= Low Anxious

2.6. INVESTIGATING CORE BELIEFS

Two approaches which have been applied within the study of psychosis in order to access participant's underlying or core beliefs about themselves will be described: (i) using measures of attributional style and (ii) using measures of information processing biases. The rationale for using these measures was based on Higgins' (1987) 'self-discrepancy theory'. According to this model individuals hold representations of three different aspects of their selves: 'the actual self', how one is perceived to be by oneself or by others; 'the ideal self', how one would like to be; and 'the self as it ought to be', how one believes one should be. Higgins investigated the association between self discrepancies and psychological symptoms and observed that the discrepancy between actual and ideal selves was associated with the degree of depression reported. Bentall, Kinderman and Kaney (1994) proposed that attributions may aim to reduce discrepancies between one's own perceptions of the self. Furthermore, Bentall and Kaney (1996) suggested that excessively rigid and perfectionist criteria for evaluating personal performance and self-worth are likely to make a person vulnerable to discrepancies between their actual self and ideal self representation, leading to low self-esteem. Based on the anecdotal and clinical reports of CFS regarding excessively rigid and perfectionist standards for self-performance, as discussed in Section 2.3, this particular discrepancy might be anticipated amongst people with CFS.

2.6.1 Measures of attributional style

Bentall and colleagues (Bentall and Kaney, 1996; Kinderman and Bentall, 1997) have applied this model to the study of people who experience persecutory delusions. They suggest that this group attempt to minimise underlying discrepancies between their ideal and actual selves (as perceived by themselves) by making

external personal attributions for negative events, i.e. attributing the cause of negative events to other people or circumstances rather than to themselves. This pattern of attribution in which positive events tend to be attributed to oneself, is described as 'self-serving' and tends to be seen, although to a lesser extent, amongst healthy participants (Zuckerman, 1979; Hewstone, 1989). This is the opposite of the attributional style that has been described amongst people who are depressed.

Depressed participants tend to make internal personal attributions for negative events, i.e. attributing the cause of negative events to oneself (Sweeney, Anderson and Bailey, 1986) as predicted by the reformulated learned helplessness model of depression (Abramson, Seligman and Teasdale, 1978). This model proposed that individuals who attribute negative events to internal, global and stable causes are predisposed to depressive reactions following negative life events.

The exact nature of the relationship between attributional style and psychopathology remains unclear. Brewin (1985) reviewed the evidence relating to the temporal and causal relationships between attributional style and depressed mood and concluded that while attributional style may be predictive of maintenance of depression, there is no evidence that a particular attributional style precedes depression. In the present study, no specific claims are being made regarding the nature of the temporal relationship between attributional style and psychopathology. Bentall, Kinderman and Kaney (1994) suggest that this attributional style acts to minimise the individual's awareness of the discrepancies between actual and ideal selves in order to maintain a positive explicit self-concept. According to this model, a discrepancy between overt and covert measures of self-esteem would be predicted, in that overt measures will not reflect actual-ideal self-discrepancies, while covert measures will detect the discrepancy.

Lyon, Kaney and Bentall (1994) adapted the Attributional Style

Questionnaire (ASQ) to be used alongside the Pragmatic Inference Test (PIT) in order to compare responses on direct and indirect measures of attribution. The ASQ (Peterson, Semmel, von Baeyer, Abramson, Metalsky *et al*, 1982) requires participants to provide possible explanations for hypothetical positive and negative events and then to rate these explanations on three scales: internality (caused by self vs. caused by others/ circumstances), stability (unchangeable vs. changeable cause) and globality (affects all areas of life vs. affects only this area). The PIT (Winters and Neale, 1985) was developed from the ASQ and requires participants to make attributional inferences when responding to an apparent memory test. Winters and Neale used the PIT with manic patients and found that, although remitted manic patients reported high self-esteem on conventional measures, they displayed an attributional style similar to depressed patients when tested on the PIT. From their review of the literature regarding cognitive approaches to delusions, Garety and Freeman (1999) conclude that this method appears to be valid. Lyon, Kaney and Bentall (1994), replicating the findings of Kaney and Bentall (1989) and Candido and Romney (1990), found that deluded patients made excessively external self-ratings of their explanations for negative events and excessively internal self-ratings of their explanations for positive events on their adapted version of the ASQ (ASQ-parallel form). On the PIT, however, like manic patients, their attributional inferences substantially resembled those of depressed participants, i.e. both groups made extremely external attributions for positive events and internal attributions for negative events whereas the healthy controls did the opposite. Comparing the responses to the direct and indirect tests of attributional style, the deluded participants 'demonstrated a dramatic transition in style between the two measures,

changing from an extremely self-serving bias to an extremely self-disparaging bias according to the method of testing' (page 642). Lyon, Kaney and Bentall (1994) tentatively suggest that this may be evidence that implicit and explicit judgements about the self are sustained by distinct cognitive mechanisms, in a similar way to other types of information (Berry and Broadbent, 1988). Power and Brewin (1991) have suggested that these distinct cognitive mechanisms may be similar to the psychoanalytic distinction between the conscious and the unconscious, and beliefs about the self may be represented at both levels.

The results from the use of the ASQ-pf and the PIT can offer no more than a tentative suggestion, as the PIT data does not allow us to determine whether the process of recognising and reporting particular attributional styles occurs at a conscious or unconscious level. It is possible that the PIT data merely confuses participants' conscious attempts at presenting themselves in a positive manner. Measures which investigate information-processing biases appear to be less susceptible to conscious self-presentation. These methods have been used to address the predicted discrepancy between overt and covert measures of self-esteem.

2.6.2 Measures of information processing bias

Within the persecutory delusions literature, two methods of examining information processing biases have been applied: (i) Self-Referent Inferential Recall Tests (SRIRT), and (ii) the Emotional Stroop Test (EST). SRIRTs are based on the 'Self-Referent Effect' which refers to the general tendency for people to preferentially recall information which relates to their view of themselves (Rogers, Kuiper and Kirker, 1977). A number of studies have found that when depressed participants are required to recall trait words, which were presented earlier as part of a personality test, they tend to recall more negative words than healthy controls

(Hanzman, Marks, Mayall and de Mayo 1985; Hanzman, Duke and Micklovitch, 1986; Dent and Teasdale, 1988). Bentall and Kaney (1996) proposed that the SRIRT is likely to provide evidence of underlying negative representations of the self even in individuals who are highly defensive. From their review, Garety and Freeman (1999) concur that the SRIRT is likely to be a valid method of accessing covert self-evaluations. In support of their hypothesis, Bentall and Kaney (1996) found that although deluded participants and healthy controls endorsed more positive words as applying to themselves than depressed participants, non-depressed deluded participants recalled the most negative endorsed words, compared to depressed participants and healthy controls (who recalled the least).

Kinderman's (1994) findings from an Emotional Stroop Test also support the presence of an underlying fragile self-concept amongst people with persecutory delusions. The Stroop Test (Stroop, 1935) requires participants to name the colour of ink that a word is written in. Attentional bias towards particular words or classes of words can be inferred from the degree of interference with the participant's performance, as measured by the speed of colour naming. Williams, Mathews and MacLeod (1996) review the numerous applications of an emotional analogue of the Stroop test in relation to psychopathology, in which patients are required to name the colour of the words printed and the words vary in their relevance to the theme of each psychopathology. Patients are often slower to name the colour of a word that relates to their clinical condition. Suicide attempters, for example, tend to be slower at naming the colour of words relating to overdosing (Williams and Broadbent, 1986); depressed patients tend to be slower on negatively toned words (Gotlib and McCann, 1984); and anxious patients show selective interference from threat related words (Mathews and MacLeod, 1985). Furthermore these interference effects appear

to relate to differences in accessing constructs relating to the psychopathology rather than reflecting a consequence of the associated affect. Gotlib and McCann (1984) used a mood induction procedure with non-depressed participants and found that the induction of depressed mood did not affect response latencies to negatively toned material, suggesting that the differences found with depressed participants are not due solely to transient mood differences. Studies have shown the Stroop effect even when participants are not aware of the material presented and it can therefore be regarded as a valid measure of individuals' covert concerns which is not dependent on conscious strategies (Williams, Mathews and MacLeod, 1996).

Kinderman (1994) studied attention to positive and negative trait words using an Emotional Stroop task with participants suffering from persecutory delusions and matched participants with depressed mood and healthy controls. Despite the participants with persecutory delusions demonstrating a significantly higher rate of endorsement for positive adjectives than negative adjectives, they showed a marked degree of interference when colour-naming both positive and negative words, but particularly negative words, in a manner similar to those with depressed mood. Kinderman concluded that there is evidence of a covert negative self-concept from the results of the Emotional Stroop test in the persecutory delusions group. Garety and Freeman (1999) supported this conclusion however they question whether the level of depression amongst the group might be a contributory factor.

Although there remains some debate within the persecutory delusions field regarding the conclusions that can be drawn from the data from this particular group of patients, there is agreement that the application of the ASQ-pf and the PIT is a valid approach to overt and covert investigation of attributions and the SRIRT and Emotional Stroop Tests are valid approaches to the study of overt and covertly

expressed self-esteem. These methods will be applied in the present study in order to investigate the hypothesis that people with CFS have underlying low self-esteem, which is not reflected by overt measures because of the existence of defensive mechanisms to maintain a positive explicit (or conscious) self-concept. The potential defence mechanisms investigated in the present study are a defensive high anxious coping style and physical illness attributions.

2.7. SUMMARY

Chronic Fatigue Syndrome describes a disorder characterised by a principal complaint of fatigue accompanied by substantial functional impairment. The aetiology of CFS has been investigated from various perspectives, in particular with regards the presence of infectious diseases, immune system dysfunction, muscular dysfunction and neuropsychology and neurophysiology. Few consistent findings have, however, been reported. The high prevalence of psychiatric disorder amongst people with CFS, particularly of depression, has led investigators to examine the relationship between these factors. A cognitive behavioral model of CFS has been proposed (Sharpe, 1997). Evidence to support the various claims of the model, however, is sparse, in particular relating to the hypothesis that people with CFS have low underlying levels of self-esteem which are protected by rigid defence mechanisms, including the tendency to make physical attributions for illness. The difficulties inherent in using conscious measures of psychological functioning to address this hypothesis have been described. The present study investigates the hypothesis that CFS occurs in people who have low levels of self-esteem and an underlying depressogenic attributional style when they are confronted with a life stressor that is incompatible with their defence mechanisms. A potential defence mechanism that is addressed is the defensive high anxious coping style.

As it is proposed that these patterns represent predisposing factors to CFS, rather than reflecting a response to chronic illness, a comparison group of people with a different chronic illness is included in the present study. Patients with insulin-treated diabetes mellitus form the comparison group as, as well as representing a chronic illness group that affects young people, there are a number of similarities between CFS and diabetes. In particular, diabetes is a 'hidden' condition that is not readily

apparent to other people. Furthermore, diabetes is a demanding condition that requires significant regulation of behaviour and imposes restrictions on lifestyle. For the present investigation it is important to include a comparison group in which levels of psychiatric disorder are equivalent to other chronic illness groups, and do not constitute a further risk for psychiatric morbidity. Although higher than in the general population, the lifetime prevalence of psychiatric disorders amongst people with diabetes is approximately equivalent to other chronic illness groups. In accordance with other chronic illness groups, patients with type 1 and type 2 diabetes have been found to have prevalence rates of psychiatric disorder between 14.4 and 32.5% (de Groot, Jacobsen, Samson and Welch, 1999).

2.8 HYPOTHESES

The major hypothesis of the present study is that people with CFS have low levels of self-esteem which are defended by physical attributions for illness and a defensive high anxious coping style.

In order to investigate this, the following specific hypotheses will be addressed:

1. Physical illness attributions are associated with high levels of self-esteem amongst people with CFS, while psychological attributions are associated with low levels of self-esteem.
2. Participants with CFS will not report low levels of self-esteem or a depressogenic attributional style when tested on overt measures when compared to a healthy control group.
3. Participants with CFS will show low levels of self-esteem and a depressogenic attributional style when tested on covert measures when compared to a healthy control group. A comparison chronic illness group will not exhibit a discrepancy between responses to overt and covert measures.
4. Participants with CFS will display a greater tendency to hold a defensive high anxious coping style, characterised by high levels of reported trait anxiety and high levels of defensiveness on self-report measures when compared to a healthy control group and comparison chronic illness group.

CHAPTER THREE

METHOD

3.1. OVERVIEW

Sixty-eight participants made up three groups: participants with Chronic Fatigue Syndrome, a comparison group of participants with a chronic medical condition (Insulin treated diabetes) and a healthy comparison group. All participants completed one standardised assessment comprised of structured interview, tests of memory and concentration and questionnaires.

3.2 PARTICIPANTS

3.2.1. CFS participants

Twenty-four participants with CFS were recruited via three tertiary referral centres that specialise in the assessment and treatment of CFS.

The age range of participants was restricted to between 18 and 30 years as many studies compare CFS participants to older comparison groups, particularly when the comparison group also experiences chronic illness. By restricting the age range it is also possible that the CFS group will be more homogeneous. All participants spoke English as their first language. Participants had not undertaken a course of cognitive-behaviour therapy, although many were on the waiting list for this. For participants who were prescribed mood-altering medication, this had not been altered within the two months preceding assessment. In accordance with the Oxford Diagnostic Criteria, all participants had experienced fatigue and accompanying symptoms for at least six months. The inclusion criteria are summarised in Table 3.1.

Letters were sent to 58 patients who were known to be between the ages of 18 and 30 years inviting them to participate in the study (see Appendix Ten). Twenty-nine patients responded to the letter (50%). Five participants did not currently fulfill Oxford Diagnostic Criteria for Chronic Fatigue Syndrome (Sharpe *et al*, 1991; see Appendix One) so were not included in the study.

3.2.2. Chronic illness comparison group

Twenty participants with insulin treated diabetes were recruited via three tertiary referral centres which specialise in the treatment of diabetes. Participants were all between the ages of 18 and 30 years and spoke English as their first language. Participants were not involved in any form of psychological therapy, including cognitive behaviour therapy. For participants who were prescribed mood-altering medication, this had not been altered within the two months preceding assessment. All patients had been diagnosed with diabetes for at least six months. The inclusion criteria are summarised in Table 3.2.

Letters were sent to 125 patients who were known to be between the ages of 18 and 30 years inviting them to participate in the study (see Appendix Eleven). Twenty eligible patients responded to the letter (16%).

3.2.3. Healthy comparison group

Twenty-four participants formed a healthy comparison group, recruited primarily via posters and e-mail advertisements within the University of London, attracting students and members of staff. Other participants were recruited via word of mouth. Participants were all between the ages of 18 and 30 years and spoke English as their first language. The distribution of gender within the group was matched with the CFS group.

Table 3.1 Inclusion Criteria for Participants with CFS

- | | |
|-------|--|
| (i) | The participant fulfils Oxford Diagnostic Criteria for CFS (see Appendix One). |
| (ii) | The participant is between 18 and 30 years of age. |
| (iii) | The participant speaks English as his/her first language. |
| (iv) | The participant is not (currently or in the past) involved in psychological interventions for CFS or any psychiatric disorder, e.g. CBT. |
| (v) | Medication has been stable for at least two months. |

Table 3.2 Inclusion Criteria for Participants with Diabetes

- | | |
|-------|--|
| (i) | The participant has Insulin-treated diabetes, which has been diagnosed for at least six months. |
| (ii) | The participant is between 18 and 30 years of age. |
| (iii) | The participant speaks English as his/her first language. |
| (iv) | The participant is not (currently or in the past) involved in psychological interventions for CFS or any psychiatric disorder, e.g. CBT. |
| (v) | Medication has been stable for at least two months. |

Table 3.3 Inclusion Criteria for Healthy Participants

- | | |
|-------|--|
| (i) | The participant is between 18 and 30 years of age. |
| (ii) | The participant speaks English as his/her first language. |
| (iii) | The participant is not (currently or in the past) involved in psychological interventions for CFS or any psychiatric disorder, e.g. CBT. |
| (iv) | Medication has been stable for at least two months. |

Participants were not involved in any form of psychological therapy, including cognitive behaviour therapy. For participants who were prescribed mood-altering medication, this had not been altered within the two months preceding assessment. The inclusion criteria are summarised in Table 3.3.

3.3. ETHICAL CONSIDERATIONS

This proposal was reviewed by the Joint UCL/UCLH Committees on the Ethics of Human Research, the The Bethlem and Maudsley NHS Trust and Institute of Psychiatry Ethical Committee (Research), St. Mary's Local Research Ethics Committee and Barking and Havering Local Research Ethics Committee. The Whittington Hospital Local Research and Ethics Committee, in association with the Joint UCL/UCLH Committees, also reviewed the proposal. Copies of the letters of approval are given in Appendix Twelve.

3.4. PROCEDURE

Each participant took part in one assessment. The assessment lasted between one and one and a half-hours. All participants were offered the opportunity for breaks during the procedure. Information was collected regarding the participants' age, gender, ethnicity, occupation, education level reached and duration of illness. The measures were then administered in a standard order, as follows.

3.5. MEASURES

3.5.1. Schedule for Affective Disorders (SADS: Spitzer, Endicott and Robins, 1978)

Each participant was assessed using a shortened version of the Schedule for Affective Disorders (SADS) (Spitzer, Endicott and Robins, 1978), in order to ascertain whether participants fulfilled Research Diagnostic Criteria for Major Depressive Disorder. Endicott (1985) reports that there have been over 60 published reports using the SADS.

3.5.2. Emotional Stroop Task (EST: Kinderman, 1994)

The EST was used as a covert measure of self-esteem. The Stroop test (Stroop, 1935) requires participants to name the colour of the ink that a word is written in. Attentional bias towards particular words or classes of words can be inferred from the degree of interference with the participant's performance, as measured by the speed of colour naming. Williams, Mathews and MacLeod (1996) review the numerous applications of an emotional analogue of the Stroop test in relation to psychopathology, as described in Section 2.6.2.

The method of administration of the EST was adapted from Kinderman (1994). Four laminated cards were constructed from white A4 size card. On each card were written ten rows of five words, in five colours of ink (black, red, blue, green and brown). The first card consisted of strings of Os, each four, four, seven, eight and nine characters long. The order of presentation of string length was quasi-random, every line included each character length. The second card consisted of five personally descriptive adjectives of positive content (CALM, WISE, CAPABLE, POSITIVE, REALISTIC). The third card consisted of five personally descriptive adjectives of negative content (LAZY,

WEAK, FOOLISH, CHILDISH, OBNOXIOUS). Kinderman (1994) determined the positive or negative nature of these words by reference to Anderson's (1968) list of value judgements of 555 personal descriptors. This study revealed a clear bipolar distribution of value judgements of personal descriptors, with few words being rated as neutral. The fourth card consisting of neutral words, therefore, employed words that would not normally be used as personal attributes. Kinderman (1994) employed the words PALE, RIPE, RESIDENT, DOMESTIC and HYDRAULIC. The words were matched, across each of the three conditions, for character length and frequency of occurrence in the English language according to Thorndike and Lorge (1944). As this study is concerned with physically ill populations, it was decided that the word PALE might have negative connotations for these groups. This word was therefore replaced with the word LEFT which matched PALE in terms of its frequency in the English language according to Francis and Kucera (1982). In each case the words were presented in a quasi-random order so that all five words from each set appeared on each of the ten lines and each of the five colours were used on each line.

Participants were first asked to identify the colours presented along the top line of card 1 to ensure that they were able to name each colour. The following instructions were then given 'Please name the colour of the ink that each word is written in. Work your way down each column naming the colour. Name the colours as quickly as you can, but try to be accurate also. If you make a mistake just correct yourself and carry on as quickly as possible'. Card 1 was presented first, but the order of presentation of cards 2,3 and 4 was counterbalanced. The time taken to colour-name each card was recorded with a stopwatch and audiotaped for verification purposes.

3.5.3. Self-Referent Inferential Recall Test (SRIRT: Bentall and Kaney, 1996)

The SRIRT was used as a second covert measure of self-esteem. SRIRTs are based on the 'Self-Referent Effect' which refers to the general tendency for people to preferentially recall information which relates to their view of themselves (Rogers et al, 1977). A number of studies have revealed that when depressed participants are required to recall trait words, which were presented earlier as part of a personality test, they tend to recall more negative words than healthy controls (Hanzman, Marks, Mayall and de Mayo 1985; Hanzman, Duke and Micklovitch, 1986; Dent and Teasdale, 1988). Bentall and Kaney (1996) proposed that the SRIRT is likely to provide evidence of underlying negative representations of the self even in individuals who are highly defensive. From their review, Garety and Freeman (1999) concur that the SRIRT is likely to be a valid method of accessing covert self-evaluations.

The procedure used followed Bentall and Kaney (1996) who based their procedure on similar tasks used with depressed patients. Participants were first given a 30 item 'Self-Statement Questionnaire' with the instruction 'Please read the words below and decide if they describe you. Place a tick in the yes or no box'. The items used were the same as those used by Bentall and Kaney (1996), Dent and Teasdale (1988) and Williams, Healy, Teasdale, White and Paykel (1990). 12 of the words were positive in content and related to the concept of 'success' (successful, capable, important, dynamic, confident, entertaining, sociable, optimistic, respected, outgoing, valuable, skillful). Dent and Teasdale (1988) selected words rated low on depressive descriptiveness in a study by Derry and Kuiper (1981). The mean depressive descriptiveness rating was 1.67 on a scale in which 1 represented 'extremely not descriptive' and 9 represented 'extremely

descriptive'. 12 of the words had negative connotations and were related to the concept of 'failure' (deficient, stupid, unloved, weak, useless, incompetent, inferior, pathetic, unwanted, failure, worthless, inadequate). These words were selected from words rated high on depressive descriptiveness in a study conducted by J. Myers (see Teasdale and Dent, 1988). The mean rating for depressive descriptiveness for these 12 words was 8.13. As in Bentall and Kaney (1996), six neutral words were included, three at the start of the list and three at the end of the list, to control for primacy and recency effects (choosy, cautious, ordinary, realistic, neutral, modern). One word in each category was also presented in the EST, so prior exposure in the EST would not be expected to have a disproportionate effect on any one category. 'Realistic' was categorised as a neutral word in this test, despite its inclusion in the EST as a positive word. This word was retained, however, as in this test 'realistic' is neutral regarding the theme of 'success'. Each trait word was followed by two boxes labeled 'yes' and 'no'. Immediately after completing the questionnaire it was removed and the participant was told 'Please try to remember as many of the words that you have just read as possible'. Subjects were prompted 'See if you can remember any more' when they appeared to give up. The experimenter recorded the number of positive, negative and neutral words recalled.

3.5.4. Pragmatic Inference Test (PIT: Winters and Neale, 1985)(see Appendix Two)

The PIT was used as a covert measure of attribution style. The PIT was developed from the Attribution Style Questionnaire (Seligman *et al*, 1979) and requires participants to make attributional inferences when responding to an apparent memory test. Winters and Neale used the PIT with manic patients and found that, although remitted manic patients reported high self-esteem on conventional measures, they

displayed an attribution style similar to depressed patients when tested on the PIT. From their review of the literature relating to cognitive approaches to delusions, Garety and Freeman (1999) conclude that this method seems to be valid for covertly assessing attributional style.

The PIT consists of 12 short scenarios presented as self-referent vignettes that describe a situation in which both an external and an internal locus of causality is implied. Half of the situations describe successful outcomes and half describe failure outcomes. An example of a situation that is successful is being complimented on your appearance. In this case taking pride in your appearance (internal cause) and a colleague wanting a favour from you (external cause) are both implied in the vignette. An example of a failure is being unemployed and having difficulty finding work. In this case a poor personal work record (internal cause) and a poor job market (external cause) are implied. The anglicised version of the PIT as used by Lyon, Kaney and Bentall (1994) was used here to ease comprehension by British participants (e.g. 'Kurt Vonnegut' became 'John Fowles' and 'Thanksgiving' became 'Christmas'). Scenarios were randomly ordered. After each scenario was presented on audiotape, participants were instructed to respond to four written multiple choice questions. In one question (target), participants were asked to select which cause they remembered to be the main contributing factor in the outcome of the situation. In another question participants were asked for memory of implied information which did not relate to causality, and in the other two questions they were asked for memory of stated facts. Winters and Neale (1985) conducted pilot work using the PIT on a group of college students to ensure that the probability of endorsing either an internal or an external cause for each target item would not differ statistically from 0.5.

Target items are scored to give response frequencies in four categories: internal and external attributions for negative events and internal and external attributions for positive events. A PIT self-serving bias score is calculated by subtracting the number of internal attributions selected for negative events from the number of internal attributions made for positive events.

3.5.5. Hospital Anxiety and Depression Scale (HADS: Zigmond and Snaith, 1983)

The HADS was used as a measure of participants' level of anxiety and depression. The HADS is a self-assessment scale that was specifically developed for detecting states of anxiety and depression amongst medical patients. The scale, therefore, excludes items that might relate to the somatization of mood or to physical illness, for example, headaches, dizziness and loss of appetite.

The HADS is comprised of two subscales. The HADS-A measures anxiety and the HADS-D measures depression. Each subscale includes seven items, rated on a four-point scale (scored from 0 to 3). An example of an item from the HADS-D is 'I still enjoy the things I used to enjoy' ~ from (0) 'definitely as much' to (3) 'hardly at all'. An example item from the HADS-A is 'I feel tense or wound up' ~ from (3) 'most of the time'; to (0) 'not at all'. Scores over 10 for each subscale determine clinical caseness. Scores between 8 and 10 are 'borderline' cases (Zigmond and Snaith, 1982).

Herrmann (1997) reviewed over 200 studies that have used the HADS in medical settings with approximately 35,000 participants and summarised the available validation data. Herrmann concludes that 'the HADS is a reliable and valid instrument for assessing anxiety and depression in medical patients'. In particular, it was noted that with few exceptions, satisfactory or good item-total correlations have been found within the two

subscales and internal consistencies (Cronbach's alphas) have been acceptable, ranging from 0.80 to 0.93 for the HADS-A and from 0.81 to 0.90 for the HADS-D. The scale also has good retest reliability, $r > 0.80$ after up to two weeks, which decreases over time allowing sensitivity to mood change over time. The validity of the two dimensions has been confirmed by factor analysis in which one depression factor and one anxiety factor have been identified which correlate highly with the corresponding subscales ($r > 0.90$).

3.5.6. Attribution Style Questionnaire- parallel form (ASQ-pf: Lyon, Kaney and Bentall, 1994)

The ASQ-pf was used as an overt measure of attribution style in contrast to the PIT. This measure is based on the Attribution Style Questionnaire (Peterson, Semmel, von Baeyer, Abramson, Metalsky *et al*, 1982) which requires participants to provide possible explanations for hypothetical positive and negative events and then to rate these explanations on three seven-point scales. The scales represent internality (caused by self vs. caused by others/ circumstances), stability (unchangeable vs. changeable cause) and globality (affects all areas of life vs. affects only this area). Five-week test-retest reliability produced respectable correlations (range 0.57 to 0.70), which support the claim that the ASQ is measuring a style (Peterson *et al*, 1982). A major difficulty of the ASQ relates to the poor internal reliability of the subscales. Peterson *et al* (1982) estimated the internal reliability of each subscale using Cronbach's alpha obtaining 0.75 and 0.72 for the composite attribution style scales for good and bad events respectively. Factor analyses have identified internality as being independent from the stability and globality dimensions (Corr and Gray, 1996). Internal consistency for the six-item subscales (separating positive and negative situations) achieved a mean alpha of 0.54 (range 0.44 to 0.69). Furthermore,

the internality dimension has consistently yielded low reliability scores (Reivich, 1995; Tennen and Herzemberger, 1985). One possible reason for this is that the internality domain may not be unidimensional. Kinderman and Bentall (1996) identified a distinction between external attributions to a person and external attributions to circumstances. In its favour, the ASQ has been systematically contrasted with alternative measurement methodologies, and explanatory style based on free verbalizations converged well with ASQ scores, particularly on the internality dimension (Peterson, Bettes and Seligman, 1985; Schulman, Castellon and Seligman, 1989). Although other measures of attribution style have since been developed to overcome the low internal reliability for the subscales these have added new dimensions which are not included in the PIT (e.g. Kinderman and Bentall, 1986). The ASQ-pf remains the only measure devised to be used alongside the PIT. Despite its limitations, therefore, it will be used in this study, although caution must be taken when interpreting the data.

As the PIT replicates certain themes from the ASQ, Lyon, Kaney and Bentall (1994) developed the ASQ-pf to use alongside the PIT. Like the ASQ, the ASQ-pf asks participants to state causes for hypothetical successes and failures. Six positive and six negative situations are presented to participants. Scores for internality, stability and globality of attributions are calculated by adding up participants' ratings for positive and negative events independently. Self-serving bias scores are calculated by subtracting internality scores for negative events from internality scores for positive events (Bentall and Kaney, 1989).

Lyon, Kaney and Bentall (1994) constructed the ASQ-pf from a draft questionnaire of 40 ASQ-type items derived from new items from Peterson and

Villanova's (1988) expanded ASQ and items drawn up by researchers. 48 undergraduate medical students completed these items and the six best positive and negative items were selected based on whole correlations for internality, lack of skew and adequate variance. Lyon, Kaney and Bentall (1994) report that, as the best positive items did not have satisfactory psychometric qualities, a further 18-item scale was constructed using the six best negative items, the six best positive items and six additional positive items devised by researchers. These items were completed by 64 medical students and the final scale of six positive and six negative items was selected from this data. As part of a separate study, the final ASQ-pf and the original ASQ were administered to a mixed group of patients with CFS, depression and muscular dystrophy. Significant correlations were observed between the ASQ and ASQ-pf on measures of internality ($r=0.29$, $p=0.02$ for positive events; $r=0.41$, $p=0.002$ for negative events) and self-serving bias ($r=0.57$, $p<0.001$). One correlation between the ASQ and ASQ-pf (stability score for positive events) failed to reach significance, therefore any interpretation of the data pertaining to stability of attributions relating to positive events should be regarded with caution.

3.5.7. Bendig short form of the Taylor Manifest Anxiety Scale (B-MAS: Bendig, 1956)

The Manifest Anxiety Scale (MAS) (Taylor, 1953) and the Bendig short form of the MAS (B-MAS) are the most frequently used measures for measuring trait anxiety in order to identify defensive coping styles (Myers, 1993). The B-MAS is comprised of the 20 most consistently valid items from the 50 item MAS. Participants are advised 'Please read each statement and decide whether you feel in general that it is mostly true as applied to you or mostly false. Please circle the appropriate letter (T- True or F- False)

directly to the right of each statement'. Typical items include 'I believe I am no more nervous than others', 'I have had periods of such restlessness that I cannot sit for long in a chair'. Bendig (1956) reported that the median internal consistency reliability of the B-MAS was 0.76 compared to 0.78 for the longer MAS. Scores on the B-MAS were also highly correlated with scores on the MAS (0.93). Bendig concluded that the B-MAS 'is more parsimonious of testing time and probably more valid than the longer MAS'(page 384).

3.5.8. Marlowe-Crowne Social Desirability Scale (MC: Crowne and Marlowe, 1960, p350)

The MC is invariably used alongside an anxiety measure such as the B-MAS, in order to identify defensive coping styles (Myers, 1993). It is comprised of thirty-three statements to which the participant must respond 'True' or 'False'. Typical items are 'I never hesitate to go out of my way to help someone in trouble' and 'There have been occasions when I took advantage of someone'. In this study the MC and B-MAS items were presented as one questionnaire, so the instructions were the same as those described for the B-MAS above. The number of socially desirable responses are totaled to give a score out of 33. Crowne and Marlowe (1964) used the Kuder-Richardson formula 20 and obtained an internal consistency coefficient of 0.88. They also report a test-retest correlation of 0.88 from fifty-seven participants who completed the scale on two occasions, one month apart. Furthermore, the construct appears to be remarkably resistant to change. Strickland and Crowne (1963) reported that test-retest correlation for clinic outpatients over a five-month period was 0.68, despite having participated in twenty to twenty-five hours of psychotherapy.

3.5.9. Identifying coping style with the MAS and MC

Weinberger, Davidson and Schwartz (1979) used the MC and the MAS in order to identify four coping styles: repressive copers (highly MC - low MAS), low anxious (low MAS- low MC), high anxious (high MAS- low MC) and defensive high anxious (high MAS- high MC). Validity of the coping constructs has been supported by the tendency reported by for the groups to respond differently on various self-report, behavioural and physiological tasks (Weinberger *et al*, 1979).

The selection criteria to define the four coping styles have varied across studies. Some studies have screened a large number of potential participants and selected extreme scorers on anxiety and defensiveness (e.g. Weinberger *et al*, 1979). Other studies, particularly those studying patient groups where sample sizes are limited, have used the entire sample and, therefore, have not used such strict criteria, usually using median splits on trait anxiety and defensiveness scores (e.g. Jamner, Schwartz and Leigh, 1988). This has led to some difficulty in making comparisons between different studies. In order to make comparisons with other patient populations, and due to limited sample size in the present study from using patient populations, median splits will be used to compare the distribution of coping styles.

3.5.10. Fatigue Scale (Chalder, Berelowitz, Pawlikowska, Watts, Wessely *et al*, 1993)

In order to assess the severity of fatigue, an eleven-item fatigue scale was used based on The Fatigue Scale in order to measure the severity of fatigue. The scale is comprised of eleven items measuring two fatigue dimensions: physical and mental fatigue. Items include 'Do you have problems with tiredness' and 'Do you have difficulty

concentrating', to which participants tick the most appropriate response out of 'I am better than usual', 'No more/worse than usual', 'Worse than usual', 'Much worse than usual'. Responses are scored from 0 to 3, with 'Better than usual' scoring 0 and 'Much worse than usual' scoring 3. Chalder *et al* (1993) reported a Cronbach's alpha of 0.89 for the scale as a whole, and 0.85 and 0.82 for physical and mental fatigue respectively. The eleven-item scale used has been used with over 1,000 patients with CFS and with community samples and has been found to show good reliability (Chalder, personal communication).

CHAPTER FOUR

RESULTS

4.1 OVERVIEW

First the participants will be described. The analyses conducted to investigate each hypothesis will then be addressed in turn. Finally some further investigations are described in a preliminary attempt to address additional issues which arose from the tests of hypotheses.

Categorical data were analysed using Chi-square test. Interval data were tested for Normality and homogeneity of variance. Data that confirmed these assumptions were analysed using parametric statistics. Group effects were investigated using analyses of variance. Tukey's HSD test was used for post-hoc analyses, as this controls effectively for Type 1 error when comparing three groups (Howell, 1992). Significant group effects were covaried for HADS anxiety and depression scores and illness duration, due to the significant differences between the groups on these factors, and their potentially confounding effect.

The distribution of two variables differed significantly from Normality: negative word endorsement on the Self Referent Inferential Recall Test (Kolmogorov Smirnoff $Z=3.07$, $p<0.001$) and internal responses to negative events on the Pragmatic Inference Test (Kolmogorov-Smirnoff $Z=1.74$, $p=0.005$). Log and square root transformations were attempted but the data still did not approach a Normal distribution. For analyses involving these variables, nonparametric statistics were used.

4.2. DEMOGRAPHIC INFORMATION

Table 4.1 represents the participants' age, gender, ethnicity, occupation, education level reached, duration of illness and severity of fatigue.

A significant difference was found between the groups with regard gender, reflecting a greater proportion of male participants within the Diabetes group. There was a significantly higher proportion of participants who described their ethnic background as 'White British' in the CFS group. There were no significant differences between the proportions of the groups who had participated in further education (beyond GCSE/O level). Although all participants were between the ages of 18 and 30 years, participants in the Diabetes group tended to be older than the other groups. The Diabetes group had also been ill for significantly longer than the CFS group. As would be expected a significantly smaller proportion of the CFS group were in employment or education than either of the other two groups.

4.3. SYMPTOMS OF CHRONIC FATIGUE SYNDROME

In order to fulfil the Oxford Diagnostic Criteria for CFS, participants must have been suffering from persistent or relapsing fatigue for at least six months and suffer persistent or recurrent reduction in short-term memory and/or concentration. Participants reported a mean of 7.63 additional symptoms (sd=1.55, range 3-10). Table 4.2. represents the percentage of CFS participants who endorsed each symptom described by the Oxford Diagnostic Criteria (Sharpe *et al*, 1991). The mean score of the CFS group on the Fatigue Scale (Chalder *et al*, 1993) was 22.96 out of a maximum score of 33 (sd=6.12, range=5-33).

Table 4.1.
Demographic Information

	CFS N=24	Diabetes N=20	Healthy N=24	Statistic
Gender %female(n)	83.3 (20)	50 (10)	83.3 (20)	$\chi^2(2)=7.90^*$
Ethnicity %W.UK(n)	95.8 (23)	75 (15)	62.5 (15)	$\chi^2(2)=7.90^*$
Occupation % in employment/ education(n)	62.5 (15)	100 (20)	95.8 (23)	$\chi^2(2)=15.51^{***}$
Education % further education(n)	79.2 (19)	90 (23)	95.8 (18)	$\chi^2(2)=3.30$
Age, years (sd, range)	24.73 (4.07, 18-30.67)	26.51 (2.89, 19.42-30.58)	23.08 (3.35, 18.75-30.58)	F (2, 65)=5.21**
Illness duration, months (sd, range)	43.42 (26.40, 5-108)	131.55 (82.57,22-271)	N/A	F (1, 42)=24.45***

* p<.05 **p<.01 ***p<0.001

Table 4.2.

Symptoms reported by CFS participants

Symptom	% (n)
Sore throat	87.5 (21)
Painful neck or armpit glands	75 (18)
Muscle discomfort or pain	95.8 (23)
Joint pain	62.5 (15)
Headaches	100 (24)
Sleep problems	62.5 (15)

Table 4.3.

Illness attributions of Participants with CFS: ‘Which one of the following best applies?’

	% (n)
‘My illness is a physical one’	37.5 (9)
‘My illness is mainly physical’	20.8 (5)
‘Both physical and psychological factors are involved in my illness’	41.7 (10)
‘My illness is mainly psychological’	0 (0)
‘My illness is psychological in nature’	0 (0)

4.4. TESTING THE HYPOTHESES

4.4.1. Physical illness attributions are associated with high levels of self-esteem amongst people with CFS. Psychological attributions are associated with low levels of self-esteem.

4.4.1.(a) Illness attributions in CFS

Table 4.3. represents the responses of CFS participants when asked which illness attribution applied to them. Just over half of the participants attributed their illness to predominantly physical factors ('physical' or 'mainly physical') (58.3%). The remaining participants cited both psychological and physical factors as being involved in their illness (41.7%). No participants reported that their illness was predominantly psychological.

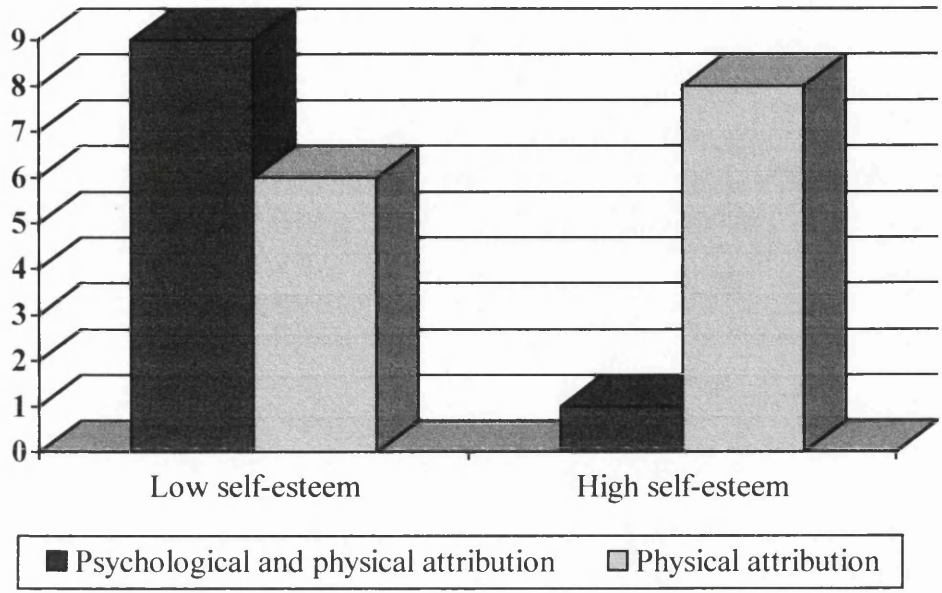
4.4.1.(b) Physical illness attributions and self-esteem

In order to investigate the relationship between illness attribution and self-esteem a composite self-esteem score was calculated from the number of positive trait words endorsed plus the number of negative trait words not endorsed in the word endorsement task of the Self-Referent Inferential Recall Test (maximum=24). The number of positive trait words endorsed and negative words not endorsed correlated highly (Spearman's $\rho=0.394$, $p<0.001$). CFS participants were divided in to two groups, above and below the median composite self-esteem score (median=20). A significant relationship was found between illness attribution and self-esteem ($\chi^2(1)=5.53$, $p=0.019$). Participants in the CFS group who had lower self-esteem (scores below the median self-esteem score) were more likely to cite both psychological and physical factors as the cause of their illness, see Figure 4.1.

Figure 4.1.

Illness Attribution and Self-Esteem amongst Participants with CFS

Frequency



4.4.2. Participants with CFS will not report low levels of self-esteem or a depressogenic attribution style when tested on overt measures when compared to a healthy control group.

4.4.2.(a) Depression amongst participants with CFS

Table 4.4 demonstrates the prevalence of a Major Depressive Episode in the three groups. Only one participant, in the Diabetes group, reported currently experiencing a Major Depressive Episode at the time of testing. A significant difference was found between the number of participants in each group who had a lifetime history of a Major Depressive Episode. This was most common amongst the participants with CFS, followed by those with Diabetes. There was not a significant difference between the number of participants with CFS and Diabetes who reported having experienced a Major Depressive Episode prior to the onset of their illness. On the Hospital Anxiety and Depression Scale, significant differences were found between the groups on anxiety and depression scores reflecting highest scores in the CFS group, followed by the diabetes group then the healthy comparison group. As shown in Table 4.5, anxiety and depression scores were not significantly correlated with length of illness. Anxiety, depression and illness duration all correlated significantly with fatigue score.

Table 4.4. Prevalence of Depression and Anxiety

	CFS N=24	Diabetes N=20	Healthy N=24	Statistic
Current RDC Major Depressive Episode % (n)	0 (0)	5 (1)	0 (0)	$\chi^2(2)=2.44$
Lifetime history of RDC Major Depressive Episode % (n)	62.5 (15)	40 (8)	20.8 (5)	$\chi^2(2)=8.62^*$
History of RDC Major Depressive Episode prior to illness % (n)	25 (6)	10 (2)	N/A	$\chi^2(2)=2.69$
HADS- Anxiety /21 Mean (sd, range)	9.42 (3.94, 2-18)	6.35 (2.94, 0-11)	5.71 (2.49, 1-10)	F (2,65) = 9.08 ***
HADS- Depression /21 Mean (sd, range)	6.29 (3.47, 1-13)	3.65 (3.18, 0-10)	1.96 (1.92, 0-8)	F (2,65) = 13.4 ***
HADS- Anxiety cases % (n)	20.8 (5) borderline 54.2 (13) case	15 (3) borderline 15 (3) case	29.2 (7) borderline 0 (0) case	$\chi^2=22.76$, df=4, p<0.001
HADS- Depression cases % (n)	20.8 (5) borderline 16.7 (4) case	15 (5) borderline 0 (0) case	4.2 (1) borderline 0 (0) case	$\chi^2=11.63$, df=4, p=0.02

* p<.05 **p<.01 ***p<0.001

Table 4.5.

Pearson Correlations of Length of Illness, Fatigue, Anxiety and Depression

	Length of illness N=44	Fatigue Score N=68	HADS-Anxiety N=68	HADS-Depression N=68
Length of illness		-0.38*	-0.29	-0.18
Fatigue Score			0.54***	0.69***
HADS-Anxiety				0.58***

* p< .05 ** p< 0.01 ***p< 0.001

Table 4.6.

Participants with CFS' reports of guilt during a Major Depressive Episode:

'During the most severe period were you bothered by feeling guilty or down on yourself?'

	Yes % (n)	No % (n)
CFS	93 (14)	7 (1)
Diabetes	75 (6)	25 (2)
Healthy comparison	20 (1)	80 (4)

4.4.2 (b) Preservation of self-esteem amongst depressed CFS participants

As shown in Table 4.6, the majority of CFS participants who reported having experienced a Major Depressive Episode in this study reported having ‘felt guilty or down on yourself’ during that episode (93%).

On the word endorsement task of the Self-Referent Inferential Recall Test, no significant differences were found in the number of positive words that were endorsed by the groups (see Table 4.8), however, the CFS participants endorsed the most negative words, followed by the Diabetes group. The group effect for the composite self-esteem score approached significance (Kruskal-Wallis $\chi^2(2)= 4.96$, $p=0.08$).

The self-esteem score was highly correlated with HADS anxiety ($\rho=-0.6$, $p<0.001$) and depression ($\rho=-0.4$ $p=0.001$), see Figure 4.3.

4.4.2 (c) Attribution style

In order to compare the attribution styles of the three groups, repeated measures analysis of variance were carried out. The repeated measures were positive and negative events. All groups made attributions which were more internal, global and stable for positive events compared to negative events (see Table 4.7). No significant group effects or interaction (group x event) were found, although the mean scores reflect a tendency towards a depressive attribution style amongst the CFS group (see Figure 4.2).

Figure 4.2.

Responses to the Attribution Style Questionnaire (Parallel Form)

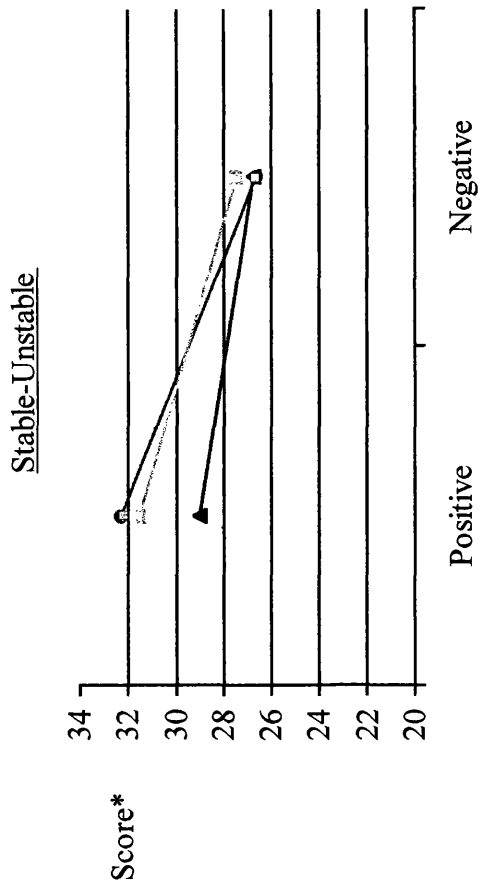
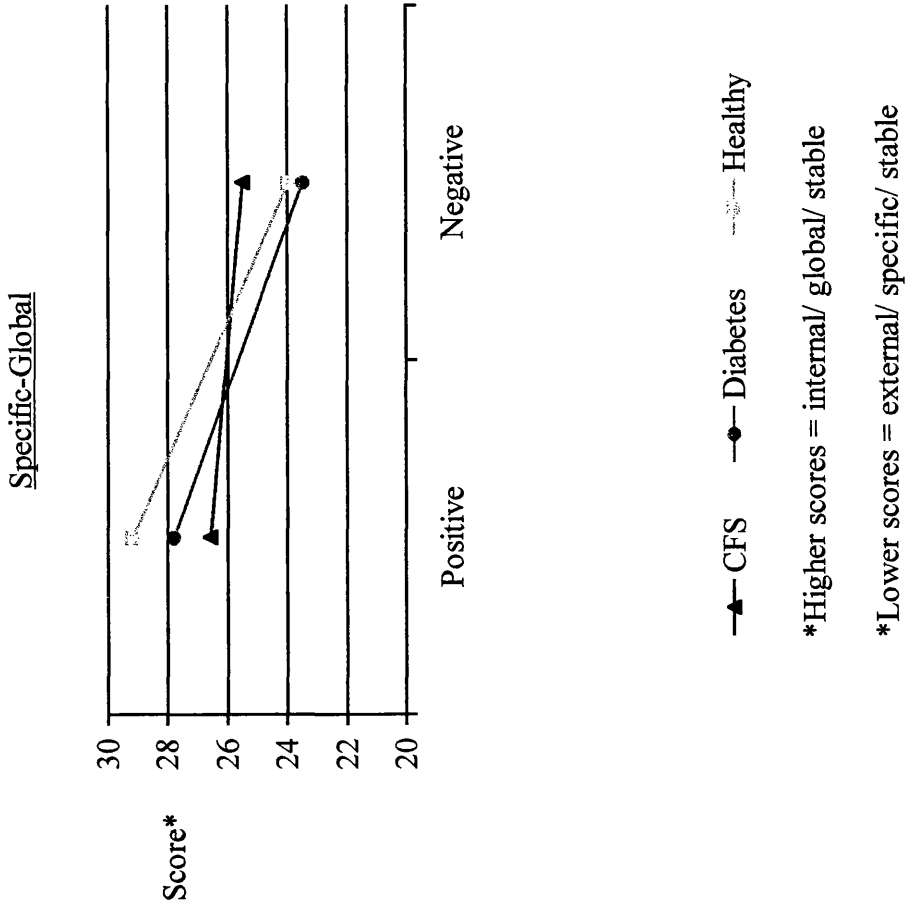


Table 4.7.

Total Ratings for Positive and Negative Events on the Attribution Style Questionnaire (Parallel Form)

Dimension	Positive events			Negative events			F (2,65)		
	CFS N=24	Diabetes N=20	Healthy N=24	CFS N=24	Diabetes N=20	Healthy N=24			
Internality Mean (sd, range)	25.46 (5.12,13-37)	27.16 (7.23, 15-36)	27.04 (4.80, 19-34)	24.29 (5.30, 12-36)	22.60 (5.34, 14-31)	23.79 (4.12, 14-30)	11.98 ***	0.16	1.33
Globality Mean (sd, range)	26.58 (6.01, 14-39)	27.79 (5.81, 17-36)	29.17 (4.55, 18-36)	25.50 (5.50, 10-34)	23.50 (8.09, 12-42)	23.96 (6.07, 15-35)	12.95 ***	0.18	1.77
Stability Mean (sd, range)	29.00 (3.89, 20-39)	32.31 (3.56, 23-37)	31.54 (4.57, 18-38)	26.75 (5.11, 14-39)	26.65 (5.73, 17-42)	27.46 (4.55, 20-38)	33.92***	1.44	2.10

* p= .05 ** p= 0.01 ***p= 0.001

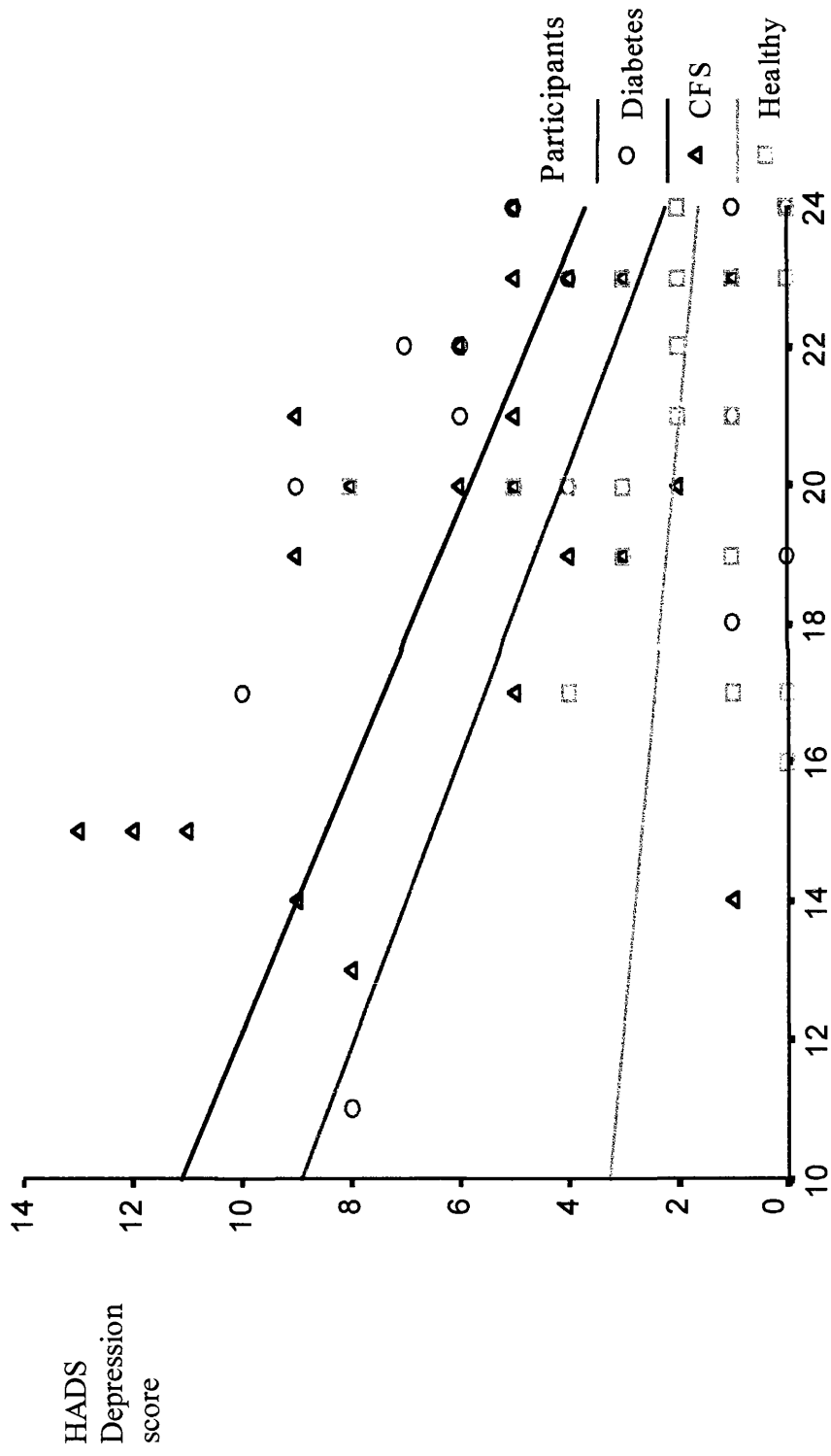
Note

Attribution ratings are made for six positive and six negative events on a seven point scale.

7 = internal/ global/ stable attribution; 0=external/ specific/ unstable attribution

Figure 4.3.

Self-esteem and Depression amongst Participants with CFS, Diabetes and a Healthy Comparison Group



Self-esteem composite (/24)

Table 4.8.

Number of Positive and Negative Words Endorsed on the Self Referent Inferential Recall Test:

	CFS N=24	Diabetes N=20	Healthy N=24	Kruskal-Wallis $\chi^2(2)$
Positive words endorsed Mean (sd, range)	8.08 (2.99, 3-12)	9.5 (2.35, 4-12)	9.25 (2.38, 5-12)	2.92
Negative words not endorsed Mean (sd, range)	11.0 (0.93, 9-12)	11.55 (1.15, 7-12)	11.79 (0.51, 10-12)	13.44***
Self-esteem score Mean (sd, range)	19.08 (3.49, 13-24)	20.85 (3.12, 11-24)	21.04 (2.56, 16-24)	4.96

* p<.05

**p<.01

***p<0.001

4.4.3. Participants with CFS will show low levels of self-esteem and a depressogenic attribution style when tested on covert measures compared to a healthy control group. A comparison chronic illness group will not exhibit a discrepancy between responses to overt and covert measures.

4.4.3.(a) Self-Referent Inferential Recall Test

Repeated measures analysis of variance revealed significant effects for word type, with all groups recalling more positive than negative words (see Table 4.9). A significant group effect was found. Post-hoc tests, controlling for Type 1 error, identified a significant difference between the CFS and healthy participants on recall of positive words (Tukey HSD, $p=0.02$). HADS-depression was a significant covariant ($F(1,64)=4.22$, $p=0.04$), however, and this accounted for the group effect found.

4.4.3.(b) Emotional Stroop Test

All groups took longer to colour name words than 'O's (see Table 4.10). A significant group effect was found. Post-hoc tests controlling for Type 1 error indicated that the CFS participants were significantly slower to colour name all word types than two comparison groups (Tukey HSD, $p<0.001$).

When the time to name positive and negative trait words was compared, a significant group effect was found ($F(2, 64)=8.75$, $p<0.001$). The effect of HADS-depression was significant ($F(1,64)=4.55$, $p=0.04$). Overtly reported self-esteem did not have a significant independent effect. After covarying for depression, the interaction between word type and group approached significance ($F(2,64)=2.75$, $p=0.07$). Figure 4.4 demonstrates a greater increase in the time to name negative words compared to positive words for the CFS group, and a slight reduction in the

time to name negative words compared to positive words for the healthy comparison group.

In order to further investigate the role of physical illness attributions in protecting self-esteem, the illness attributions of CFS participants (predominantly physical versus physical and psychological) were compared for those who were slower to colour-name positive words and those who were slower to colour-name negative words. No significant relationship was found between these factors ($\chi^2(1)=0.97, p=0.30$).

Table 4.9.

Number of Positive and Negative Words Recalled on the Self-Referent Inferential Recall Test

	Positive words		Negative words		F (2,65)		
	Mean (sd, range)	Diabetes N=20	Healthy N=24	Mean (sd, range)	CFS N=24	Diabetes N=20	Healthy N=24
Recall	3.96 (1.68, 1-8)	4.60 (2.01, 1-9)	5.29 (1.40, 3-8)	2.88 (1.80, 0-8)	3.20 (1.67, 0-7)	3.79 (1.14, 1-6)	29.89 ***
							4.00 *
							0.008

* p= .05

** p= 0.01

***p= 0.001

Table 4.10

Time to Colour-Name (Seconds) on the Emotional Stroop Test

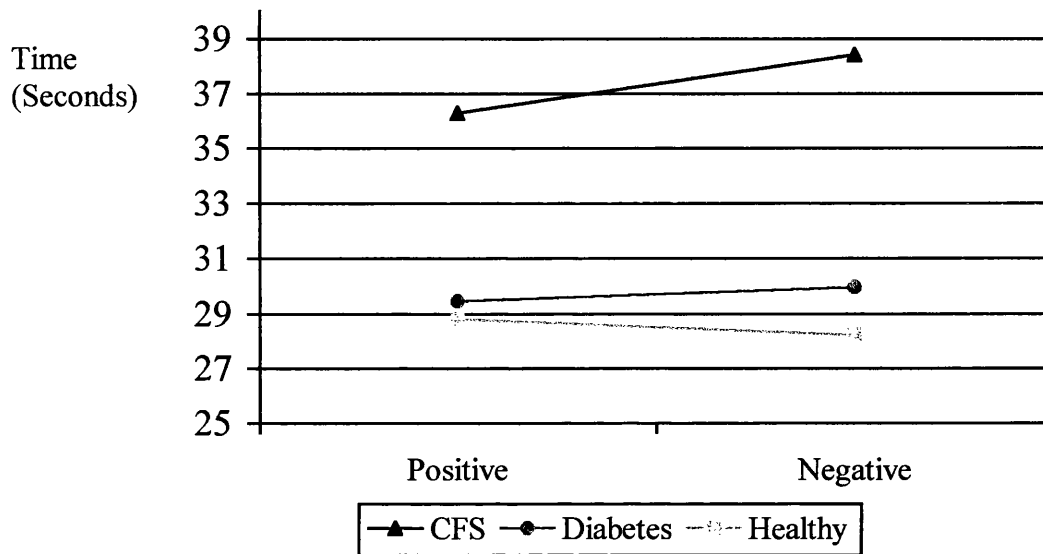
	CFS N=24	Diabetes N=20	Healthy N=24	Word F(3,61)	Group F(2,63)	Group x Word F(6,124)
Os				6.88 ***	8.46 **	1.78
Time (Secs)	30.91 (5.12,	25.40 (4.16,	25.35 (4.08.			
Mean (sd, range)	22.22-43.47)	18.94-35.75)	19.91-34.16)			
Neutral words	37.71 (7.89,	29.97 (4.36,	28.39 (5.04,			
Time (Secs)	24.81-56.21)	22.88-39.97)	20.0-39.06)			
Mean (sd, range)						
Positive words	36.29 (6.78,	29.46 (3.81,	28.80 (5.06,			
Time (Secs)	25.25-55.6)	23.53-36.44)	19.56-39.37)			
Mean (sd, range)						
Negative words	38.42 (7.89,	29.97 (4.36,	28.23 (4.19,			
Time (Secs)	24.81-56.21)	22.88-39.97)	20.72-35.75)			
Mean (sd, range)						

* p= .05 ** p= 0.01 ***p= 0.001

Figure 4.4.

Time to Colour-Name Positive and Negative Words (Seconds) on the Emotional

Stroop Test



4.4.3.(c) Pragmatic Inference Test

The number of internal responses made by each group were compared using the Kruskal-Wallis Test. Significant group effects were not found for positive or negative words (see Table 4.11).

In order to compare the self-serving bias found by the PIT and the ASQ-pf, discrepancy indices was calculated as follows:

$$\frac{\text{ASQ-pf internalising score} - \text{PIT internalising score}}{\text{Total possible ASQ-pf internalising score} - \text{Total possible PIT internalising score}}$$

Where 'ASQ-pf internalising score' =

Total internalising scores for positive events – total internalising scores for negative events

'PIT internalising score' =

Total internalising responses for positive events - total internalising responses for negative events

The discrepancy indices were split in to those above and below 0, where indices below 0 represent more internalising for positive events on the PIT, and scores above 0 represent more internalising for positive events on the ASQ-pf. A significant group effect was not found ($F(2,64)=1.12, p=0.33$).

Table 4.11.

Number of Internalising Responses on the Pragmatic Inference Test

	CFS N=24	Diabetes N=20	Healthy N=24	Kruskal-Wallis $\chi^2(2)=$
Positive events				
Mean (sd, range)	2.33 (1.24, 0-6)	2.65 (1.18, 1-5)	1.17 (1.49, 0-5)	0.69
Negative events				
Mean (sd, range)	2.08 (1.21, 1-5)	1.7 (1.17, 0-5)	2.25 (1.22, 1-5)	1.41

* p < .05

** p < 0.01

*** p < 0.001

4.4.4. Participants with CFS will display a greater tendency to hold a defensive high anxious coping style, characterised by high levels of reported trait anxiety and high levels of defensiveness on self-report measures compared to a healthy control group and comparison chronic illness group.

As shown in Table 4.12, a significant group effect was found on the Manifest Anxiety Scale ($F(2, 65)=11.58, p<0.001$). Post-hoc tests, controlling for Type 1 error, revealed significantly higher scores in the CFS group, compared to the Diabetes (Tukey HSD, $p=0.003$) and the Healthy comparison groups (Tukey HSD, $p<0.001$). The groups did not differ significantly on the Marlowe-Crowne Social Desirability Scale ($F(2,65)=0.84, p=0.44$).

The groups were divided into four coping styles, using the median splits method. The median score on the Manifest Anxiety Scale was 17, and the median score on the Marlowe-Crowne Social Desirability Scale was 8. B-MAS scores greater than 8 were defined as high anxiety. MCSDS scores greater than or equal to 17 were defined as high social desirability. Table 4.13 and Figure 4.5 display the proportion of participants within each group with each coping style. A significant difference was found between the groups for the proportion of Defensive High Anxious participants ($\chi^2(2)=6.53, p=0.038$), reflecting a greater number of Defensive High Anxious participants in the CFS group compared to the two comparison groups

Table 4.12

Manifest Anxiety Scale and Marlowe-Crowne Social Desirability Scale Scores

	CFS N=24	Diabetes N=20	Healthy N=24	F (2,65)
Marlowe-Crowne Social Desirability Scale	17.38 (5.98, 7-26)	15.25 (5.47, 4-24)	15.92 (5.38, 6-27)	0.84
Mean (sd, range)				
Bendig-Manifest Anxiety Scale	11.96 (4.45, 2-20)	7.30 (4.53, 1-17)	6.13 (4.28, 1-14)	11.59 ***
Mean (sd, range)				

* p < .05 ** p < 0.01 *** p < 0.001

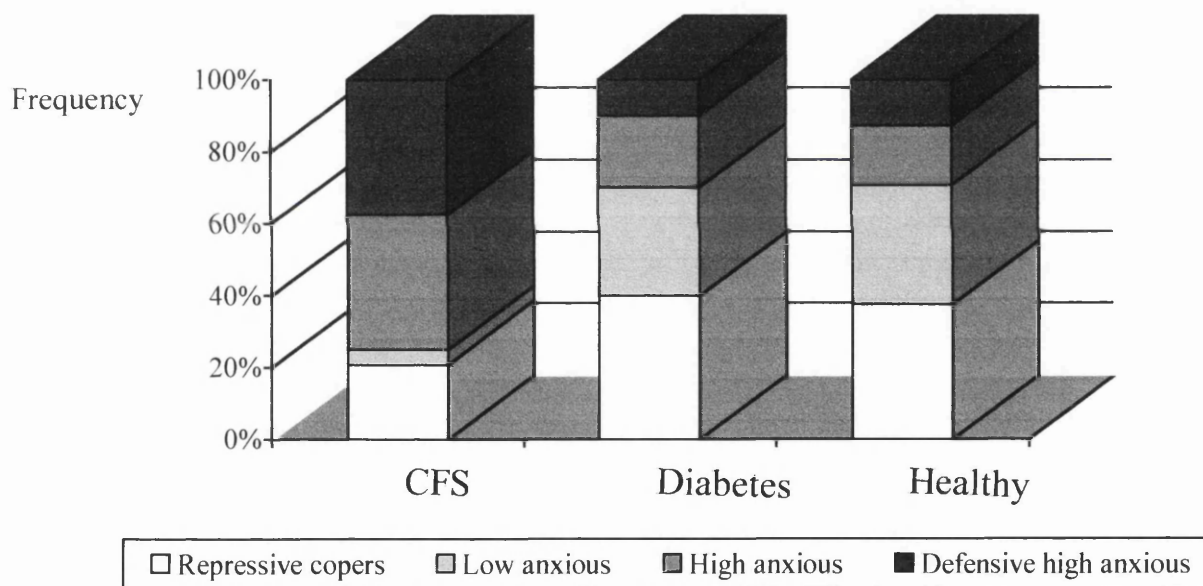
Table 4.13

Coping Styles

	Defensive high anxious % (n)	Repressive copers % (n)	Low anxious % (n)	High anxious % (n)
CFS (/24)	37.5 (9)	20.8 (5)	4.2 (1)	37.5 (9)
Diabetes (/20)	10 (2)	40 (8)	30 (6)	20 (4)
Healthy(/24)	12.5 (3)	37.5 (9)	33.3 (8)	16.7 (4)

Figure 4.5.

Coping Styles



4.5. FURTHER INVESTIGATIONS

4.5.1. The relationship between illness attributions and attribution style

Table 4.14 summarises participants with CFS's responses to the question 'What do you think is the cause of your fatigue?' The three most common themes related to the presence of a virus or other illness, behaviour in response to illness (i.e. overdoing it) and stress. There was no difference in the number of participants in each group who cited a virus as contributing to the cause of their illness (79% 'Physical/Mainly physical' attribution; 80% 'Both physical and psychological' attribution). More people in the 'Physical/ Mainly physical' group cited a role of behaviour ('overdid it') (57% compared to 40%). More people in the 'Both physical and psychological' group cited a role of 'stress' (60% compared to 21%).

A significant relationship was found between the presence of a self-serving attribution style and illness attributions amongst participants with Chronic Fatigue Syndrome ($\chi^2(1)=4.03$, $p=0.045$). As shown in Figure 4.6, those participants who held a self-serving attribution style (i.e. a tendency to attribute positive events to internal causes and negative events to external causes) were more likely to make a physical attribution for their illness. Those who held a self-disparaging (depressive) attribution style (i.e. a tendency to attribute negative events to internal causes and positive events to external causes) were more likely to attribute a role to psychological factors in their illness.

Table 4.14

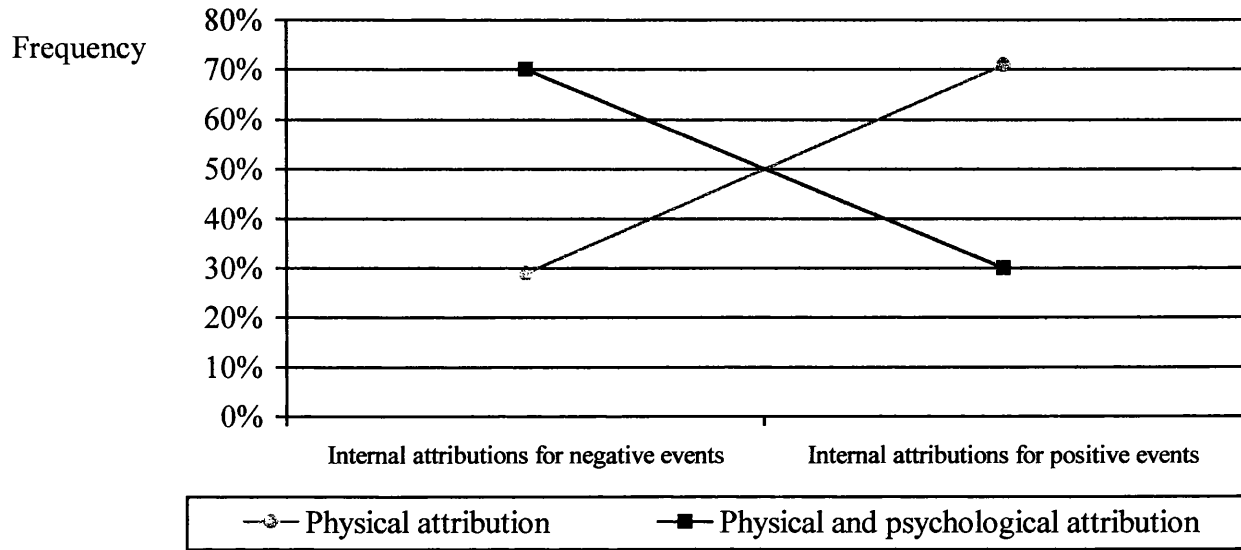
Summary of participants with CFS responses to ‘What do you think is the cause of your fatigue?’

Attribution classified by participant as physical/mainly physical (N)	Attribution classified by participant as: both physical and psychological (N)
Virus (4)	Virus and antibiotics (1)
Virus and overdid it (1)	Virus and overdid it (1)
Virus, stress and overdid it (3)	Virus, stress and overdid it (2)
Virus and overdid it (personality) (1)	Virus and stress (3)
Other illness and overdid it (2)	Other illness and overdid it (1)
Overdid it (1)	
Trauma (1)	
Accident and pregnancy (1)	Other illness and stress (1)
	Stress (1)

Figure 4.6.

Internal Attributions for Positive and Negative Events on the Attribution Style

Questionnaire (Parallel Form) and Illness Attributions of Participants with CFS



4.5.2. Characteristics of participants with CFS who have not experienced a Major Depressive Episode

Comparisons within the CFS group, of those who did or did not report having experienced a Major Depressive Episode, involve small numbers of participants.

Figure 4.7 displays the coping styles of these two groups of participants. Although the relationship between coping style and report of a Major Depressive Episode does not reach statistical significance ($\chi^2(2)=5.42, p=0.14$) a greater proportion of those who had not experienced a Major Depressive Episode held a repressive coping style (4/9) than those who had experienced a Major Depressive Episode (1/15). A smaller proportion of those who had experienced a Major Depressive Episode are classified as Defensive High Anxious (2/9 compared to 6/15).

There are no clear differences between the two groups with regards interference effects from positive or negative words on the Emotional Stroop test. Figure 4.8, however, displays the discrepancy indices for the PIT and the ASQ-pf. A greater proportion of those who did not report having experienced a Major Depressive Episode held a greater self-serving bias on the ASQ-pf (6/9) than on the PIT, compared to those who did report having experienced a Major Depressive Episode (5/15).

Figure 4.7.

Coping Styles of CFS Participants who have and have not experienced a Major Depressive Episode (MDE)

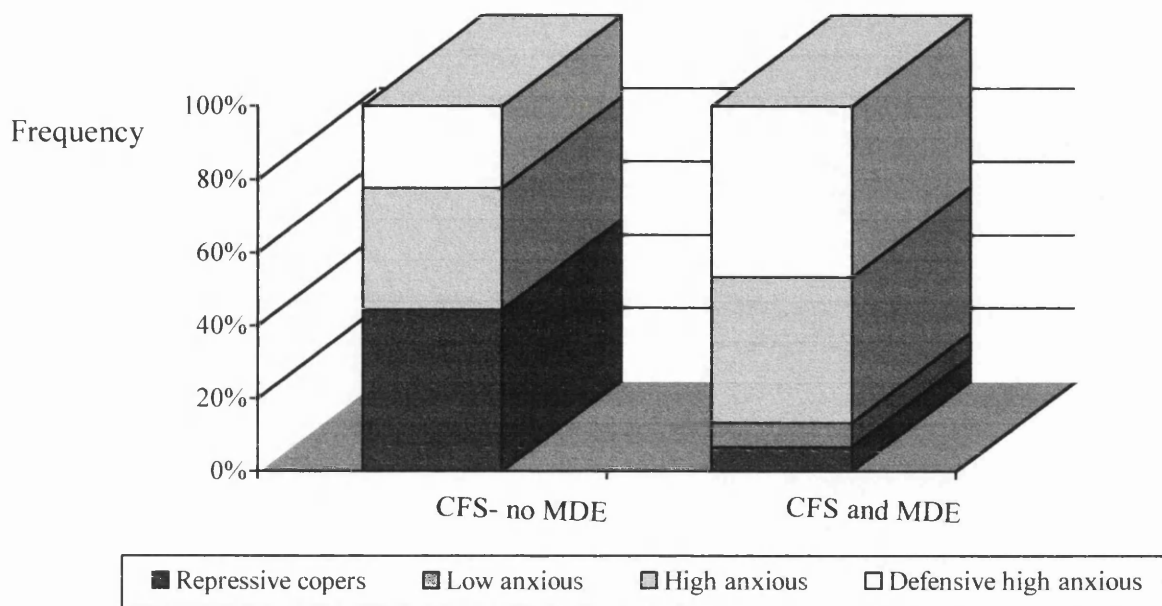
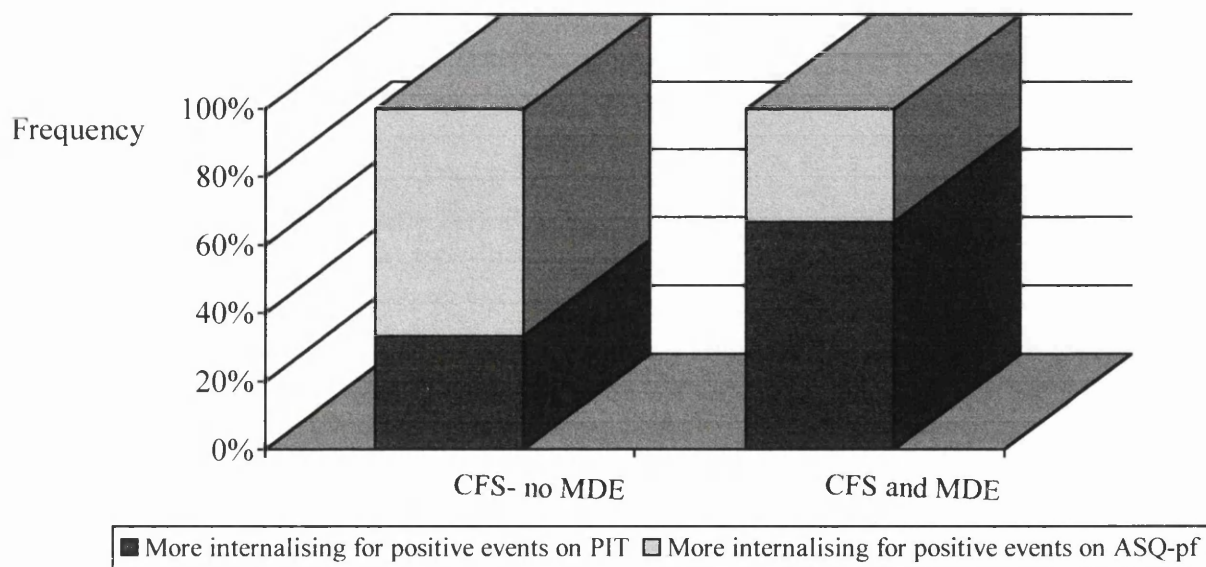


Figure 4.8.

Discrepancies between Internalising Attributions on the Attribution Style Questionnaire (Parallel Form) and the Pragmatic Inference Test for CFS Participants who have and have not experienced a Major Depressive Episode (MDE)



CHAPTER FIVE

DISCUSSION

5.1. OVERVIEW

The present study aims to investigate the hypothesis that people with CFS have low levels of self-esteem which are defended by physical attributions for illness and a defensive high anxious coping style. The findings are consistent with the hypothesis that people with CFS have a tendency to have low underlying levels of self-esteem. The findings did not suggest that physical illness attributions act as a defence mechanism, although participants with CFS were more likely to hold a defensive high anxious coping style.

The main findings are summarised as follows:

1. Physical illness attributions were associated with higher overtly reported self-esteem in participants with CFS. Physical illness attributions were not associated with lower underlying levels of self-esteem but reflected participant's general attributional style.
2. Participants with CFS experienced higher levels of anxiety and depression than the healthy and chronic illness comparison groups. Guilt, self-esteem and attributional style appeared to be associated with depression.
3. Participants with CFS demonstrated greater interference from negative words on the Emotional Stroop Test, a covert measure of self-esteem, than the two comparison groups. This was not fully accounted for by depression or overtly reported self-esteem.
4. A greater proportion of participants with CFS displayed a Defensive High Anxious coping style compared to the two comparison groups. A greater

proportion of participants with CFS who had not been depressed held a Repressive Coping style, than those who had been depressed.

The four hypotheses will be discussed in turn. Possible explanations for differences from previous studies and limitations of the current study are discussed in relation to each hypothesis. More general limitations are then reviewed. Finally the implications of the findings of the present study for future research and clinical practice will be discussed.

5.2. HYPOTHESIS ONE

Physical illness attributions are associated with high levels of self-esteem amongst people with CFS. Psychological attributions are associated with low levels of self-esteem

5.2.1. *Illness attributions in CFS*

The present study replicated the findings of Euba *et al* (1996) who found that 56% of patients with CFS in their hospital-based sample cited a predominantly physical cause for their illness. In the present study 58.3% of the sample made a predominantly physical attribution. This is a significantly smaller proportion than had been found in older studies which reported that over 80% of CFS participants made physical illness attributions (e.g. Powell, Dolan and Wessely, 1990; Wessely and Powell, 1989). These studies recruited participants from highly specialised settings that generally received referrals from medical consultants. The settings from which CFS participants were recruited in the present study generally receive referrals directly from General Practitioners. In the last ten years, CFS has also received a great deal of research and media interest, changes in people's understanding of CFS is highly likely to influence illness attributions.

Ray, Jeffries and Weir (1995) suggest that how the participants are asked about the cause of their illness may affect their response, in particular using the words ‘non-physical’ rather than ‘psychological’ may increase the acceptability of this response. Ray, Jeffries and Weir found that a lower rate of participants attributed their fatigue to ‘wholly physical’ or ‘mainly physical’ causes (34.9%). Participants were hospital outpatients. In the present study participants were asked about ‘physical’ and ‘psychological’ causes so it is possible that the results may overestimate the tendency to make ‘physical’ illness attributions.

The differences between the studies may reflect differences in the participants recruited. In the present study illness attributions were not found to be associated with duration or severity of fatigue, or gender. The present study was restricted to participants between the ages of 18 and 30 years. Younger people with CFS may be more accepting of a role of psychological factors in illness, due to cultural changes amongst patients and medical professionals, which have increased the acceptability of seeking help for psychological difficulties. If this is so it might suggest that for this group psychological illness attributions may not represent a negative moral evaluation, protecting self-esteem from threat, but simply a labeling process. This will be discussed further in Section 5.9.

5.2.2. Physical illness attributions and self-esteem

A significant relationship was found between illness attribution and levels of self-esteem, as measured by endorsement of positive and negative words, amongst participants with CFS. Participants with CFS who had lower levels of self-esteem were more likely to make an illness attribution that involved both physical and psychological factors. This finding is consistent with Powell, Dolan and Wessely’s (1990) suggestion that physical illness attributions defend vulnerable levels of self-

esteem amongst people with CFS, as those people who made physical illness attributions tended to report higher levels of self-esteem. Clearly, there are other explanations for this relationship. For example, those people with CFS who do experience depression may find that this exacerbates their symptoms of fatigue, and hence make an illness attribution which involves both physical and psychological factors. Alternatively, if a person with CFS experiences depression, they may be more likely to hold themselves responsible for the occurrence of negative events (self-disparaging attributional style). It would be consistent with this style to blame oneself (make a psychological attribution) for the occurrence of illness (a negative event). Discussion of this relationship will be returned to in Section 5.6.

5.2.3. Summary

58.3% of participants made a predominantly physical attribution for their illness. Physical illness attributions were associated with higher levels of self-esteem. This pattern would be expected if illness attributions serve to protect self-esteem against potential threat. Alternative functions of illness attributions will be discussed in Section 5.9.

5.3. HYPOTHESIS TWO

Participants with CFS will not report low levels of self-esteem or a depressogenic attributional style when tested on overt measures when compared to a healthy control group.

5.3.1. Depression and Anxiety amongst participants with CFS

The present study found a similar lifetime history of depression amongst participants with CFS as Wessely and Powell (1989). In their study 67% of participants with CFS, according to Oxford Diagnostic Criteria, in a U.K. hospital

setting had experienced a Major Depressive Episode, based on the Schedule for Affective Disorders and Schizophrenia (SADS) (Spitzer, Endicott and Robins, 1978).

In the present study, 62.5% of CFS participants reported a lifetime history of depression, using the same assessment instrument. It is striking that this figure is similar despite the lower age of participants in the present study (the participants in Wessely and Powell's (1989) study were between the age of 35 and 40 years). This appears to reflect, however, the tendency for a Major Depressive Episode to first occur after the onset of CFS, with only 25% of CFS participants reporting a Major Depressive Episode prior to their illness (although this is still higher than would be expected in the general population).

Although only one participant, in the Diabetes group, fulfilled criteria for a current Major Depressive Episode, current levels of anxiety and depression were higher amongst the CFS group than the two comparison groups. Over half (54.2%) of the CFS participants fulfilled clinical caseness criteria of Anxiety and 16.7% fulfilled criteria for clinical caseness of Depression on the Hospital Anxiety and Depression Scale (HADS)(Zigmond and Snaith, 1983). The HADS is likely to be more sensitive to mood changes than the SADS, as responses are rated on a scale rather than in a 'yes' or 'no' format. Participants also completed the HADS in questionnaire form and the SADS in interview form, which may have led to different responses (see Section 2.6).

Anxiety and Depression scores on the HADS correlated highly with severity of fatigue as measured by the Fatigue Scale (Chalder *et al*, 1993). The HADS was designed for use with physically ill populations, and hence puts less emphasis on the somatic symptoms of anxiety and depression. There is still some overlap, however, with items in the fatigue scale, in particular, the two items 'I feel as if I am slowed

down' (Depression) and 'I feel restless as if I have to be on the move' (Anxiety). Interestingly, this anxiety item would be expected to have an inverse relationship with fatigue. Although depression may be over-estimated in CFS by the HADS, it is unlikely that the relationship between anxiety and fatigue is accounted for solely by overlap of items in the two scales.

The duration of fatigue had an inverse relationship with fatigue severity, anxiety and depression. This suggests that people who had CFS longer in this sample may have had less severe symptoms, experienced improvement in their symptoms or rated their experience differently due to the longer experience of fatigue. Lower levels of anxiety and depression may reflect the relationship with lower ratings of severity of fatigue, as discussed above.

5.3.2. Preservation of self-esteem amongst depressed participants with CFS.

5.3.2.(a) Guilt

In contrast to Powell, Dolan and Wessely (1990), who found that 78% of depressed participants with CFS reported experiencing no guilt, the majority of participants with CFS who reported having experienced a Major Depressive Episode in the present study reported that they had been 'bothered by feeling guilty or down on [themselves]' (93%).

5.3.2.(b) Self-esteem

Powell, Dolan and Wessely (1990) also reported that the majority of CFS participants, including depressed participants did not report reduced levels of self-esteem. This was not replicated by the present study, in which only 37.5% of participants with CFS did not report reduced levels of self-esteem, compared to 75% of the participants with Diabetes and 83.4% of the Healthy comparison group.

Powell, Dolan and Wessely (1990) used the SADS to investigate self-esteem. This involves asking participants questions such as ‘How do you feel about yourself?’ in an interview format. In the present study self-esteem was measured as part of the Self-Referent Inferential Recall Test in which participants report whether positive and negative trait words apply to them, in a questionnaire format. This measure was used so that a direct comparison could be carried out with an indirect measure of self-esteem based on word recall. It is possible, however, that the differences with the findings of the present study relate to the mode of questioning. Although the word-endorsement test has been classified as a ‘direct’ measure of self-esteem, it is possible that measures may fall along a continuum of directness, and that the word-endorsement test used here is less direct than the structured interview method adopted by Powell, Dolan and Wessely (1990). This is consistent with Myers (in press) who reported that participants with a repressive coping style made conflicting reports about their relationships with their fathers depending on whether they were responding to interviews or structured questionnaires.

In the present study self-esteem, as assessed by positive and negative word endorsement, correlated highly with anxiety and depression scores on the HADS for all groups of participants. This is in contrast to the findings of Powell, Wessely and Dolan (1990) who found that impaired self-esteem was not associated with depression in participants with CFS. This may reflect differences in the populations studied, as discussed in Section 5.2, or in the mode of assessing self-esteem as discussed above. The findings of the present study are consistent with Johnson *et al* (1997) and Moss-Morris and Petrie (1996) who concluded that, on the whole, people with CFS report good self-esteem, unless they are depressed.

5.3.2.(c) *Attributional style*

The attributional style scores of the CFS participants for internality, stability and globality did not differ significantly from the two comparison groups. The general pattern of responses did appear to be more self-disparaging than the other two groups, i.e., more internal, stable and global attributions for negative events. This would be expected due to higher levels of depression amongst the CFS group.

The Attributional style Questionnaire (parallel form) was used in the present study as it can be used alongside the indirect attributional style test, the Pragmatic Inference Test. This version has been found to correlate highly with the original Attributional Style Questionnaire on all dimensions except stability. The Attributional Style Questionnaire has been used extensively, however, low reliability scores have been found for the internality dimension. It has been suggested that this reflects the presence of distinct dimensions of externality, for example attributing an event to a person or to circumstances. Despite these limitations, the results of the present study resemble those of another larger study using this measure with participants with CFS. Chubb *et al* (1999) found that responses of CFS patients with concurrent depression resembled those of depressed participants, whilst the responses of non-depressed CFS patients resembled those of healthy subjects. This study involved 50 participants with CFS, 37 participants with Depression and 100 healthy controls and, hence, had greater power to detect a difference than the present study. This issue will be returned to in Section 5.7.

5.3.3. *Summary*

A greater number of participants in the CFS group than the two comparison groups reported a lifetime history of a Major Depressive Episode. Current levels of depression and anxiety were also higher amongst the participants with CFS. Levels

of guilt, self-esteem and attributional style were consistent with the reported degree of depression, in contrast with the hypothesis that participants with CFS do not report impairments on these measures, despite being depressed.

5.4. HYPOTHESIS THREE

Participants with CFS will show low levels of self-esteem and a depressogenic attributional style when tested on covert measures when compared to a healthy control group. A comparison chronic illness group will not exhibit a discrepancy between responses to overt and covert measures.

5.4.1. Self-Referent Inferential Recall Test

Participants with CFS recalled a smaller number of positive words compared to negative words than the two comparison groups. The higher HADS depression scores accounted for this group effect. There is no suggestion, therefore, that CFS is associated with lower levels of self-esteem on covert tests that cannot be accounted for by reported levels of depression.

Bentall and Kaney (1996) carried out separate analyses on their data from participants with persecutory delusions and depression looking at recall of words that had or had not been endorsed in the earlier phase of the task. This analysis was, unfortunately, not possible in the present study due to the ceiling effect found for positive word endorsement and the floor effect for negative word endorsement, and the lower number of words recalled by the participants with CFS.

5.4.2. Emotional Stroop Test

Participants with CFS were slower to colour-name all words (and non-words) on the Emotional Stroop Test, which is consistent with reports of slower processing speed on effortful tasks amongst people with CFS (Tiersky *et al*, 1997). The

participants with CFS were also slower to name negative words compared to positive words, than the two comparison groups. Although this effect was partially accounted for by HADS depression scores, the group effect continued to approach significance. Overtly reported self-esteem did not have a significant independent effect. This finding is consistent with the hypothesis that participants with CFS have underlying low levels of self-esteem, which are not fully accounted for by their levels of depression or overtly reported self esteem.

In order to investigate the role of physical illness attributions in protecting self-esteem, the illness attributions of CFS participants (predominantly physical versus physical and psychological) were compared for those who were faster to colour-name positive words and those who were faster to colour-name negative words. No significant relationship was found between these factors. This does not support Powell, Dolan and Wessely's (1990) suggestion that by holding on to physical attributions for CFS, individuals avoid a challenge to their consciously held view of themselves as a worthwhile person which overlies unconscious feelings of inadequacy.

5.4.3. Pragmatic Inference Test

Participants with CFS did not differ significantly to the two comparison groups on their responses to the Pragmatic Inference Test. Discrepancy Indices were calculated in order to compare participants' responses on the direct measure, the Attributional Style Questionnaire (parallel form) and the indirect measure, the Pragmatic Inference Test. The CFS participants did not differ from the two comparison groups with regards the discrepancy between the direct and indirect measure of internal attributions for positive and negative events.

5.4.4. Summary

Discrepancies between overt and covert measures were not found using the Self Referent Inferential Recall Test or the Pragmatic Inference Test. On the Emotional Stroop test, however, participants with CFS displayed greater interference from negative words than positive words. Depression scores were significantly associated with the degree of interference, but the group effect continued to approach significance even after covarying for this. This supports the hypothesis that covert testing reveals lower levels of self-esteem than would be expected from the level of depression, unlike the levels of self-esteem revealed by overt testing.

5.4.5. *Why were results of the covert tests inconsistent?*

The Pragmatic Inference Test (PIT) has been used in previous studies with participants with persecutory delusions (Lyon, Kaney and Bentall, 1994) and participants with mania (Winters and Neale, 1985). Both of these groups demonstrated a discrepancy between their ‘intact’ responses to overt self-esteem or attribution measures and their ‘depressive’ response pattern on the PIT. Lyon, Kaney and Bentall (1994) suggest that the discrepancy found may reflect distinct ‘implicit’ and ‘explicit’ representations of the self. It is possible, however, that responses to the PIT *are* susceptible to response bias from ‘explicit’ self-representations in other groups of patients who are not convinced that the test is purely testing memory. A number of participants did, indeed, comment that the test did not seem to be testing their memory. Interestingly, very few participants in the CFS group commented on this. This may reflect their reduced confidence in their cognitive abilities, or greater willingness to please the experimenter (as would be consistent with higher, though not significantly, scores on the Marlowe-Crowne Social Desirability Scale). There is, therefore, reason to be cautious about the results from the PIT, as in the groups being

tested in the present study the test may have been more susceptible to the same conscious processes as the Attributional style Questionnaire.

Measures of information-processing biases are likely to be less susceptible to self-presentation bias. Two such measures were used in the present study: the Self-Referent Inferential Recall Test (SRIRT) and the Emotional Stroop Test. The tests both compare performance on words associated with 'positive' and 'negative' self-esteem. In the present study the same words were used for the SRIRT as have been used with other patient groups (Dent and Teasdale, 1988; Williams *et al.*, 1990; Bentall and Kaney, 1996). The results of the SRIRT in the present study were limited, however, as, the full analysis following Bentall and Kaney (1996) could not be conducted. As in the current study, Bentall and Kaney did not find a group effect on the total number of positive and negative words recalled, the expected group differences were only found when only words which were endorsed were looked at. They tentatively suggest that recall of endorsed adjectives will give a better indication of self-schemata as these words were involved in a process of elaboration and organisation. In the current study this analysis was confounded by the overall tendency for participants to endorse positive words and not endorse negative words, and the generally low recall scores amongst the CFS participants.

The hypothesised group effect on the Emotional Stroop test approached significance. More highly significant results may have resulted from a larger sample size (see Section 5.7) and some procedural changes. One word was changed in the neutral condition of the Emotional Stroop from those used by Kinderman (1994), changing 'pale' to 'left', due to the possible association of 'pale' with illness. The words which made up the positive and negative conditions were based on value ratings made by healthy volunteers (Anderson, 1968). Whether the same value

judgements would be made now, thirty years later, and by patients with medical conditions is not certain. Anecdotal reports have suggested that people with CFS place increased value on achievement (Ware and Kleinman, 1992). Particular words may, therefore, have particular resonance, such as 'weak' and 'failure' and others might not, such as 'calm', 'realistic'. In order to address this, a computerised version of the Emotional Stroop could be used in which the time to colour-name can be evaluated independently for each word.

5.5. HYPOTHESIS FOUR

Participants with CFS will display a greater tendency to hold a defensive high anxious coping style, characterised by high levels of reported trait anxiety and high levels of defensiveness on self-report measures when compared to a healthy control group and comparison chronic illness group.

5.5.1. Trait anxiety

As expected, participants with CFS scored significantly more highly on the measure of trait anxiety than the two comparison groups.

5.5.2. Defensive High Anxious Coping Style

When the groups were divided according to Weinberger, Schwartz and Davidson's (1979) classification of coping styles, a greater proportion of the participants with CFS were Defensive High Anxious as predicted. In fact the proportion of Defensive High Anxious CFS participants (37.5%) was not only higher than the chronic illness comparison group in this study (10%), it was also substantially higher than has been found in other patient populations. Jensen (1987) used the same cut off points with median splits as the present study, yet found lower levels of Defensive High Anxious participants in participants with advanced breast cancer (16%), recurrence free breast cancer (26%) and patients undergoing minor

surgery (23.5%). The result does not, therefore, appear to simply be an artefact of increased anxiety associated with having a physical illness, although genuine High Anxious coping was also more prevalent amongst participants with CFS than the two comparison groups.

5.5.3. Summary

In support of the hypothesis, more participants with CFS exhibited a Defensive High Anxious coping style. It has been suggested that Defensive High Anxious individuals represent 'Failed Repressors'. Section 5.6.3 describes the characteristics of participants with CFS who reported never having experienced a Major Depressive Episode in order to begin to investigate whether this is because these participants continue to 'effectively' use the Repressive Coping style. The implications of holding defensive coping styles for health are discussed in Section 2.5.

5.6. FURTHER INVESTIGATIONS

5.6.1. The relationship between illness attributions and attributional style

As described in Section 5.2, the relationship found between illness attributions and self-esteem is in the direction which would be predicted were illness attributions a form of defence mechanism against vulnerable underlying self-esteem. Powell, Dolan and Wessely (1990) suggest that by holding on to physical attributions for CFS, individuals avoid a challenge to their consciously held view of themselves as a worthwhile person which covers up underlying feelings of inadequacy. The results of the current study, however, suggest that participants with CFS did not necessarily report low levels of guilt associated with depression, and reported less than optimal levels of self-esteem, concordant with levels of depression. Results

from the Emotional Stroop test suggest that low underlying self-esteem is not fully accounted for by depression and overtly expressed self-esteem. No association, however, was found between interference on the Stroop Test from positive or negative words and illness attribution.

Other functions of illness attribution might account for the relationship between reported self-esteem and illness attribution. Brewin and Antaki (1987) draw attention to the separate functions of ordinary explanation, which include labelling or description, moral evaluation, causal attribution and self-presentation. Within CFS research, there has been a tendency to view illness attributions as moral evaluations, in which a psychological attribution is seen as a sign of weakness of character. Individuals maintain a physical illness attribution to protect against this threat to self-esteem (e.g., Powell, Dolan and Wessely, 1990). The responses of participants in the current study have not supported the view that illness attributions protect vulnerable self-esteem. More recent studies have suggested that illness attributions are labels that may influence behaviour. Physical illness attributions may lead to avoidance of exercise, which has a deleterious effect on prognosis unless this is successfully challenged (Deale, Chalder and Wessely, 1998).

If illness attributions reflected a labelling process, the relationship between more psychological attributions and lower self-esteem might result from the experience of depression exacerbating symptoms of fatigue. If illness attributions reflect a process of moral evaluation about the self, people with CFS who experience depression, may be more likely to hold themselves responsible for the occurrence of negative events in general (self-disparaging attributional style). It would be consistent with this style to blame oneself (make a psychological attribution) for the occurrence of illness (a negative event). The current study is not in a position to

adequately address this. In order to have a tentative look at these possibilities, however, the qualitative explanations for CFS which were given by participants with CFS who made more or less physical illness attributions were compared. The three most common themes related to the presence of a virus or other illness, behaviour in response to illness (i.e. overdoing it) and stress. Viral attributions were cited equally by both groups. Those who rated their attribution as predominantly physical cited behaviour in response to illness more frequently and those who rated their attribution as 'both physical and psychological' cited stress more frequently. It is striking that there is not a great deal of difference between the explanations offered and frequently the same explanation, e.g. the combination of having a virus, being under stress and overdoing it, might be labelled 'physical' by some and 'physical and psychological' by others.

This might suggest that the attribution rating given was more representative of a general attribution or coping style. The significant association found between attributional style on the Attributional Style Questionnaire (parallel form) and illness attribution supported this. Those participants with CFS who held a self-disparaging attributional style (i.e. a tendency to attribute negative events to internal causes and positive events to external causes) were more likely to include psychological factors in their illness attribution. This suggests that how a participant rates their illness attribution may, therefore be a function of negative affect (via a general attributional style). These results are difficult to interpret at present. It has been suggested that participants who make physical illness attributions tend to have worse outcomes from cognitive behavioural therapy (Sharpe *et al*, 1992). Some treatment outcome studies, however, have found that those with lower mood (associated here with *less* physical illness attributions) have worse outcomes from cognitive behaviour therapy

(Wilson *et al*, 1994; Butler *et al*, 1991), although others have not (Bonner *et al*, 1994; Deale *et al*, 1997).

Research findings relating to illness attributions have been extremely inconsistent in relation to prognosis from treatment. Based on the findings of the current study it is suggested that this may be because illness attributions may not reflect a judgement which is stable over time, but like attributional style are liable to change, particularly in accordance with mood (e.g. Golin, Sweeney and Schaeffer, 1981). Furthermore, the function, and hence the impact, of the illness attribution may vary from person to person. Given the lack of attention to these distinctions within the field of outcome studies of CFS it is, perhaps, not surprising that the findings have been inconsistent. It is important that future research pays more attention to the function of the illness attribution for individuals in order to identify whether it is likely to have an impact on prognosis.

5.6.2. *Summary*

Illness attributions have received a great deal of attention within the CFS research literature, in particular because some studies have reported an association between outcome from therapy and illness attributional style, although results have been inconsistent. The current study found that illness attributions tend to mirror an individual's general attributional style, which is likely to reflect their mood at the time of testing. It is suggested that the function of illness attributions needs to be more carefully addressed, in order to ascertain whether there are some individuals for whom illness attributions play a greater role in recovery and to inform appropriate clinical interventions.

5.6.3. Characteristics of participants with CFS who have not experienced a Major Depressive Episode

It has been suggested that individuals with CFS attempt to protect vulnerable levels of self-esteem. In the current study, however, participants with CFS did report high levels of depression and anxiety, reduced self-esteem and higher levels of guilt than the two comparison groups. In the majority of participants with CFS, therefore, if they did attempt to maintain high levels of self-esteem, their attempts had not been wholly successful. There was a minority of participants with CFS, however, who reported not having experienced a Major Depressive Episode. Indeed, these participants' responses to questions about depression were similar to those described by a number of authors, for example 'I'm not the sort of person to get depressed' (Surawy *et al*, 1995; Chalder, Power and Wessely, 1996; Powell, Dolan and Wessely, 1990; Katz and Andiman, 1988).

Those participants with CFS who did and did not report having experienced a Major Depressive Episode were compared. The comparisons involved small numbers of participants, and so the conclusions drawn are only speculative at this stage. The findings suggest a tendency for those who reported not having had a Major Depressive Episode to hold a Repressive Coping style, while those who reported having had a Major Depressive Episode were more likely to be Defensive High Anxious. This is particularly interesting in relation to the suggestion that Defensive High Anxious individuals may represent 'failed repressors', i.e. repressors for whom these coping mechanisms have become ineffective. It has been suggested that both these coping styles may impinge on physical well being, particularly via the hypothalamic-pituitary adrenal axis (Jamner, Schwartz and Leigh, 1988). This is clearly relevant to CFS, as increasing evidence is being provided that patients with

CFS have an abnormal physiological responses to stress related to reductions in hypothalamic-pituitary- adrenal axis functioning and neurotransmission (Wessely, Hotopf and Sharpe, 1997).

As might be anticipated to occur alongside a Repressive Coping style, the group who reported not having had a Major Depressive Episode were also more likely to have a more self-disparaging bias when their attributional style was assessed indirectly than directly. Just as Repressive Copers tend to report low levels of anxiety, although physiological measures suggest high levels of anxiety (Asendorpf and Scherer, 1983), this discrepancy suggests participants outwardly report higher self-competence than genuinely reflects their underlying beliefs. The mechanisms responsible for this discrepancy are currently unclear, although it has been suggested that Repressive Copers appear to act consistently with a process of self-deception rather than other-deception (Derakshan and Eysenck, 1999).

5.6.4. Summary

Participants with CFS who reported having not experienced a Major Depressive Episode, were more frequently categorised as Repressive Copers. Those who reported having experienced a Major Depressive Episode were more frequently Defensive High Anxious. This is consistent with the view that Defensive High Anxious individuals are repressors who no longer find their coping strategy effective. Both defensive groups have been found to have abnormal hypothalamic-pituitary-adrenal axis functioning, as have individuals with CFS. The convergence of these two areas of research in future studies would be of great interest.

5.7. LIMITATIONS OF THE PRESENT STUDY

5.7.1. Sample size

Group differences approached significance on the Attributional Style Questionnaire (parallel form) and the Emotional Stroop Test. More significant effects may have been found with a larger sample size. The sample size in the present study was determined by a power analysis based on data obtained by Kinderman (1994) using the Emotional Stroop test. This test is an indirect measure of self-esteem, and hence is crucial to this study's hypotheses. Although the measure has not been used with participants with CFS, Kinderman (1994) administered the test to three groups including individuals who experience persecutory delusions which were hypothesised to develop as a means of coping with low self-esteem, just as rigid beliefs in CFS are proposed to develop as a means of coping with low self esteem. Unfortunately the study by Kinderman (1994), and other studies using indirect measures of self-esteem and attribution, was based on a small number of participants (16 in each group). Based on this study, a power of 80% would be achieved from a sample size of 10 participants in each group. Because of the small sample used by Kinderman, and hence high probability of error, a larger sample size was aimed for and at least twenty participants formed each group. The present study attempted to reduce the amount of error between the groups by restricting the samples to people between 18 and 30 years of age, and matching the participants for gender as far as possible. Due to practical constraints, while it was possible to match the healthy comparison and CFS groups for gender, it was not possible to match the diabetes group. Tests of the hypotheses of the present study did not, however, reveal significant effects of gender on the measures used. Future investigations would benefit from a larger sample size.

5.7.2. Type 1 Error

Multiple comparisons were kept to a minimum in order to avoid Type 1 error. When multiple comparisons were carried out, statistical corrections were conducted. Tests were restricted to the hypotheses based on previous research and theory, with the exception of the further investigations described. The conclusions of which should remain tentative.

5.7.3. Confounding variables

Significant group effects were covaried for anxiety, depression and duration of illness. The groups also differed in the distribution of gender and ethnic background. Gender was not entered in to the analyses as the CFS and healthy comparison groups were matched for gender and only the group with Diabetes differed. The Diabetes group was not found to be different to the other two groups in any of the tests of hypotheses.

Although more of the CFS group described their ethnic background as ‘White British’ than the two comparison groups, a requirement of the study was that the participants spoke English as their first language and all participants were born in the United Kingdom or Eire.

People with CFS have been found to exhibit cognitive deficits, in particular on tests relating to speed or efficiency of processing (Tiersky *et al*, 1997). In accordance with this, the present study found that CFS participants were slower to colour-name on the Stroop task and recalled less words on the Self-Referent Inferential Recall Test. In order to test the hypotheses, therefore, the relationship between performance with positive and negative material was assessed so the results were not confounded by cognitive deficits. The low level of word recall on the Self-

Referent Inferential Recall Test did, however, limit the extent of the analysis that could be carried out, as described in Section 5.4.

5.7.4. Generalisation of the results

The present study selected patients with CFS who fulfilled the Green College, Oxford criteria as this definition has been most widely used within the British population and does not make any assumptions about the psychiatric status of people with CFS. Some controversy continues regarding necessary and sufficient conditions for the diagnosis of CFS. The results of this study can not, therefore, be generalised to other chronically fatigued groups who do not fulfil the Oxford criteria, without further investigation.

The age of participants was restricted to 18 to 30 years in this study for two reasons. First, a limitation of many studies of CFS is the tendency for the CFS participants to be younger than depressed or chronic illness controls. The present study, therefore, restricted the age range of all three groups and recruited comparison participants who have a chronic medical condition that affects young people (Insulin treated diabetes). Second, as discussed by Ray (1991) CFS may be a heterogeneous condition. Although at this time, it is not clear how the overall category of CFS may be divided, one possibility is that a different condition affects older and younger people with CFS. Although, this is highly speculative, it may be that by restricting the age range of the group, a more homogeneous sample results. In support of this, the descriptions of symptoms provided by the CFS participants do reflect an overall tendency for them to experience similar somatic symptomatology. By restricting the sample studied, in this way, however, it is not possible to generalise the results to all people with CFS, in particular those who are not in this age range.

Recent studies have attempted to investigate whether there are differences in the presentation of CFS amongst those who have experienced CFS for a longer or shorter duration. Friedberg, Dechene, McKenzie and Fontanetta (2000) reported that their long-duration CFS (4-7 years) group had significantly higher CFS symptom severity scores, largely attributable to increased cognitive difficulties. Illness duration was not, however, found to be a significant factor in the hypotheses investigated in the present study, although severity of fatigue measured by the Fatigue Scale did vary widely.

A number of studies have found differences between samples recruited from primary and tertiary care services, in particular, socio-economic status and the tendency to make physical illness attributions (Euba *et al*, 1996). Due to the differences found between these groups the current study was restricted to participants recruited through specialist clinics for people with CFS. Referrals to these services were generally received from General Practitioners. The rate of physical illness attributions was equivalent to that found in the tertiary care group by Euba *et al* (1996). Female participants were over represented in the present study, as in other studies. Euba *et al* (1996) report that 68.4% of their hospital sample were female. In this study 83.3% of participants were female, which may reflect a female bias towards expressing interest in participating in the study. It is not possible at this stage, therefore, to generalise the conclusions of this study to all patients, in particular those seen outside tertiary services, without further investigation.

The participants in the present study were informed from the outset that a psychologist was conducting this study. It is possible that higher rates of reporting guilt and low self-esteem were found due to the participation of patients who are more open to psychological theories about CFS. A number of participants described

being 'wary' of taking part because of previous experiences of delegitimation. It is highly likely that many other potential participants did not volunteer to take part for similar reasons, and that these people might be more wary about reporting guilt and low self-esteem.

5.8. IMPLICATIONS FOR FUTURE RESEARCH

A number of areas have been identified in the current study that would be of interest in future research. The following areas, in particular, remain largely untouched by the literature to date. The current study, however, suggests that they are likely to be important areas for further investigation.

- (i) The function of illness attributions in CFS in relation to their impact on recovery.
- (ii) The prevalence of Defensive High Anxious and Repressive Coping styles amongst individuals with CFS.
- (iii) The relationship between defensive coping styles and depression in people with CFS.
- (iv) The relationship between hypothalamic-pituitary- adrenal axis functioning and defensive coping styles in CFS.
- (iii) The discrepancies between overt and covert measures of self-esteem and attribution amongst individuals with CFS who have not been depressed.

A number of general implications are also apparent. First, the current study supports previous findings that some participants will respond differently to different modes of data collection (e.g. Myers, in press). There may not be a simple distinction between measures presented 'overtly' and 'covertly', and very subtle differences

may affect a participant's response. In large studies, these differences would be expected to be cancelled out across the groups. Unless, of course, one group is particularly sensitive to particular modes of assessment, such as repressive copers and, perhaps, non-depressed participants with CFS. It is essential for researchers to clearly describe their methods of data collection, and to consider the role that the methods may have in obtaining certain results.

A chronic illness control group was included in the current study to help identify whether the group differences are a result of the experience of chronic illness or are more specific to CFS. The inclusion of comparison groups who are depressed or anxious, but do not have CFS, would help further define which aspects are specific to CFS. Comparison with other chronic illness groups that have features in common with CFS, such as Chronic Pain Syndrome which is also a poorly understood condition, will also be useful. Furthermore, it cannot be assumed that those factors that appear to be specific to individuals with CFS *predispose* a person to CFS. Prospective studies are necessary. A very good example of such a study is that of Imboden, Canter and Cluff (1959) who carried out psychological assessments as an influenza epidemic was expected to hit the country. Studies will also need to take account of the potentially additive effects of different factors, and their reciprocal effects on each other (Ray, 1991).

It is important to also acknowledge the additional points made by Ray (1991), who stated that any working model of CFS should recognise that (i) CFS may be a non-specific response, with multiple somatic and psychological causes; (ii) CFS may be a heterogeneous condition. The present study has identified psychological processes that may be more common amongst participants with CFS, but are by no means universal. Further research will need to identify more clearly whether people

with CFS can be classified in ways which guide more effective treatment. In the mean time an individualised approach is warranted in therapy, as discussed further below.

A further implication for future research comes from the apparent convergence of the separate fields of study of neuroendocrinology, immunity and coping mechanisms in the general population and in individuals with CFS. The need for multi-disciplinary research has, perhaps, never been so great as it is in the study of CFS.

5.9. IMPLICATIONS FOR CLINICAL PRACTICE

The findings of the current research are consistent with the claim of Sharpe's CBT model, that people with CFS tend to have low underlying levels of self-esteem. It is suggested that a defensive coping style may be utilised to protect against this. It is tentatively suggested that some individuals with CFS are able to successfully protect their self-esteem using a Repressive Coping style. Others were less successful at protecting their self-esteem and this was associated with a Defensive High Anxious style. No evidence was found to suggest that illness attributions act as a defence mechanism, although the different functions that they may serve for different individuals has been discussed.

As described in Section 2.4, participation in Cognitive Behaviour Therapy (CBT) has been found to be associated with improved physical functioning and mood and reduced fatigue. The findings of the present study support the involvement of cognitive components of therapy, although these should be individually tailored to each patient. It is important to include therapeutic processes that enhance feelings of self-esteem and self-competence, even if these concerns are not explicitly voiced by

the patient. The probability of high levels of depression and, particularly, anxiety should also be addressed. With regards the relationship between depression and anxiety, it is tentatively suggested that a subgroup of individuals with CFS may be vulnerable to anxiety and depression and adopt defensive coping strategies to manage this. Illness or other stressful life events may then trigger the breakdown of these coping mechanisms, leading to an increased rate of anxiety and depression amongst people with CFS.

It is likely that a defensive attributional style may be adaptive in certain situations, particularly if an individual can successfully repress signs of distress. In a work situation, for example, an individual might be able to continue to perform in a difficult environment without (consciously) feeling under pressure. This coping style might no longer be adaptive, however, if the individual were struck down by an illness such as a virus, which necessitated a period of recuperation. The individual might keep on working, not acknowledging the impact that it is having on her physical well-being until she can simply no longer function. In fact, during the study the model being investigated was discussed with participants. The most common response was for participants to concur with an explanation of CFS which involved a tendency to keep going in a situation which they have since acknowledged was difficult (for example due to ill health or high work load), although this had not been appreciated at the time. In this situation cognitive components of CBT might be beneficial in addressing schema and core beliefs which might lead to the development of a defensive coping style. Clinical research is necessary to establish effective strategies for working clinically with defensive coping styles, particularly in the light of research suggesting repressive coping is a process of self (rather than other) deception (Derakshan and Eysenck, 1999). Schwartz (1990) describes the

process of psychotherapy with a number of teenagers ‘whose biological, psychological and social functioning was compromised as a consequence of pervasive repression’.

‘Patients were encouraged to reinterpret their prior coping styles as being remarkably adaptive...They were helped to discover that this coping strategy was no longer necessary or adaptive... With time patients became more hopeful, more realistic and more open minded and saw therapy as a positive challenge that in a safe context could free them.’ (Schwartz, 1990; p 430)

This description of psychotherapy with teenagers who held defensive coping styles, also draws attention to the point that there is no a priori reason why CBT will be the only successful intervention for people with CBT, although it may be necessary for a successful intervention to promote alternative coping styles. Interventions that involve behavioural pacing exercises may promote an alternative coping style sufficiently for some individuals

Fulcher and White (1997) found a comparable improvement amongst patients who completed a graded exercise programme to that found in trials of CBT. Wessely, Hotopf and Sharpe (1999) suggest that the graded exercise programme also addresses patients’ fears relating to attribution of their illness to a physical cause, such as the thought that exercise is damaging. This is consistent with the suggestion that illness attributions are simply a label, and providing a new label for patients allows them to practice more adaptive behaviour. This is similar to the process hypothesised to take place in patients with Panic Disorder who are assisted to discover that hyperventilation is a symptom of panic and not a sign of an impending heart attack (Clarke, 1986). The current study has found that illness attributions reflect general

attributional style. It has been proposed that attributional style may serve various functions, and these might differ for different people in different situations. With regards illness attributions in CFS labelling and moral evaluation functions have been implied in the literature. In clinical practice it will be important to assess the function of illness attributions with individual patients. If, for example, the illness attribution represents a label, graded exercise may be sufficient to overcome fears associated with the physical illness label and subsequent avoidance of activity. If, on the other hand, the illness attribution represents a moral evaluation the cognitive components of therapy might need to be worked on before a patient is willing to undergo graded activity. It is likely that patients may drop out of therapy that encourages them to act in conflict with their attributions if their attributions reflect mechanisms for protecting their view of themselves.

Finally, it is important to stress that although patterns relating to psychological processes have been identified, these represent trends amongst the CFS population studied. Although more of the CFS participants may have displayed certain attributes, these were by no means universal to the whole population studied. It is crucial to acknowledge the points made by Ray (1991), as discussed above, in particular: (i) CFS may be a non-specific response with multiple somatic and psychological causes, and (ii) CFS may be a heterogeneous condition. The primary clinical implication, therefore, is that therapeutic approaches for people with CFS should involve a thorough individualised formulation. The psychological mechanisms described in the present study are likely to have reciprocal effects with other variables to a greater or less extent for different individuals.

5.10 CONCLUSIONS

The findings of the present study provide greater understanding of cognitive aspects of CFS. As well as having implications for cognitive-behavioural treatment of CFS, the findings emphasise the importance of multi-disciplinary research and treatment of CFS. Previous studies have suggested a relationship between defensive coping styles and immunological functioning. The identification of high rates of defensive coping styles amongst people with CFS, provides more evidence against the mind-body dualism which has pervaded through people's understanding. It is hoped that multi-disciplinary research and practice will continue to offer new hope for people suffering from CFS.

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APPENDIX ONE

The Green College, Oxford Definitions and Recommendations (Sharpe *et al*, 1991)

Broad clinical syndromes:

- Chronic Fatigue Syndrome (CFS)
 1. Principal symptom is fatigue with definite onset and not lifelong
 2. Fatigue is severe, disabling, and affects physical and mental functioning
 3. Fatigue for at least 6 months, during which it is present at least 50% of the time
 4. Other symptoms may be included, such as:
 - Myalgia
 - Mood disturbance
 - Sleep irregularity
 5. Definite medical and psychiatric exclusions:
 - Established medical conditions known to produce chronic fatigue
 - Schizophrenia
 - Manic depressive illness
 - Substance abuse
 - Eating disorder
 - Organic brain disease

- Postinfectious fatigue syndrome (PIFS)
 1. A subtype of CFS which follows or is associated with an infectious illness
 2. Patients must fulfill criteria for CFS as specified above, and, in addition, have
 - (a) definite evidence, including laboratory corroboration, of infectious illness at onset
 - (b) Full syndrome is present for at least 6 months after onset of infection

APPENDIX TWO

The Pragmatic Inference Test (PIT: Winters and Neale, 1985)

Vignettes presented on tape

A You decide to open your own dry cleaning shop in a small but growing town near the border. Your store will be the only one of its kind for miles around. In the first year of business, the town's population doubles and your business prospers. Your ad campaign is a big success and reactions from customers indicate that the cleaning is quality work. Your gross sales exceed expectations. You wonder whether it would be to your advantage to open a chain of stores, so you go to the bank and apply for a loan. As you hoped, the bank approves the loan.

- A1. What kind of store do you open?
 - A. Hardware
 - B. Dry cleaning
- A2. In what part of the country is the town located?
 - A. Midwest
 - B. South
- A3. Where is the loan obtained?
 - A. Loan agency
 - B. Bank
- A4. What is the reason for the success of your business?
 - A. You are a smart businessperson
 - B. You had no competition

B. You have been looking unsuccessfully for a job as a factory worker. The unemployment rate has risen lately, and jobs are especially tight in your field. Sales have been hurt because of foreign competition. You decide to talk to a friend about the situation. He reminds you that you have had difficulties with management in the past because of tardiness and a poor performance record. Your search for a job is frustrating and you go four weeks without finding a job.

- B1. Why do you discuss your situation with a friend?
 - A. You need advice
 - B. Your friend is hiring
- B2. How long do you go without finding work?
 - A. Four weeks
 - B. Four months
- B3. Why do you have trouble finding work?
 - A. Poor work record
 - B. Poor job market
- B4. What kind of job interests you?
 - A. Big company
 - B. Small company

C. You pride yourself on your appearance. You recently spent some money on new clothes and a new hairstyle. The next day you receive a number of compliments at work, especially from one colleague. However, this person angers you later on in the day by asking you for a lift home. This is a great inconvenience because this person lives quite a distance from your destination.

- C1. Why do you receive a compliment from a colleague?
 - A. Your appearance is genuinely perceived as worthy of a compliment.
 - B. This person needs a favour from you.
- C2. Why do you spend money on your appearance?
 - A. Self pride
 - B. You enjoy compliments
- C3. Who gives you the most compliments at work?
 - A. Same sexed people
 - B. Opposite sexed people
- C4. On what do you spend your money?
 - A. Shoes
 - B. Hairstyle

D. A neighbour mentions to you that their teenager is having a drinking problem. You wonder if the neighbour is going to ask you for advice. The neighbour is an independent and headstrong person who rarely seeks advice from others. You are uncomfortable because you do not have children of your own and you are not very good at counselling people. The neighbour leaves without asking for your advice.

- D1. Who comes to you for advice?
 - A. Fellow worker
 - B. Neighbour
- D2. What is the nature of the problem?
 - A. Stealing
 - B. Drinking
- D3. What gender is the person with the problem?
 - A. Male
 - B. Female
- D4. Why doesn't the neighbour ask you for advice?
 - A. The person is the type to not ask for advice
 - B. You are inexperienced in this area

E. You and a colleague decide to go out one night for a bite to eat. You wonder whether you will have a good time since your colleague is a moody person. The night starts out poorly when you forget to call a taxi for the both of you and you also fail to make dinner reservations. You and the colleague wait for an hour at the restaurant and there is still no table. You both decide to go elsewhere for a meal. The food and service is unsatisfying at the other place, especially for the colleague. On the trip home, the colleague asks you a lot of questions about how you were able to receive a recent promotion from the boss, and mentions that no one else in the office has received a promotion in over two years. The questioning indicates a hostile tone.

E1. Where do you and the colleague go?

- A. To the cinema
- B. To a restaurant

E2. At what time of day does the activity take place?

- A. Afternoon
- B. Evening

E3. Why does the colleague act hostilely to you?

- A. The person is jealous of you
- B. The person is angry that you forgot to call a taxi and make a reservation

E4. Who initiates the activity?

- A. You
- B. The colleague

F. You have a date with someone new. You go to the cinema and your date does not have much of an opinion about it. And for most of the evening, your date does not say much. You also do not initiate much conversation, and when you do talk, you have a difficult time keeping up your end of the conversation. When the evening is over, your date expresses disappointment about how the evening went.

F1. With whom do you have a date?

- A. Close friend
- B. New acquaintance

F2. Where do you go on the date?

- A. To the cinema
- B. To dinner

F3. Why does the date go badly?

- A. The date was a boring person
- B. You were not interesting enough for the person

F4. Where did you go after the date?

- A. For a drive
- B. Nowhere

G. A lonely, aged person sits next to you on a park bench while you are reading a book and begins to talk to you. You are not surprised by this, since strangers are often friendly towards you. After some small talk, you find out that this person is down on hard times and needs help. You and the person talk for quite some time, and it seems to you that the person continues to enjoy your company.

- G1. Who starts the conversation with you?
 - A. A tourist
 - B. A stranger
- G2. Why does this person talk to you for so long?
 - A. You are friendly
 - B. This person wants your help
- G3. What are you doing when you are approached by the individual?
 - A. Reading a newspaper
 - B. Reading a book
- G4. Why is this person down on hard times?
 - A. Illness
 - B. Deserted by family

H. The company you work for is always busy around holiday time. It is the day before the Christmas holiday and everyone in the office is exhausted. On short notice you decide to throw an office party. You prepare an interesting mix of gin and fruit punch, which draws a number of compliments from others. Everyone seems to enjoy themselves. You make friends with a couple of new colleagues and many people laugh at your jokes.

- H1. Why was the party a success?
 - A. Your colleagues are in the mood to unwind
 - B. You know how to throw a good party
- H2. What is popular at the party?
 - A. Liquor
 - B. Food
- H3. At what time of year is the party?
 - A. Easter
 - B. Christmas
- H4. Is the party well attended?
 - A. Yes
 - B. No

I. You give an important talk on a controversial topic to a group of town residents. You present a point of view that in the short run is unpopular but will likely benefit the town over the long term. The audience reacts negatively, especially to your suggestion that the town ought to purchase more trucks. The next speaker presents a view that is opposite to your own. As you listen to the speech, you notice that the individual is a very fluent and persuasive speaker. It becomes quite obvious to you that the second speaker receives a positive reaction from the audience.

- I1. Where do you give the speech?
 - A. Political convention
 - B. Town hall meeting
- I2. Why does the audience react negatively to your speech?
 - A. You are an ineffective speaker
 - B. The second speaker took the less controversial viewpoint
- I3. How do you learn about the audience's reaction to the second speaker?
 - A. Someone tells you
 - B. You witness it
- I4. What is being discussed at the meeting?
 - A. Road repair
 - B. Rubbish removal

J. Recently, you haven't done all the work that your boss expects of you. The boss begins to complain about your performance. The job is sometimes difficult for you because it is quite technical and the hours are a burden. Also, you recently discover through the office grapevine that the boss' nephew is interested in your position.

- J1. With whom do you talk about the problems at work?
 - A. No one
 - B. Your spouse
- J2. What kind of skill does this job require?
 - A. Manual
 - B. Technical
- J3. Why does your boss complain about your work performance?
 - A. You have poor technical skills
 - B. The boss wants you to quit to make room for a relative
- J4. What shift do you work?
 - A. Day
 - B. Night

K. You take a college course in English Literature because you like to write. One of your assignments is to write a paper on one famous contemporary English author. You choose John Fowles, a decision which is met with praise by the teacher who is a big fan of Fowles. The teacher tells you that Fowles is perhaps the most influential contemporary writer. You work hard on the paper and think that it is well written. You are pleased when the paper is returned the teacher comments that your interpretation of Fowles' work is consistent with her own and you receive an excellent grade.

- K1. What kind of course do you take?
 - A. English literature
 - B. Writing course
- K2. Why do you take the course?
 - A. Need the credits
 - B. Pleasure
- K3. Why does the teacher like your paper?
 - A. You are a good writer
 - B. Your viewpoints are similar to the teachers
- K4. Why do you choose to write about Fowles?
 - A. He is your favourite author
 - B. The teacher tells you to

L. You recently receive a salary increase at work. While you are a bit surprised by this since you had no prior notice about such a raise, you do not feel that you have been a reliable worker. Indeed, others have received wage increases in the past when you did not. The day after you receive the news, a memo is sent to all workers indicating that in the last few months a number of employees have voluntarily left the company. The company's owner offers to be sensitive to suggestions for improving job satisfaction.

- L1. What kind of income raise do you receive?
 - A. Bonus money
 - B. Wage increase
- L2. How do you hear about the raise?
 - A. Memo
 - B. Told personally
- L3. Why do you get the raise?
 - A. The company wants to prevent further resignations
 - B. You deserve the raise because of good performance
- L4. Who else gets a raise?
 - A. No one
 - B. Everyone

APPENDIX THREE
Information Sheet for Participants with CFS

CHRONIC FATIGUE SYNDROME STUDY
INFORMATION

CONFIDENTIAL

Thank you for your interest in this study. You are invited to participate in a study about the thought processes of people when they are experiencing Chronic Fatigue Syndrome. As you may know, various studies have been carried out that suggest that for many people ‘cognitive-behaviour therapy’ (CBT) can be a useful treatment for Chronic Fatigue Syndrome (CFS). ‘Cognitive-behaviour therapy’ involves working on how people think and what people do that might have an effect on their illness. With more information about this we hope to be able to make cognitive behaviour therapy for CFS more effective.

This study aims to investigate some aspects of how people with CFS think, and whether this is different from other groups of people. The study will involve filling out some written questionnaires about how you have been feeling recently, and also carrying out some simple tests of memory and concentration.

The study will take less than one hour. This will only involve one appointment, which will include a break for refreshments. Additional expenses that are incurred by you because of taking part in the study, will be reimbursed. Please let the researcher know about these when an appointment is arranged.

You do not have to take part in this study if you do not want to. If you decide to take part you may withdraw at any time without having to give a reason. Your decision whether to take part or not will not affect your care and management in any way. All information that is collected as part of this study will remain entirely confidential.

All proposals for research using human subjects are reviewed by an ethics committee before they can proceed. This proposal was reviewed by the Joint UCL/UCLH Committees on the Ethics of Human Research, The Bethlem and Maudsley NHS Trust and Institute of Psychiatry Ethical Committee (Research), St. Mary’s Local Research Ethics Committee and Barking and Havering Local Research Ethics Committee.

Please retain this information sheet. If you would like to discuss the research further, please contact:

Cathy Creswell
Clinical Psychologist in training
Sub-department of Clinical Health Psychology
Tel:
email: c.creswell@ucl.ac.uk

APPENDIX FOUR
Information Sheet for Healthy Volunteers

CHRONIC FATIGUE SYNDROME STUDY
INFORMATION

CONFIDENTIAL

Thank you for your interest in participating in this study. You are invited to participate in a study about the thought processes of people when they are experiencing Chronic Fatigue Syndrome. You have been asked to participate in this study as part of a comparison group of healthy volunteers who do not have Chronic Fatigue Syndrome.

Recently studies have been carried out that suggest that for many people psychological therapy can be a useful treatment for Chronic Fatigue Syndrome (CFS). Psychological therapies often involve working on how people think and what people do that might have an effect on their illness. With more information about this we hope to be able to make psychological therapy for CFS more effective.

This study aims to investigate some aspects of how people with CFS think, and whether this is different from other groups of people. The study will involve filling out some written questionnaires about how you have been feeling recently, and also carrying out some simple tests of memory and concentration.

The study will take less than one hour. This will only involve one appointment, which will include a break for refreshments. You will be reimbursed five pounds to cover any expenses that you have had to incur.

You do not have to take part in this study if you do not want to. If you decide to take part you may withdraw at any time without having to give a reason. All information that is collected as part of this study will remain entirely confidential.

All proposals for research using human subjects are reviewed by an ethics committee before they can proceed. This proposal was reviewed by the Joint UCL/UCLH Committees on the Ethics of Human Research (study number: 99/0055).

Please retain this information sheet. If you would like to discuss the research further, please contact:

Cathy Creswell
Clinical Psychologist in training
Sub-department of Clinical Health Psychology
Tel:
email: c.creswell@ucl.ac.uk

APPENDIX FIVE
Information Sheet for Participants with Diabetes

CHRONIC FATIGUE SYNDROME STUDY
INFORMATION SHEET

CONFIDENTIAL

Thank you for your interest in participating in this study. You are invited to participate in a study about the thought processes of people when they are experiencing Chronic Fatigue Syndrome. **You have been asked to participate in this study as part of a comparison group who also have to attend the doctors regularly, but do not have Chronic Fatigue Syndrome.**

Recently various studies have been carried out that suggest that for many people psychological therapy can be a useful treatment for 'Chronic Fatigue Syndrome' (CFS). Psychological therapies often involve working on how people think and what people do that might have an effect on their illness. With more information about this we hope to be able to make psychological therapy for CFS more effective.

This study aims to investigate some aspects of how people with CFS think, and whether this is different from other groups of people. The study will involve filling out some written questionnaires about how you have been feeling recently, and also carrying out some simple tests of memory and concentration.

The study will take less than one hour. This will only involve one appointment. You will be paid five pounds in return for participating in the study to cover any expenses that you may incur.

You do not have to take part in this study if you do not want to. If you decide to take part you may withdraw at any time without having to give a reason. Your decision whether to take part or not will not affect your care and management in any way. All information that is collected as part of this study will remain entirely confidential.

All proposals for research using human subjects are reviewed by an ethics committee before they can proceed. This proposal was reviewed by the Joint UCL/UCLH Committees on the Ethics of Human Research (study number: 99/0055), the Institute of Psychiatry Ethical Committee (Research) (067/99), St. Mary's Local Research Ethics Committee (98/BM/313) and The Whittington Hospital Local Research and Ethics Committee (99/75).

Please retain this information sheet. If you would like to discuss the research further, please contact:

Cathy Creswell

Clinical Psychologist in training

Sub-department of Clinical Health Psychology

Tel:

email: c.creswell@ucl.ac.uk

APPENDIX SIX

Consent Forms for Healthy Volunteers

Investigator: Cathy Creswell

CHRONIC FATIGUE SYNDROME STUDY
CONFIDENTIAL

Have you read the information sheet about this study? Yes/ No

Have you had an opportunity to ask questions and discuss this study? Yes/ No

Have you received satisfactory answers to all your questions? Yes/ No

Have you received enough information about this study? Yes/ No

Who have you spoken to about this study? _____

Do you understand that you are free to withdraw from this study
at any time Yes/ No

without giving a reason for withdrawing Yes/ No

Do you agree to take part in this study? Yes/ No

Name:.....

Address:.....
.....

Signature
(participant):.....

(researcher):

APPENDIX SEVEN

Consent Form for Participants with CFS or Diabetes

Investigator: Cathy Creswell

CHRONIC FATIGUE SYNDROME STUDY
CONFIDENTIAL

Have you read the information sheet about this study? Yes/ No

Have you had an opportunity to ask questions and discuss this study? Yes/ No

Have you received satisfactory answers to all your questions? Yes/ No

Have you received enough information about this study? Yes/ No

Who have you spoken to about this study? _____

Do you understand that you are free to withdraw from this study
at any time Yes/ No
without giving a reason for withdrawing Yes/ No
without affecting your medical care? Yes/ No

Do you agree to take part in this study? Yes/ No

Name:.....

Address:.....
.....

Signature
(participant):.....

(researcher):

APPENDIX EIGHT

Letter to Doctors Working with Patients with CFS

Dear Doctor

A research project is being carried out to investigate specific aspects of the cognitive behavioural model of Chronic Fatigue Syndrome (CFS). The model has successfully guided psychological interventions with people with CFS, yet certain aspects of the model remain under-investigated. It is anticipated that clarification of the key aspects of the model will lead to further refinement, and hence greater efficacy, of cognitive behavioural interventions with people with CFS.

Participants will carry out tests of attention and memory as well as filling out checklists regarding fatigue, anxiety and depression symptomatology, self-esteem and demographic variables. Participation will take no longer than one hour per participant, and any expenses will be reimbursed.

I would be grateful if you would assess whether patients fulfill the attached inclusion criteria. If a patient does fulfill the criteria could you please inform the patient that the study is currently underway, and ascertain whether he or she might be willing to be involved. If the patient is interested and would like further information, I would be grateful if you would contact me and inform me of the patient's name and a contact telephone number. Where possible I will attempt to carry out the research assessment when the participant is due to visit your department.

In accordance with ethical guidelines, participants will be free to withdraw from the study at any point and will be given debriefing information after participation. They will be given full assurance that their involvement in the study will not affect their medical care.

This research project is being carried out as a requirement for completion of a doctorate degree in Clinical Psychology at University College London. Data will be collected up to January, 2000. The study has been approved by the Institute of Psychiatry Ethical Committee (Research) (067/99).

Thank you very much for your co-operation

Yours sincerely

Cathy Creswell
Clinical Psychologist in training
Contact:
telephone
email c.creswell@ucl.ac.uk

Criteria for inclusion in the study

- (i) The participant must be between 18 and 30 years of age.
- (ii) The participant's first language is English.
- (iii) Participants should not (currently or in the past) be involved in psychological interventions for CFS, e.g. CBT. The participant should not currently be involved in psychiatric interventions for any other psychiatric disorder.
- (iv) Oxford Diagnostic Criteria for CFS will be followed:
 1. The patient has been suffering from persistent or relapsing fatigue for at least six months and less than five years which:
 - a) is of new definite onset; b) is unrelated to bed rest; c) requires bed rest after previously tolerated exertion; d) is not alleviated by rest; e) requires substantial reduction in previous levels of activity.
 2. The patient is suffering from persistent or recurrent reduction in short-term memory and/ or concentration sufficient to interfere with normal activities.
 3. The patient has at least three of the following concurrent symptoms, which must not predate CFS and have been persistent or recurrent for at least six consecutive months; recurrent sore throat, painful neck or armpit glands, muscle discomfort/ pain, joint pain but no swelling or redness, headaches, difficulty in falling asleep or maintaining sleep, malaise lasting over 24 hours following exertion, allergies (of new type or severity), nausea, imbalance.
 4. There is no active medical condition that may explain the presence of chronic fatigue.
 5. There is no previous medical condition that cannot be excluded and may explain the chronic fatigue, e.g. malignancy, Hepatitis B or C.
 6. There is no past history of the following psychiatric disorders: major depression with psychosis or melancholia, bipolar affective disorder, schizophrenia or delusional disorder, chronic dementia.
 7. There has been no alcohol or other substance misuse within five years prior to the onset of chronic fatigue or subsequently.
 8. There are no unexplained laboratory or imaging test abnormalities suggesting the presence of another condition that may explain the chronic fatigue

APPENDIX NINE

Letter to Doctors Working with Patients with Diabetes

Dear Doctor

A research project is being carried out to investigate specific aspects of the cognitive behavioural model of Chronic Fatigue Syndrome (CFS). **Patients with diabetes are being recruited as a comparison group who have a chronic illness.**

The cognitive behavioural model has successfully guided psychological interventions with people with CFS, yet certain aspects of the model remain under-investigated. It is anticipated that clarification of the key aspects of the model will lead to further refinement, and hence greater efficacy, of cognitive behavioural interventions with people with CFS.

Participants will carry out tests of attention and memory as well as filling out checklists regarding fatigue, anxiety and depression symptomatology, self-esteem and demographic variables. Participation will take no longer than one hour per participant, and five pounds will be paid to cover any expenses incurred.

I am looking for volunteers aged between 18 and 30 years, who have insulin treated diabetes and who speak English as their first language. They should not be experiencing any complications associated with their diabetes and diabetes should have been diagnosed for at least six months. Medication should have been stable for at least two months.

If a patient does fulfil these criteria could you please pass on a letter with information sheet and stamped, addressed envelope to the patient during their clinic appointment. Where possible I will attempt to carry out the research assessment when the participant is due to visit your department.

In accordance with ethical guidelines, participants will be free to withdraw from the study at any point and will be given debriefing information after participation. They will be given full assurance that their involvement in the study will not affect their medical care. This research project is being carried out as a requirement for completion of a doctorate degree in Clinical Psychology at University College London. Data will be collected up to January, 2000. The study has been approved by the Joint UCL/UCLH Committees on the Ethics of Human Research (study number: 99/0055), the Institute of Psychiatry Ethical Committee (Research) (067/99), St. Mary's Local Research Ethics Committee (98/BM/313) and The Whittington Hospital Local Research and Ethics Committee (99/75).

Thank you very much for your co-operation

Yours sincerely

Cathy Creswell

Clinical Psychologist in training

Contact: telephone

email: c.creswell@ucl.ac.uk

Criteria for inclusion in the study

- (i) The participant will have Insulin-dependent diabetes which has been diagnosed for at least six months.
- (ii) The participant will be between 18 and 30 years of age.
- (iv) The participant's first language is English.
- (v) Participants will not (currently or in the past) be involved in psychological interventions e.g. Cognitive Behaviour Therapy.
- (vi) Medication will have been stable for at least two months.

APPENDIX TEN
Letters to Potential Participants with CFS

Dear [name of patient]

Your specialist has provided me with your name in case you might be interested in participating in a research project that is being carried out in to Chronic Fatigue Syndrome.

I have enclosed an information sheet about the study and I would be very grateful if you would consider taking the time to participate.

Please return the slip below to let me know if you might be interested in taking part. A stamped addressed envelope is provided. If you feel that you might be interested, I will then contact you to discuss the study further and to arrange an appointment that is convenient for you should you agree to participate.

Thank you very much for taking the time to read this information.

Yours sincerely

Cathy Creswell
Clinical Psychologist in training
Contact:
telephone
email c.creswell@ucl.ac.uk

*Return to C. Creswell, Sub-dept Clinical Health Psychology, UCL, Gower St.,
London WC1E 6BT*

I would/ would not like to be contacted about participating in the CFS research study.

Signature: _____ Name (print): _____

Address: _____

Contact telephone number: _____

APPENDIX ELEVEN

Letters to Potential Participants with Diabetes

Dear [name of patient]

Your doctor has provided me with your name in case you might be interested in participating in a research project that is being carried out. The study is investigating Diabetes and Chronic Fatigue Syndrome.

I have enclosed an information sheet about the study and I would be very grateful if you would consider taking the time to participate.

Please return the slip below to let me know if you might be interested in taking part. A stamped addressed envelope is provided. If you feel that you might be interested, I will then contact you to discuss the study further and to arrange an appointment that is convenient for you should you agree to participate.

Thank you very much for taking the time to read this information.

Yours sincerely

Cathy Creswell
Clinical Psychologist in training
Contact:
telephone
email c.creswell@ucl.ac.uk

*Return to C. Creswell, Sub-dept Clinical Health Psychology, UCL, Gower St.,
London WC1E 6BT*

I would/ would not like to be contacted about participating in the CFS research study.

Signature: _____ Name (print): _____
Address: _____

Contact telephone number: _____

APPENDIX TWELVE

Letters of Approval from Local Research Ethics Committees

1. Barking and Havering Local Research Ethics Committee
2. The Whittington Hospital Local Research Ethics Committee
3. St Mary's Local Research Ethics Committee
4. The Institute of Psychiatry Ethical Committee (Research)
5. The Joint UCL/UCLH Committees on the Ethics of Human Research

**Barking and Havering
Health Authority**

Professor L. Findley
Consultant Neurologist & Clinical Director
Department of Neurology
Harold Wood Hospital
Gubbins Lane
HAROLD WOOD
ESSEX RM3 OBE

3rd March 2000

Dear Professor Findley

Re: CFS Research study

Thank you for your letter of the 17th January 2000 enclosing amended patient letter relating to this study. I am pleased to confirm that this was approved by the Barking & Havering Local Research Ethics Committee on the 1st March 2000 and you may proceed with the trial.

The Committee has agreed that any amendments to studies should be presented on the Amendment Form and I enclose copy for your information.

The Committee looks forward to receiving a final report of your research findings in due course.

Yours sincerely

Mrs. J. Carter
Administrator LREC



Telephone: 0171-288 5675 (24 hr service)
0171-288 5676
0171-288 5677
Fax Number: 0171-288 5674

 The
Whittington
Hospital

Highgate Hill
London N19 5NF

12th November 1999

Dear Ms Creswell

99/75 Self-esteem, Attribution and Coping in Chronic Fatigue Syndrome

I refer to your submission of the above project to the Local Research and Ethics Committee. I am pleased to inform you that your study has been approved by Chairman's Action under reciprocal agreement with UCL/UCLH joint Local Research Ethics Committee. However, I should be grateful if you would complete the enclosed research registration form in order that we are able to monitor research activity within the Trust.

Approval is for two years from the date of this letter. Extension of this period will be dependent on the submission of a brief synopsis of the project together with an estimation of the time required for its ultimate completion.

The Ethics Committee will require an annual report on the progress of the study, and a copy of the completed study together with any consequent publication. In addition, the Committee must be informed, by the completion of the relevant form, of any untoward or adverse events that occur during the course of the study. The person who provided independent review of the original protocol should also be sent information regarding adverse events.

The Ethics Committee must be informed of, and approve, any proposed amendment to your initial application that has a bearing on the treatment or investigation of patients or volunteers.

A copy of the patient consent form and information sheet must be lodged in the clinical notes.

An
NHS
Trust


UCL
MEDICAL
SCHOOL

Cathy Creswell
Page 2
99/75
12th November 1999

I am sure that every effort is already made to preserve the confidentiality of any patient information used in this study. Please ensure that the team of investigators are aware that everyone who has access to patient information appreciates the importance of maintaining confidentiality particularly in respect of the use of computers and the statutory regulations laid down in the Data Protection Act 1984

In terms of the managerial and financial implications associated with the study, where these relate to additional costs for the Trust, Mr Rob Hurd (Management Accountant, Finance Department, Whittington Hospital), will be in contact with you to discuss the Trust's schedule of charges for research projects. **Approval of these issues must be obtained from your general manager before the study commences.**

In any correspondence regarding the study please quote the above Ethics Committee reference number.

Yours sincerely

Mr John Farrell
Chairman - Local Research and Ethics Committee

Ec161
17.3.99



RESEARCH AND DEVELOPMENT OFFICE

(Mailbox 121) 2nd Floor, Mint Wing, St Mary's Hospital, Praed Street, London W2 1NY
Direct Line R&D: 0171-886 1330/2014 Ethics: 0171-886 6514 Fax: 0171-886 1529

ST MARYS' LOCAL RESEARCH ETHICS COMMITTEE

March 29, 1999

Ms C Creswell

Dear Ms Creswell

98/BM/313 Self esteem, attribution and coping in Chronic Fatigue Syndrome
R & D NUMBER MUST ALWAYS BE USED IN CONNECTION WITH THIS RESEARCH

On behalf of the members I am pleased to say that the above project documents, detailed in the following grid, have now been given ethical approval by the St Mary's LREC. This approval is given on the understanding that the research team will observe strict confidentiality over the medical and personal records of the participants. It is suggested that this be achieved by avoidance of the subject's name or initials in the communication data. In the case of hospital patients, which can be done by using the hospital record number and in general practice, the National Insurance number or a code agreed with the relevant GP.

Document	Original date	Amended date
LREC application form (date circulated)	17.3.99	-
Patient information sheet	17.3.99	-
GP Letter	17.3.99	-
Questionnaire	17.3.99	-

Chairman's initials

The St Mary's NHS Trust comprises:
St Mary's Hospital, the Western Eye Hospital, and acute services at St Charles Hospital



**Institute of
Psychiatry**

at The Maudsley

**Ethical Committee
(Research)**

**Research Ethics Co-ordinator
Margaret M Chambers**

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KING'S
College
LONDON
Founded 1829

ETHICAL COMMITTEE (RESEARCH)

Tel: (0171 919) 2892

22 April, 1999

Dr T Chalder
Dept of Psychological Medicine
Institute of Psychiatry

Dear Dr Chalder

**Re: Self-esteem, attribution and coping in Chronic Fatigue Syndrome
(067/99)**

The Ethical Committee (Research) considered and approved the above study at its meeting on 16 April 1999. **This approval is dependent on the completion of the enclosed Student Form and the typographical error (2nd line) of the letter to doctors. Please return these documents to this office.**

Initial approval is given for one year. This will be extended automatically only on completion of annual progress reports on the study when requested by the EC(R). Please note that as Principal Investigator you are responsible for ensuring these reports are sent to us.

Please note that projects which have not commenced within two years of original approval must be re-submitted to the EC(R).

Please let me know if you would like to nominate a specific contact person for future correspondence about this study.

Any serious adverse events which occur in connection with this study should be reported to the Committee using the attached form.

Please quote Study No. 067/99 in all future correspondence.

Yours sincerely,

Margaret M Chambers
Research Ethics Coordinator

University College London Hospitals is an NHS Trust incorporating The Eastman Dental Hospital, The Hospital for Tropical Diseases, The Middlesex Hospital, The National Hospital for Neurology & Neurosurgery, The United Elizabeth Garrett Anderson Hospital and Hospital for Women, Soho, and University College Hospital.