# The Development of a Work Instability Scale for Multiple Sclerosis

By

**Estelle Mc Fadden**

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I hereby confirm that the work submitted is my own and to the best of my knowledge appropriate credit has been given where reference has been made to the work of others. I have not submitted this material, either in full or in part, for a degree at this or any other institution.

## Abstract

Multiple Sclerosis (MS) is a chronic inflammatory demyelinating disease of the central nervous system leading to progressive impairment of various neurological functions (Kesslering et al, 2002). It is the commonest cause of chronic neurological disability among young adults in the western world (Mc Donnell et al, 2001). The physical and psychological effects of multiple sclerosis can impact heavily on any of these important life activities and so people with multiple sclerosis are more likely to be unemployed than the general population (Solari et al, 2001).

Premature unemployment can have significant financial and social consequences in adulthood. Like the majority of the general population, many of those who have multiple sclerosis will still have dependants and regular financial commitments in their forties and fifties, such as a mortgage. Therefore, unemployment may be disastrous for the family, especially if it is the main wage earner who loses their job.

### Background

As well as the financial gain from work, it can also be an enjoyable activity that can define people and help them fulfil a social role. In recent years there has been more awareness of the burden of disease, not just physically but emotionally and financially and the disease and demographic factors contributing to job loss in the multiple sclerosis population have been identified. The effect of vocational rehabilitation in chronic diseases has also been explored to a degree and the Organisation for Economic Co – operation and Development (OECD) report in 2003 stated that early vocational rehabilitation intervention is the most effective measure against the dependence on benefits that is a consequence of long-term ill health (Frank, 2003). However at present there is no outcome measure to assess the need for intervention and it still remains to be seen if costly interventions, such as, the disease modifying therapies or vocational rehabilitation have any effect on the working life of those with multiple sclerosis.

Work disability is premature work cessation due to a health problem or disability. Prior to work disability there may be a period of work instability. Work instability is a state in which the consequences of a mismatch between an individual's functional abilities and the demands of his or her job can threaten continuing employment if not resolved. During this time the patient is most at risk of job loss and timely intervention in the work place can facilitate job retention (Gilworth et al, 2003).

The aim of this thesis was to produce a valid and reliable Work Instability Scale (WIS) for multiple sclerosis in order to be able to predict those at risk of job loss.

## Methods

A multiple methodological approach, involving both qualitative and quantitative research techniques was used and a disease specific, work instability scale, the MS-WIS was developed and tested for appropriate psychometric properties. In depth, semi-structured interviews were conducted with people with MS in order to explore the issues of working while having MS. Qualitative analysis of these interviews using a grounded theory approach yielded themes that described the issues of working with MS. Further qualitative analysis of the interviews using content analysis methodology was being done alongside this which identified the items suitable for inclusion in the scale, a process known as item generation. The scale was then subjected to in-depth psychometric testing using Rasch analysis. Rasch analysis was chosen as this would ensure that a single construct was being measured and allowed the validity and the reliability of the scale to be tested.

## Results

Twenty seven people were interviewed for the qualitative study, fifteen men and twelve women and the interviews transcribed and studied. Four main themes emerged as factors contributing to work instability on qualitative analysis of the data:

i) The impact upon work of the physical and cognitive aspects of the disease

ii) The extent to which the environment and organisational aspects of working life can affect job ability and retention

iii) The social aspects of the working environment

iv) The psychological aspects of working.

Item generation initially yielded a scale consisting of one hundred and twenty two items.A postal questionnaire yielded 109 responses, the data from which then had to undergo in-depth Rasch analysis, during which items were discarded. Vocational assessment was used as a gold standard. This process resulted in a valid, reliable twenty one item scale - the Multiple Sclerosis Work Instability Scale (MS –

WIS). The final scale is quick and easy to complete. It is scored in 3 bands indicating low, medium and high risk of Work Disability. The medium – risk threshold has 88% sensitivity and 60% specificity and the sensitivity and specificity of the scale reaches 100% at the high – risk threshold with regards to the need for work place intervention.

## Conclusion

MS has been shown to have a considerable impact on the working life of the individual. A disease specific outcome measure to quantify the risk of job loss in an individual with MS with good psychometric properties has been developed. The MS-WIS has been derived from a strong conceptual framework and a strong mathematical measurement model. It has been rigorously tested for its psychometric properties and has high sensitivity and specificity for risk of job loss.

# Acknowledgements

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Dr Majella Mc Fadden of Sheffield Hallam University offered expert advice regarding qualitative methodology and was also involved in the analysis of the transcripts. As ever my family and husband Andrew fully supported and applauded my endeavours.

The final and most important thanks go to all the people who participated in this study. I truly appreciated the time they gave up to be interviewed and assessed and hope that they find some value in this piece of work.

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# Chapter 1: Introduction

## 1.1 Hypothesis

It is possible to identify people who are at risk of losing employment due to multiple sclerosis by developing a valid and reliable work instability scale (WIS) for Multiple Sclerosis.

## 1.2 Aim of the thesis

The aim of this thesis was to devise an outcome measure that could be used to assess the risk of job loss in those with multiple sclerosis, that is, a work instability scale (WIS) for multiple sclerosis (MS). The study design comprises a number of discrete stages:

**Stage one:** Qualitative interviews

**Objective:** To generate the item pool for the patient completed Multiple Sclerosis Work Instability Scale (MS – WIS)

**Stage two:** Item selection

**Objective:** To produce a draft questionnaire

**Stage three:** Cognitive debriefing

**Objective:** To assess the face and content validity of the draft WIS

**Stage four:** Internal Construct Validity

**Objective:** To test the structural properties of the scale

Item reduction

**Stage five:** Criterion Validity

**Objective:** Comparison of the scale against a "Gold Standard" assessment to assess the reliability and sensitivity of the draft measurement. The pre-existing method of determining work instability is to carry out a full vocational assessment.

**Stage six:** Reliability

**Objectives:** To assess the test- reliability and internal consistency of the scale

To further confirm the validity of the questionnaire

## 1.3 Objectives of the thesis

To achieve the aim of devising a work instability scale for multiple sclerosis, the following objectives needed to be met:

- A critical review of the literature relating to the causes and consequences of multiple sclerosis as defined by the ICF model, paying particular attention to participation and job loss and the role of vocational rehabilitation
- Conduct semi – structured interviews with people with multiple sclerosis who were in employment or had left their jobs within the previous six months to determine the issues of working with multiple sclerosis and to identify potential items for a patient completed scale

- Subject the scale to Rasch analysis which would test a range of psychometric properties, including unidimensionality and its consistency over time

## 1.4 Outline of the thesis

The thesis is comprised of 7 chapters:

*Chapter1* introduces the area to be studied, discusses the background to this and provides the broad aims and objectives of the thesis

*Chapter 2* reviews the existing literature relating to the thesis topic, employment in multiple sclerosis. It also outlines the socioeconomic consequences of multiple sclerosis and vocational rehabilitation interventions.

*Chapter 3* comprises the qualitative research methodology used for the purposes of the project. It explains the theoretical approach to qualitative research and the actual methodology used.

*Chapter4* comprises the quantitative research methodology employed for this project. It explains the theoretical approach behind quantitative research and the actual methodologies used.

*Chapter5* presents the qualitative results obtained. It comprises the qualitative analysis of the data from the interviews and the items chosen for the scale.

*Chapter6* presents the quantitative results obtained. It gives the final 21- item Work Instability Scale after the initial items have been subjected to rigorous statistical analysis in the form of Rasch analysis.

*Chapter7* is the discussion chapter for the entire project, covering the limitations of the study and the conclusions drawn from the various chapters. The areas where the scale would be used best are discussed along with future directions for further research in this area.

# Chapter 2: Definition, Aetiology, Pathology and Consequences of Multiple Sclerosis; A Literature Review

## 2.1 Definition of Multiple Sclerosis

Multiple Sclerosis (MS) is a chronic inflammatory demyelinating disease of the central nervous system leading to progressive impairment of various neurological functions (Kesslering et al, 2002). A key defining feature of MS is that lesions are disseminated in both space and time, i.e. they occur at more than one site and develop on more than one occasion (O Connor, 2002). It is the commonest cause of chronic neurological disability in young adults in the western world (Mc Donnell et al, 2001).

## 2.2 Aetiology of multiple sclerosis

The mean age of onset is about 30 years old. Approximately 70% of cases occur between the ages of 20 and 40 years. Onset before the age of 15 years or after the age of 50 is unusual. It has been reported that 10% of cases occur in childhood and 20% arise when the patient is aged greater than 40 (Vukusic et al, 2001).The disease has a female predominance of approximately 2:1 (Ebers et al, 1997). A recent United Kingdom national study estimated the incidence of MS at 5.5 per 100,000 with age standardized rates for females of 7.2 and 3.1 per 100,000 for males (Alonso et al, 2007).

The exact cause of MS is unknown however some environmental and genetic factors have been established which have been shown to influence the epidemiology of the disease.

### 2.2.1 Environmental factors

The prevalence of MS varies widely around the world and Kurtzke classified different areas as, high, medium or low risk. Areas that are classified as high risk have a prevalence of > 30/100,000, medium risk areas 5 - 30/100,000 and low risk <5/100,000. High risk areas include northern and central Europe (except northern Scandinavia), Italy, the northern United States, Canada, south – eastern Australia, parts of the former Soviet Union and New Zealand. Medium risk areas include southern Europe (excluding Italy), the southern United States, northern Australia, northern Scandinavia, other parts of the former Soviet Union, South Africa (white population only) and possibly Central America. Low risk areas are, the rest of Asia and Africa, the Caribbean and possibly northern South America (Kurtzke, 1991).

The geographic variation in the prevalence of MS appears to be due to both environmental and genetic factors (Ebers et al, 1997). The evidence for an environmental influence includes the increased risk of developing MS with increasing latitude even in countries that are relatively racially homogeneous (Hammond et al, 1988, Kuroiwa et al, 1983). There have been several reports of immigrants to a country developing a prevalence rate similar to that of the indigenous population (Gale, 1995). Conversely, several epidemiological studies have shown that persons who migrate from a high risk to a low risk zone carry with them at least part of the risk of their country of origin, even though the disease may not become apparent for a number of years. The risk of developing the disease depends upon the age of migration i.e. it remains high if the individual left the high risk area for a lower risk area after their mid-teens and supports the role of environmental factors in the aetiology of MS (Compston et al, 1999).

There is a hypothesis that MS develops as a result of infection by a virus or another pathogen. Many antibodies to infectious agents have been detected at elevated levels in the serum or CSF of patients with MS, for example, Chlamydia pneumoniae, the Epstein Barr virus and the measles, mumps and influenza viruses among others (Cook et al, 1995). Certain viruses have caused demyelination in experimental animal models and some human myelopathies similar to MS are caused by viral infection or the resultant immune activity. However the role of infection in MS remains unclear as the evidence is not conclusive (O Connor, 2002).

### 2.2.2 Genetic factors

There is unequivocal evidence that there is a familial association in MS. The absolute risk of the disease in a first-degree relative of a patient with MS is less than 5% however this is 20-40 times the risk in the general population (Sadonovick et al, 1988). The concordance rate of 31% among monozygotic twins is approximately six times the rate among dizygotic twins which is 5 % (Sadonovick et al, 1993). However, within families with more than one affected member, no consistent pattern of Mendelian inheritance has emerged (Compston at al, 1999).

Certain histocompatabilty antigens (HLAs) are more frequent in patients with multiple sclerosis than in control subjects. It has been known since 1973 that the presence of the HLA-DR 2 allele increases the risk of MS (Jersild et al, 1973). Compston's analysis of the subject, which was done over a 25 year period, concluded that the gene that confers susceptibility to multiple sclerosis is DR2 (DR15), although its biological contribution to susceptibility is relatively small (Compston et al, 1999).

## 2.3 Pathogenesis

The epidemiological data points to a relationship between MS and an environmental factor that is encountered in childhood and, after years of latency; either evokes the disease or contributes to its causation. If the initial event in the development of MS is a viral or other infection of the central nervous system, then a secondary factor must operate later in life to activate the neurological disease and cause exacerbations. It is thought this secondary mechanism is an autoimmune reaction, attacking some components of myelin and causing axonal damage and destruction (Compston et al, 1999). The pathogenesis of MS is thought to be an organ or antigen-specific autoimmune reaction and although MS has long been thought of as a demyelinating condition, it is more likely that the permanent neurological disability that is accrued is due to axonal damage and that this damage begins early in the course of the disease (O Connor, 2002)

## 2.4 Diagnosis

No single clinical feature or diagnostic test is sufficient for the diagnosis of multiple sclerosis. The diagnosis is based on clinical criteria but investigations, such as the analysis of cerebrospinal fluid and magnetic resonance imaging (MRI) are used to support the diagnosis (McDonald et al, 2001). A diagnosis of relapsing remitting MS is given when neurological dysfunction becomes "disseminated in space and time" (Noseworthy et al, 2000).

From 1982 until July 2000, "The Poser Criteria" for the diagnosis of MS was in use. During this time degrees of diagnostic certainty were identified by categories ranging from clinically definite diagnosis to laboratory-supported definite MS, clinically probable MS, and laboratory supported probable MS.

In July 2000 the International Panel on the Diagnosis of MS was convened in London, United Kingdom, under the auspices of the U.S. National Multiple Sclerosis Society and the International Federation of MS Societies to reassess existing diagnostic criteria and to recommend appropriate changes. The aim was to create diagnostic criteria that could be used by the physician, but could also be adapted for clinical trials, to include magnetic resonance imaging (MRI) as a diagnostic tool, to provide a scheme for the diagnosis of primary progressive disease and to simplify the diagnostic classification of MS. The new criteria have been widely accepted and are known as "The Mc Donald Criteria" (Mc Donald et al, 2001).

**Table 2.1 Diagnostic Criteria for Multiple Sclerosis: The McDonald Criteria**

| **Clinical Presentation** | **Additional Data Needed for MS Diagnosis** |
|---|---|
| | |
| Two or more attacks; objective clinical evidence of 2 or more lesions | None(i) |
| Two or more attacks; objective clinical evidence of 1 lesion | Dissemination in space, demonstrated by MRI *or* Two or more MRI-detected lesions consistent with MS plus positive CSF *or* Await further clinical attack implicating a different site |
| One attack; objective clinical evidence of 2 or more lesions | Dissemination in time, demonstrated by MRI *or* Second clinical attack |
| One attack; objective clinical evidence of 1 lesion (monosymptomatic presentation; clinically isolated syndrome) | Dissemination in space, demonstrated by MRI *or* Two or more MRI-detected lesions consistent with MS plus positive CSF *and* Dissemination in time, demonstrated by MRI *or* Second clinical by MRI *or* Second clinical attack |
| Insidious neurological progression suggestive of MS | Positive CSF *and* Dissemination in space, demonstrated by1) Nine or more T2 lesions in brain *or* 2) 2 or more lesions in spinal cord, *or* 3) 4-8 brain plus 1 spinal cord lesion *or* abnormal VEP associated with 4-8 brain lesions, or with fewer than 4 brain lesions plus 1 spinal cord lesion demonstrated by MRI *and* Dissemination in time, demonstrated by MRI *or* Continued progression for 1 year |

If criteria indicated are fulfilled, the diagnosis is multiple sclerosis (MS); if the criteria are not completely met, the diagnosis is "possible MS"; if the criteria are fully explored and not met, the diagnosis is "not MS."

(i) No additional tests are required; however, if tests [magnetic resonance imaging (MRI), cerebral spinal fluid (CSF)] are undertaken and are *negative,* extreme caution should be taken before making a diagnosis of MS. Alternative diagnoses must be considered. There must be no better explanation for the clinical picture.

### 2.4.1 Clinical course of the Disease

**Table 2.2 Course of the Disease**

| Disease Type | Relapses | Disease Progression |
|---|---|---|
| 1. Relapsing – remitting multiple sclerosis (RRMS) | Clearly defined acute attacks followed by acute or partial recovery to the pre – existing level of disability | No disease progression between attacks |
| 2. Primary progressive multiple sclerosis (PPMS) | None | From onset, with or without occasional plateaus or temporary minor improvements |
| 3. Secondary progressive multiple sclerosis (SPMS) | Initial relapsing – remitting phase | Characterised by disease progression with or without occasional relapses, minor remissions and plateaus |
| Progressive relapsing multiple sclerosis (PRMS) | Clearly defined acute attacks followed by acute or partial recovery to the pre – existing level of disability | Disease progression from onset punctuated by relapses |

(Adapted from Kremenchutsky et al, 1999)

Relapsing – remitting multiple sclerosis (RRMS) accounts for approximately 85% of the cases of MS at onset (Miller, 2001) and there is a 2.5:1 ratio of female to male for this type of MS (Alonso A et al, 2007). In this disease type, symptoms and signs evolve over a few days, reach a nadir, stabilise, and then often improve spontaneously or after treatment with steroids in a number of weeks. The ability of steroids to shorten the length of relapses often diminishes with time (Noseworthy et al, 2000). Secondary progressive MS occurs after the initial relapsing – remitting phase. (Noseworthy et al, 2000).

Patients with MS whose disease course is progressive from onset and who have no periods of relapse or remission are relatively rare (Thompson et al, 1997). According to Ebers 1997 work, 20% of cases of MS are primary progressive with a gradual accumulation of disability (Ebers et al, 1997). However Thompson recognised that the precise proportion varied between studies from 7.7 – 38% and concluded that should be an average figure of 10%should be taken as the proportion of people with MS who have PPMS. The difference in these series is probably influenced by the diagnostic criteria set, the strictness to which they are adhered and the effect of retrospective verses prospective data collection, in that an initial relapse may well be forgotten over time (Thompson et al, 1997). The incidence of primary progressive MS is similar in men and women (Ebers et al, 1997, Alonso et al, 2007).

The presenting symptoms seen in RRMS are usually sensory disturbances, unilateral optic neuritis, diplopia (internuclear opthalmoplegia), L'Hermitte's sign (trunk and limb parathesias provoked by neck flexion), limb weakness, clumsiness, gait ataxia and bladder and bowel disturbance. Fatigue is a frequent symptom often complained of and can worsen as the day goes on. Uhthoff's phenomenon is the transient worsening of symptoms associated with an increase in body temperature and is strongly suggestive of MS, as are symptoms in the post partum period.

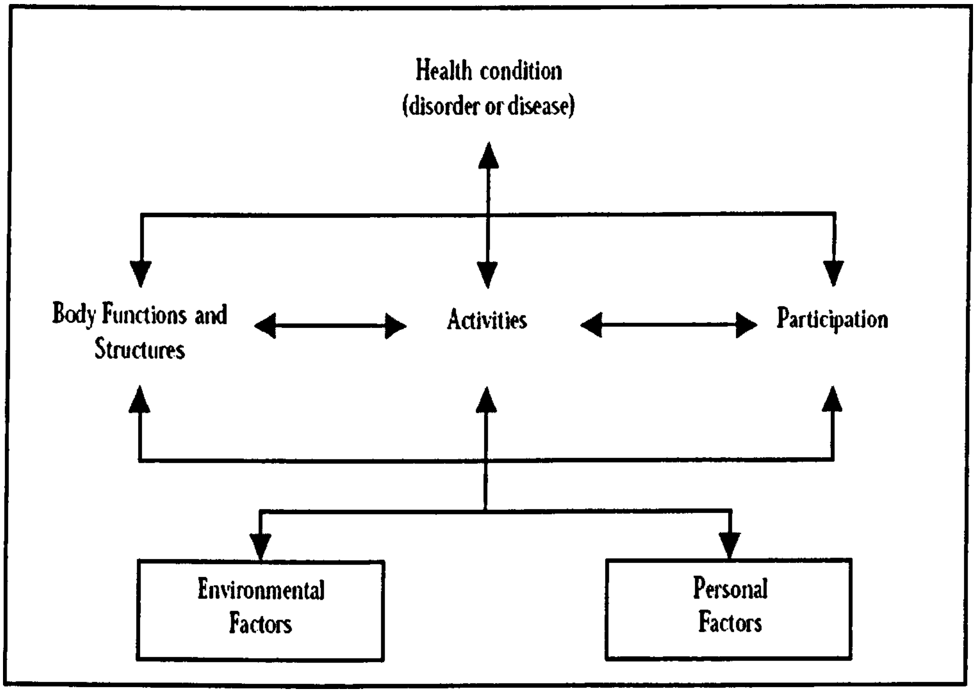
Cognitive impairment, depression, emotional lability, dysarthria, dysphagia, vertigo, progressive quadraparesis and sensory loss, ataxic tremors, pain, sexual dysfunction, spasticity and other manifestations of central nervous system dysfunction may also dominate the clinical picture (Noseworthy et al, 2000).

## 2. 5 Consequences of multiple sclerosis

The World Health Organisation's International Classification of Functioning (WHO ICF, 2001) (Figure 2.1) is a biopsychosocial framework for describing the situation of someone with long-term ill health (Wade et al, 2003). This was previously known as the International Classification of Impairments, Disability and Handicaps (ICIDH, WHO 1980).

The nomenclature has changed, thus impairment is described as problems associated with body function or structure, disability has become limitation in activity, that is, difficulties an individual may have in performing activities, and handicap has become restriction in participation, which is, problems an individual may experience in involvement in life situations, namely, at home, socially or when at work (Yorkston et al, 2003).

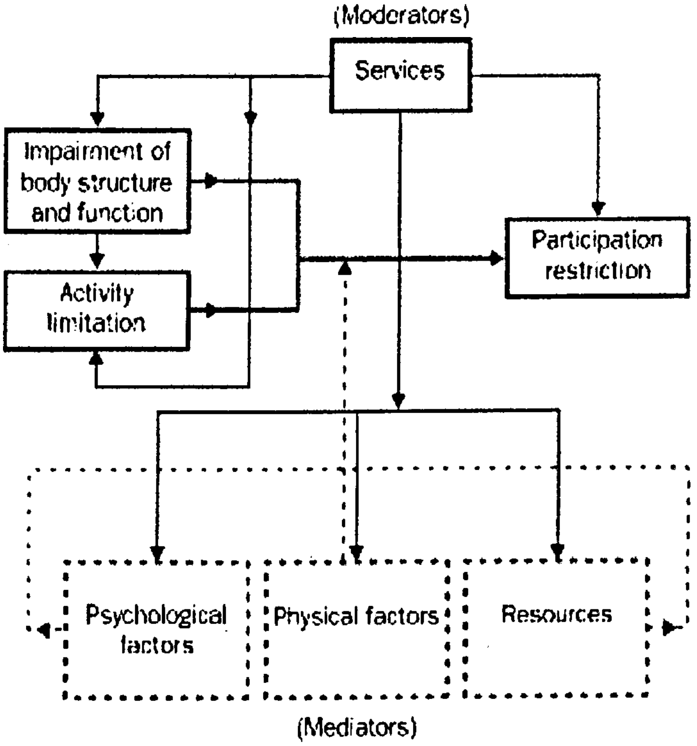
**Figure 2.1 The WHO ICF (Adapted from WHO guidelines, 2001)**



MS is responsible for problems in body function and structure, such as limb weakness, spasticity, impaired vision, double vision, sensory disorders, fatigue, bladder and bowel incontinence and cognitive dysfunction (Compston et al, 1999). The limitation of activities and level of participation depends on the individual, on society, on their attitude towards the illness and their health and social care provision.

These effects are known as moderators and mediators (Figure 2.2) and are widely used in the field of social medicine (Baron et al, 1986).

**Figure 2.2 Conceptual Model of Moderators and Mediators**



(Bent et al, 2002)

Moderators are health-care and social services that have a direct effect on the way a person deals with his or her impairment, limitations in activity and participation restriction. Drugs such as beta-interferons and glatiramer acetate act as moderators through their effect in modifying the long term course of the disease by decreasing relapse rate (Kesslering et al, 2002). Mediators include resources and psychosocial factors e.g. self esteem - that act as filters and determine the degree to which a person's problems with bodily function influence their limitation in activity (Bent et al, 2002).

Free will is another example of a mediator. In most everyday situations (including many aspects of their illness) patients experience a sense of control and influence over their behaviour by choosing between different courses of action. Free will, therefore, should join other phenomena such as motivation at the level of the person. The notion of free will adds an important and often neglected dimension to the ICF, and may explain illness behaviour not produced by disease, injury, psychopathology or psychosocial factors (Wade et al, 2003).

One of the strengths of the ICF is that it highlights the influence of conceptual factors when determining limitation on activities and restrictions on participation (Figure 2.2) and this confirms the observation that how someone behaves is influenced by many factors (Wade et al, 2003). In addition to these aspects of function and disability, environmental factors must also be considered, particularly when the patient is at work (Yorkston et al, 2003). The ICF emphasises the importance of social participation for humans and that goal-directed behaviour is the core construct of all level of activities (World Health Organisation International Classification of Functioning, 2001).

**Table 2.3 An overview of the ICF**

| | **Part 1: Functioning and Disability** | |
|---|---|---|
| **Components** | **Body Functions and Structures** | **Activities and Participation** |
| **Domains** | Body functions<br>Body structures | Life areas (tasks, actions) |
| **Constructs** | Change in body functions (physiological)<br>Change in body structures (anatomical) | Capacity: Executing tasks in a standard environment<br>Performance: Executing tasks in the current environment |
| **Positive aspect** | Functional and structural integrity | Activities Participation |
| | Functioning | |
| **Negative aspect** | Impairment | Activity limitation<br>Participation restriction |
| | Disability | |

**Adapted from WHO guidelines, 2005**

*Activity* is the execution of a task or action by an individual. *Activity limitations* are difficulties an individual may have in executing activities. *Participation* is involvement in a life situation. *Participation restrictions* are problems an individual may experience in involvement in life situations. In the 2001 World Health Organisation International Classification of Functioning introduction the domains for the Activities and Participation component are given in single list that covers the full range of life areas (from basic learning or watching to composite areas, such as interpersonal interactions or employment). The domains covered are learning and applying knowledge, general tasks and demands, communication, mobility, self – care, domestic life, interpersonal interactions and relationships, major life areas and community, social and civic life. There is an obvious overlap between Activities and Participation but the domains can be qualified by introducing the concepts of *performance* and *capacity*.

*Performance* describes what an individual does in his or her current environment. This includes a social role and so performance can also be discussed as "involvement in a life situation" or "the lived experience". *Performance* also includes all aspects of the environment from the physical environment and the social life of the individual to the attitudes of those surrounding the person. *Capacity*, however, describes the person's ability to carry out a task or action. Capacity aims to define the highest level of functioning that an individual may reach in a given domain at any single moment at a given time in an environment. Ability depends very much upon the impact of the environment on the individual. Therefore capacity reflects the environmentally adjusted ability of the individual.

The gap between *capacity* and *performance* illustrates the difference between the impact of the current environment and a more uniform environment and provides a very useful guide as to what can be done to the environment of the individual to improve performance.

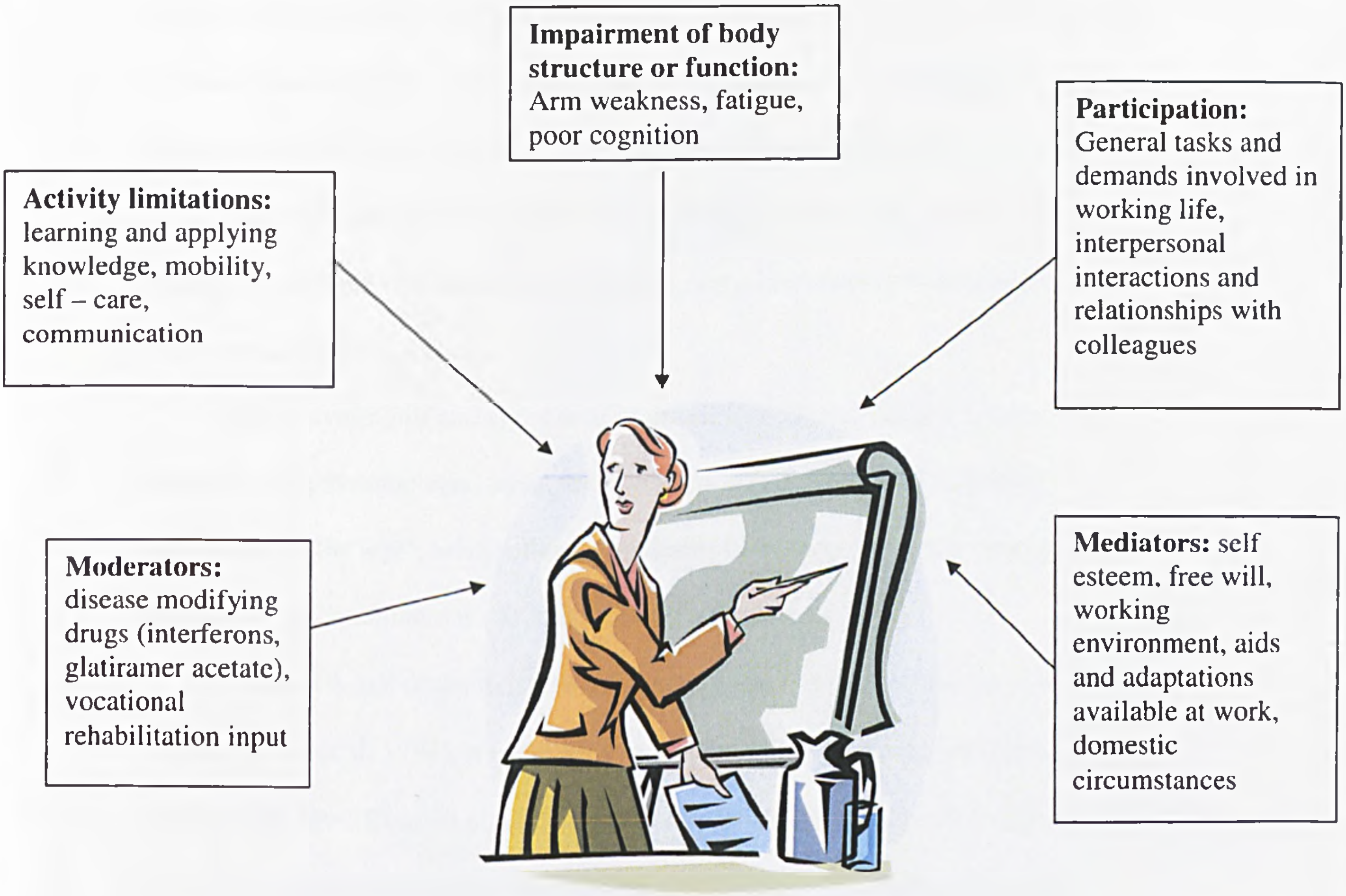
This illustrates that it can be difficult to distinguish between *Activities* and *Participation* when discussing the domains of everyday life. The ICF has offered guidance to users to differentiate the two in a manner that best suits their research purposes. The four ways they have suggested are:

1. To designate some domains as activities and others as participation, not allowing any overlap
2. Same as above, but allowing partial overlap
3. To designate all detailed domains as activities and the broad category headings as participation
4. To use all domains as both activities and participation (World Health Organisation guidelines, 2001).

The third approach has been chosen for the purposes of this research project. Therefore *Activities* (previously known as disability under the old classification) is comprised of learning and applying knowledge, communication, mobility and self – care. *Participation* (previously known as handicap) encompasses general tasks and demands, domestic life, interpersonal interactions and relationships, community, social and civic life and other major life areas, such as, employment or work.

The ICF model is helpful in attempting to understand the complex phenomenon of work within the context of MS (Yorkston et al, 2003). Problems in body function, for example leg weakness causing poor mobility, could lead to some limitations in activity such as walking around the workplace. This means the individual is unable to carry out their normal work activities and may have to give up work, rendering them unemployed and experiencing participation restriction. Adaptation of the workplace or the use of a walking aid acts as a mediator and may keep the person in employment for longer (Figure 2.3).

**Figure 2.3 Conceptual model of a person with MS at work**



## 2.6 Problems associated with body function or structure

Problems associated with body function or structure was previously known as impairments when discussed in the ICIDH. The symptoms and signs of MS are very variable but in general reflect the involvement of those parts of the CNS that are most heavily demyelinated and so affect the *architecture* or *structure* of the brain (Compston et al, 1999). Multiple sclerosis is characterised clinically by episodes of a localised disorder of the optic nerves, spinal cord, and brain. Commonly there are relapses and remissions, followed by progression at variable rate and extent (Sadovnik et al, 1993, Weinshenker BG, 1994). Paty and Ebers described the list of clinical findings seen in patients attending MS clinics at the Universities of British Columbia and Western Ontario, Canada.

Initial symptoms and signs seen in the clinic included fatigue, optic neuritis, internuclear opthalmoplegia, nystagmus, vertigo, gait disturbances, sensory loss (commonly in the legs), brisk reflexes, weakness in the legs, spasticity and bladder disturbance or combinations of these symptoms (Paty et al, 1997).

Fatigue is one of the most common symptoms of MS, affecting at least 75% of patients (Freal et al, 1994), it is reported in all clinical phenotypes of MS and affects people of all ages (Comi et al, 2001). Fatigue in MS is experienced as a subjective lack of physical and/or mental energy, perceived by the individual or caregiver to interfere with usual and desired activities (Johansson et al, 2008). It can be caused by the disease process (primary fatigue), by problems such as insomnia, infections or depression (secondary fatigue) or a combination of these and so can be multifactorial in nature. However it can be difficult to measure as it is a subjective experience and this is why self-reported scales to measure this are currently in use, for example, the

Fatigue Severity Scale (Krupp et al, 1989: appendix 1). Such tools help clinicians and researchers measure fatigue (Johnson SL, 2008). Mills et al provided a medical definition of fatigue in multiple sclerosis, in terms of a common framework, typical of a medical history in their 2008 work, which entailed a qualitative phase followed by a cross-sectional questionnaire study. Forty patients with clinically definite MS were involved in the research and a definition of fatigue in MS was produced. In summary, fatigue could be defined as reversible motor and cognitive impairment, with reduced motivation and a desire to rest. It could appear spontaneously or may be brought on by mental or physical activity, humidity, acute infection and food ingestion. It was relieved by daytime sleep or rest without sleep. It could occur anytime but was usually worse in the afternoon (Mills et al, 2008).

Fatigue can be one of the worst and most debilitating of the clinical features of the disease and is a major reason for work absenteeism and a major cause of unemployment in people with MS (Ziemssen T, 2009). Dyck's survey in 2000 focusing on women with MS and employment issues indicated that 34% reported frequent problems at work related to fatigue (Dyck et al, 2000). In 2008 Diamond et al looked at relationships between information processing, depression, fatigue and cognition in MS. It was found that slower processing, lower verbal fluency, fewer recall of words and digits and poor recall of visual-spatial information was correlated with higher levels of fatigue. Depression and physical fatigue had the greatest influence on the association between processing speed and more effortful tasks, such as, immediate recall and word list learning (Diamond et al, 2008).

It was demonstrated by Holtzer et al in their 2009 work that fatigue and executive control are uniquely related in MS by providing the first behavioural evidence of this. Twenty individuals with MS were compared against twenty controls case matched for age, sex, education and intellect (using IQ measurements) by looking at executive control processes using a Delayed Item Recognition (DIR) paradigm and assessed levels of fatigue using the Fatigue Severity Scale (Krupp et al, 1989; appendix 1). The linear regression model which was used revealed that increased fatigue levels were related to delayed DIR reaction time and accuracy performance only in the MS group (Holtzer et al, 2009).

However the relationship between fatigue in MS and employment can also be a very positive one. A longitudinal study of variations in and predictors of fatigue in MS was carried out by Johansson et al (Johansson et al, 2008).

This study investigated the predictive value of the variables of sex, sense of coherence, mood, living with a partner and work status on variations in fatigue levels in 219 individuals attending and out patient clinic over a two year period. Sense of coherence (SOC) gives an indication of an individual's capacity to cope with stressful life events (Antonovsky, 1993). Fatigue levels were again measured using the Fatigue Severity Scale (Krupp et al, 1989; appendix 1) at six monthly intervals. Independent predictors of increased fatigue were depressive symptoms, a weak or moderate sense of coherence, living with a partner and not working. Independent predictors of decreased fatigue were absence of depressive symptoms, a strong sense of coherence, living alone and working.

Cognitive impairment and depression also occur in multiple sclerosis and are not always late features of the disease. Research into this area has shown that impairment of memory and the ability to learn can be affected early in the course of the disease. In their 1987 research, Truelle et al showed that 55% of patients with disease duration of less than 5 years had marked memory impairment. In the same year, Van den Burg et al, assessed 40 patients with mild physical disability and found

7 (17.5%) to be cognitively impaired. In another study 84 patients who were in remission were examined, they were not on any medication and had an expanded disability status scale (EDSS) of less than 6 (Kurtzke, 1983; appendix 2). It was found that 26 (30%) were intellectually impaired when compared with controls (Good et al, 1992).

The lifetime prevalence of major depression in MS is approximately 50% (Minden et al, 1990) and may be more common in MS than in other chronic neurological conditions (Schubert et al, 1993). Risk factors for major depression in MS were identified in Pattens study in 2000 and included the female gender, age less than 35 years, family history of major depression and a high level of stress. Chwastiak et al identified younger age, less education and lack of social support as associations with depressive symptoms in MS (Chwastiak et al, 2002) however these factors are also recognized as associations with depression in the general population and in other medical illnesses (Eaton et al, 1989). In 2002 Chwastiak et al carried out an epidemiological study of depressive symptoms in a large community sample of 739 Canadians with MS. They examined the prevalence of clinically significant depressive symptoms in a large community sample of patients with multiple sclerosis using the Center for Epidemiological Study Scale for Depression (CES – D Scale) (Radloff, 1977). This instrument has been validated and widely used in medical settings to screen for depression; however for the purposes of the study Chwastiak et al performed an additional analysis to ensure only symptoms due to depression were being measured. This analysis removed items from the scale that could be due to either depression or multiple sclerosis. Four items from the scale were deleted: "I had trouble keeping my mind on what I was doing" "I felt that everything I did was an effort" "My sleep was restless" and "I could not get going" as it was felt these symptoms were not exclusive to depression and would be heavily influenced by multiple sclerosis. The results from the revised scale still revealed that 41.8% of this population had clinically significant depressive symptoms, higher than seen in either the general population or in primary care populations (Chwastiak et al, 2002). The rates of depression were also higher than those seen in persons with other chronic medical illnesses, such as, HIV (Lyketsos et al, 1993). In Chwastiaks study the severity of MS in terms of disability was more strongly associated with depressive symptoms than duration of illness or pattern of progression. The lower prevalence of severe depressive symptoms in patients with longer durations of MS indicates an adaptation to illness as time passes whereas subjects who were within a year of the diagnosis were more often severely depressed (Chwastiak et al, 2002). Similar results were found when investigating quality of life (QoL) which showed improved levels of QoL with longer disease duration and it was concluded that a measure of subjective QoL may reflect adjustment to disease (Ford et al, 2001).

Structural brain abnormalities in MS are thought to be linked with major depression. In 2004 Feinstein et al studied two groups of patients with clinically definite multiple sclerosis. One group had defined major depression, as defined by the Diagnostic and Statistical manual of Mental Disorders (4th Edition) and the other did not. The depressed and non depressed groups were matched for disease and cognitive measures and for demographic characteristics. The essential findings from Feinstein's 2004 work were, patients who were depressed had a greater T2 – weighted lesion volume seen on MRI. However, the authors did recognise that while both lesion burden and atrophy are important in the pathogenesis of depression in multiple sclerosis, psychosocial influences also play a considerable role (Feinstein et al, 2004). Another study found that people who have MS which mainly affects the spinal cord and cerebellum have a lower rate of depression than do those with predominantly cerebral involvement (Schiffer et al, 1983).

Therefore the explanation for the high rate of depressive symptoms seen in MS is likely to be multifactorial and include direct effects of MS on limbic system structures and the negative effects of functional impairment on life satisfaction and self – esteem (Chwastiak et al, 2002).

## 2.7 Limitation in activity

Limitation in activity describes the difficulties an individual may have in performing activities and was previously known as disability under the terms of the ICIDH (Yorkston et al, 2003). *Activity* comprises domains such as learning and applying knowledge, general tasks and demands, communication, mobility and self – care. Difficulties or problems in these domains can arise when there is a qualitative or quantitative alteration in the way in which an individual carries out these domain functions. These difficulties are known as *limitations* or *restrictions* and are assessed against a generally accepted population standard. The standard or norm against which an individuals capacity and performance is compared is that of an individual without a similar health condition (disease, disorder or injury, etc) and the *limitation* is the difference between the observed and the expected performance. The expected performance is that of people without a specific health condition (World Health Organisation guidelines, 2001). For example, an individual with multiple sclerosis may have to use a stick to walk or have memory problems and so would have limitations in activity regarding mobility and cognition which can easily lead to problems within many aspects of daily life, including employment. People with multiple sclerosis experience a range of problems with body function or structure and can also have marked limitation in activities. Ford et al found this to be the case for retaining work, as a mixture of problems with body function (impairment), limitation in activity (disability) and progressive disease course were predictive of not working (participation) (Ford et al, 2001).

### 2.7.1 Work in multiple sclerosis

Work can be defined in a number of ways; as paid employment at a job or a trade, occupation or profession (Collins Dictionary, 2003) or as an accomplishment, that is the product of overcoming obstacles to achieve an objective (Yorkston et al, 2003). The capacity to work is often altered in persons with chronic illness such as multiple sclerosis (Gulick et al, 1989).

### 2.7.2 Physical reasons for job losses

A significant number of patients with MS lose their jobs because of the limitation in activity associated with the disease (Hakim et al, 2000). There is a recognised correlation between more severe physical disability (EDSS>6.5) and an increased risk of premature unemployment due to the physical symptoms encountered (Solari et al, 2001). In one study of 305 patients with MS, none of the patients with severe MS (EDSS score of 7 or more) were in paid employment. The employment rate for those with moderately severe disease was 27% compared with 68% whose had less severe disease based on an EDSS score of 4.5 or less (Hakim et al, 2000).

Dyck's survey in 2000 focusing on 534 women with MS and employment issues indicated that 44% of the 252 women who were employed at the time of the study were limited in the type and amount of work they were able to do because of

MS symptoms. 34% reported frequent problems at work related to fatigue. Frequent difficulty with standing for a period of time and difficulty climbing stairs was reported by 16% of all employed women. Frequent problems with job duties involving walking, writing and memory or concentration were identified by 15%, 12% and 11% respectively. Difficulty with symptoms of MS was cited by 77% of the 163 women who were no longer employed as the main reason for job loss. Insufficient energy to combine work, in paid employment and at home were reported by 60% as the primary reasons for leaving employment (Dyck et al, 2000).

Impairments and activity limitations identified as having the greatest effect on employment were fatigue, physical problems, such as muscle weakness, walking difficulties and lack of hand coordination. Other reported problems included visual disturbance, poor bladder and bowel control, memory loss and emotional lability (Kornblith et al, 1986, Jackson et al, 1991).

### 2.7.3 Cognitive reasons for job losses

Various cognitive deficits occur in MS. The rate at which one learns new information, skills, and procedures can be slower (Franklin et al, 1989). Both short and long term memory and abstract reasoning abilities have been reported as being diminished (Rumrill, 1996). In Roessler and Rumrill's 1995 Work Experience survey 44% of participants had difficulties with thought processing, 40% with short term memory and 26% with long term memory. Less common cognitive problems also occurred, with 12% of participants reporting difficulty with problem solving, 10% with organisation and 6% with planning (Roessler et al, 1995).

40-70% of MS patients have significant cognitive impairment and a number of studies suggest that this can also contribute to the loss of employment in people with

MS (Rao et al, 1991, Beatty et al, 1995, Edgley et al, 1991). Patients with impaired cognition were less likely to be gainfully employed or have leisure activities and they reported more physical and psychiatric symptoms than those with a similar degree of disability and duration of illness but with intact cognitive function (Hakim et al, 2000).

Rao et al found that 48 MS patients judged to be cognitively impaired by neuropsychological testing were less likely to be employed than 52 cognitively intact patients. The two groups did not differ significantly in age, education, estimated premorbid IQ, highest attained occupational status, overall physical disability, gender distribution, or disease duration or course. It was found that the group deemed cognitively intact was more likely to be working, be independent in their ADLs and have a better social life than the cognitively impaired group (Rao et al, 1991). Wild et al compared patients whose lesions were classified as predominantly cerebral or predominantly spinal based on neurological examination and magnetic resonance imaging. Although the patients with the spinal lesions were substantially more physically disabled than the patients in the cerebral group, they were more likely to be employed (Wild et al, 1991). Beatty et al studied the demographic, clinical and cognitive characteristics of multiple sclerosis patients who continue to work. To determine the factors that contribute to maintaining employment in MS patients they compared 38 patients who were still working to 64 patients who had retired prematurely. The employed group was younger, better educated, had less severe physical disability, a shorter duration of disease, an earlier age at diagnosis, and performed significantly better on nearly all neuropsychological variables examined (Beatty et al, 1995).

## 2.8 Participation

Restriction in participation describes the problems an individual may experience in involvement in life situations, namely at home, socially or when at work and replaces the term handicap used in the ICIDH (Yorkston et al, 2003). *Participation* encompasses general tasks and demands, domestic life, interpersonal interactions and relationships, community, social and civic life and other major life areas, such as, employment or work, leading to unemployment. The majority of people develop multiple sclerosis in their third, forth of fifth decades but this is usually the stage when people are at their most productive in terms of their work, home and social lives and having multiple sclerosis can affect all of the above.

Studies have shown that 50-80% of MS patients are unemployed within 10 years of disease onset (Bourdette et al, 1993, Gronning et al, 1990, LaRocca et al, 1985, Kornblith et al, 1986). Another study showed 73% of MS patients were unemployed in an average of 17 years after the first symptom (Kornblith et al, 1986).

### 2.8.1 Work Disability

Work disability is premature work cessation due to a health problem or disability. Prior to work disability there may be a period of work instability. Work instability is a state in which the consequences of a mismatch between an individual's functional abilities and the demands of his or her job can threaten continuing employment if not resolved. During this time the patient is most at risk of job loss and timely intervention in the work place can facilitate job retention (Gilworth et al, 2003).

### 2.8.2 Demographics of work in MS

Research has also been done in the area of work loss in musculoskeletal disorders and neurological disorders other than multiple sclerosis. It has been reported that approximately one third of patients with rheumatoid arthritis will not be working three years following the diagnosis (Barrett et al, 1992). The fact that in one study 30% of those with disabling knee and hip disease left their original job mainly or partly because of their joint disorder is of concern as there is a rising prevalence of serious joint disease in Britain (Palmer et al, 2005). Employment profiles in 60 people with progressive neuromuscular diseases were studied by Fowler et al in 1997. They found that 40% were in paid employment, 50% had been employed in the past and 10% had never worked. The important factors that determined employment status were level of education, type of occupation, intellectual capacity and psychosocial adjustment to their impairment (Fowler et al, 1997).

Various factors are related to employment of individuals with MS and can be divided into disease and demographic characteristics (Kornblith et al, 1986). It seems the connection between level of impairment and level of participation is not clear cut, and is influenced by many factors (Yorkston et al, 2003). Greater degree of disability and longer disease duration are associated with unemployment (Kornblith et al, 1986). Those with chronic progression from onset had the lowest employment rates, those with relapses and remissions, the highest and those with relapses and progression were in-between (Bauer et al, 1965). Gronning et al carried out a multivariate analysis of factors associated with unemployment in people with multiple sclerosis and found similar results. Diagnostic category, clinical course, occupation group and age were the strongest predictors of time to unemployment. The study concluded that patients with a non-remittent clinical course, aged 30 years and over and doing heavy physical work should be considered at high risk for early unemployment due to MS (Gronning et al, 1990).

Busche et al found the demographic variables often associated with unemployment to be older age and lower level of education. They also demonstrated the importance of job specific and social factors, for example, manual labour, private sector employment and lack of accessible transportation to get to and from work were associated with unemployment (Busche et al, 2003). In fact in one study 12.1% of unemployed MS patients reported transportation difficulties as one of the reasons for leaving their last job (Kornblith et al, 1986). Solari et al found that males were more likely to be employed than females; the female sex increasing the risk of premature unemployment. 46% of the 163 women studied were unemployed compared with an unemployment rate of 33% in the 88 men involved in the same study. Both sexes had similar levels of disease severity (Solari et al, 2001). In one study, being male increased the probability of being employed by 11% (LaRocca et al, 1992).

A 1991 study of 210 registrants of the Vancouver Island Multiple Sclerosis Society showed that in relation to employment status over two-thirds (66.7%) reported a change in their job status since the onset of MS, and almost half (44.8%) reported having to make a forced change in their field of employment. The greatest decrease in employment occurred in health related professions and service industries. Proportionally less people working in clerical positions left their jobs as illustrated in the study. There were fourteen people whose job was in medicine or an associated field when diagnosed with MS but at the time of the study only two of these were in still in health care (14.2%) and both were working on a part time basis. Thirteen of the forty one persons (32%) whose most recent job was clerical were still working in their chosen field, five on a full time basis and eight part timers (Jackson et al, 1991). In a

1986 study, 8% of participants stated that MS had caused them to change the type of work they did and over 40% indicated that due to MS they had to leave or were dismissed from a job (Kornblith et al, 1986).

In summary, the disease characteristics, which have been shown to be significant for continued employment, are degree of physical impairment, age at diagnosis, duration and course of the disease.

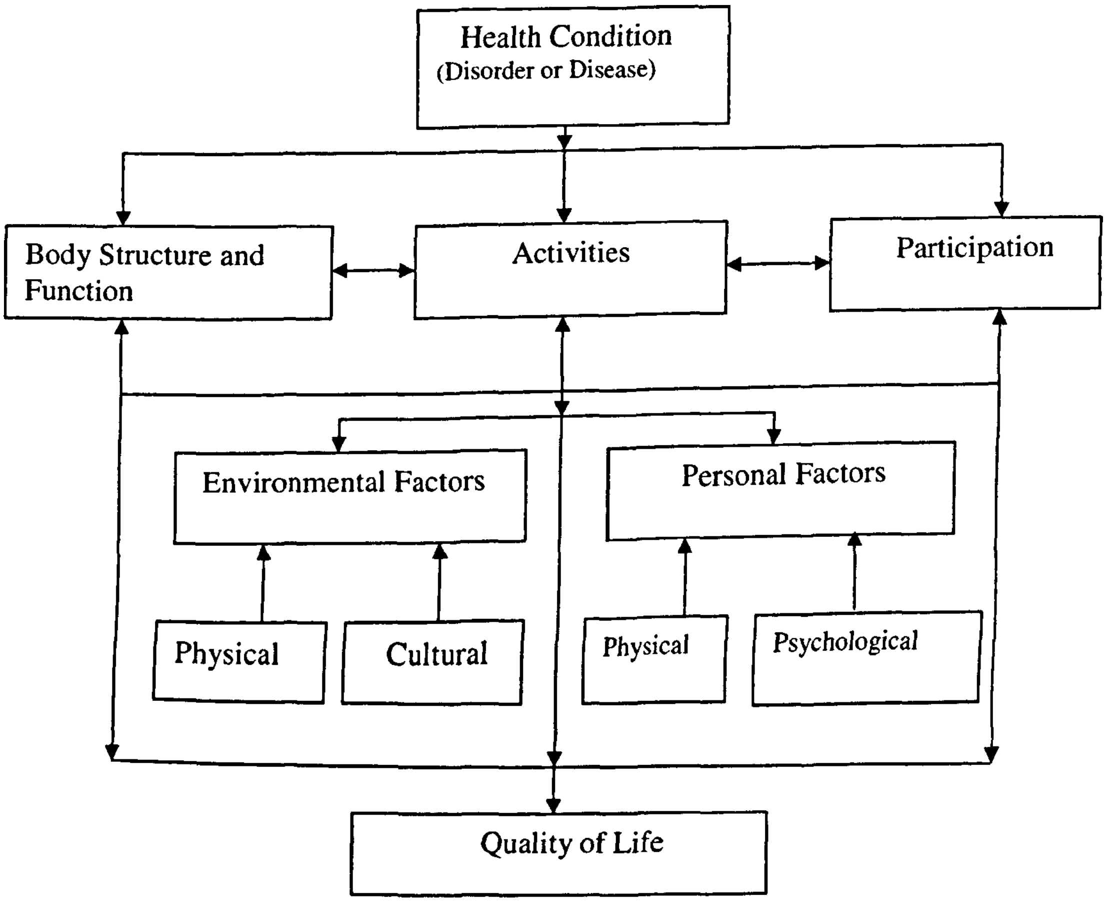
### 2.8.3 Working Environment

Job demands and the occupational environment have an important effect on the ability of patients with MS to remain in gainful employment. Yorkston found that the environmental factors of physical surroundings, social atmosphere and attitudes of colleagues and employers influenced the study participant's ability to work (Yorkston et al, 2003). Verdier – Taillefer and colleagues carried out a case – control study. Cases were patients unemployed for less than five years before the study (n = 77) and controls were currently in employment (n = 94). All study participants were being followed up in neurology clinics, had clinically definite multiple sclerosis and a score between 3 – 7 on the Expanded Disability Status Scale. Odds ratios for the relationship under review were adjusted for sex, age, disease form and educational level. The results showed that protective factors against unemployment were work in the public sector, a desk job, a sitting position at work and the knowledge that some specific arrangements at work could be obtained. Some patients were able to keep working as they could make provisions for a progressive disability, for example, loss of mobility leading to wheelchair use could be accommodated within the workplace Risk factors for unemployment were daily work duration of over 8 hours, a job requiring physical strength or manual precision, a rigid work schedule and frequent moves. Logistic regressions were performed on job characteristics significantly related to unemployment and showed that the factors having independent associations were that work in the public sector was protective whereas a rigid work schedule and work requiring physical strength were risk factors. The reason that the independent protective factor was a job in the public sector was that those jobs were mostly sedentary (Verdier-Taillefer et al, 1995).

### 2.8.4 Quality of life in multiple sclerosis

Quality of life (QoL) is a measure of "well-being". It has been described by Chibnall & Tait as *satisfaction with one's key psychosocial life areas,* by Chubon et al as *a general sense of physical and psychological well-being* and by Ware as *the extent to which one is able to function in his or her role, has emotional and social support, and is free from emotional distress* (Chibnall et al, 1990, Chubon et al, 1995, Ware, 1984). Subjective well being or QoL is not incorporated into the International Classification of Functioning (ICF) model but, can be viewed as a separate construct as outlined below in the biopsychosocial model (Figure 2.4) which explains how the ICF framework and QoL may fit together in the context of this research (Tennant A, Personal Communication 2009). In this model work instability is at the interface of activity limitation and participation, as work and maintaining a job are clearly major life areas.

**Figure 2.4: The Biopsychosocial Model incorporating QoF into the ICF (Tennant A, Personal Communication, 2009)**



Prior to 2001, the existing measures of quality of life in MS e.g. the MSQoL-54 and the Functional Assessment of Multiple Sclerosis (FAMS) reflected impairment, disability and handicap and so, were measuring health status rather than QoL. The need for a disease specific QoL measure for MS was recognized by Ford et al as they wanted to capture the concept of well-being. They developed a patient-completed disease-specific measure of quality of life in MS and validated it in a community- based population of people with MS. They recognized that QoL should be measured through ascertaining the subjective experiences of the individual (Pearlman et al, 1988). The items in the scale were generated from the discussions in focus group sessions of people with MS and then the scale was tested and re-tested on random samples from a population register of people with MS in Leeds. The Leeds MSQoL scale was found to be an appropriate, reliable and valid tool to assess quality of life in MS, measuring a construct separate from health status and acting as an adjunct to other outcome measures used in MS (Ford et al, 2001).

Once it had been established that QoL and health status were two separate entities, Ford et al demonstrated that there is not a straightforward exchange between health status and quality of life. They found that older individuals with a longer disease duration, and greater disability were more likely to report a good quality of life than a younger less disabled person with a shorter disease duration. From this, it could be concluded that subjective self perceived QoL incorporated the individual's adjustment to their disease (Ford et al, 2001).

Measurement of quality of life (QoL) could be an important measure in clinical trials when assessing the benefits of a new therapy, as it may be used to supplement impairment scales, such as the EDSS. The EDSS measures impairment and disability in terms of the physical deficits produced by MS and the effect on mobility, whereas a QoL scale may reflect the impact of such an illness on the patient's life (Murphy et al, 1998).

Rothwell felt that there were two main arguments against relying solely on measurements of impairment and disability. The first is that general well being, social function and psychological function are more important to patients in terms of overall health status than impaired physical function (Rothwell et al, 1997). Side effects are reported in trials but often do not influence the primary outcome measure and, therefore the second argument is that measuring QoL affords the opportunity to do so, by seeing if treatment benefits outweigh side effects, not just in terms of neurological impairment but in the other important domains *(Rothwell et al, 1998).*

### 2.8.5 Work and quality of life in multiple sclerosis

In 2001 Koch, Rumrill et al hypothesised that employment, educational attainment, marital status and household income were positively related to QoL and number of symptoms, symptom persistence and age were negatively related to QoL. A study was undertaken to confirm or refute the hypotheses. 227 study participants completed a 60-item questionnaire, which had three principal sections. The first focused on demographics and the illness-related experiences of the participants. The second dealt with employment and focused on training, education, job acquisition, employers attitudes, access to reasonable accommodation, wages and benefits. In the third section the study participants completed a Quality of Life scale, rating their satisfaction in seven areas of life. The scale was developed by Chibnall and Tate for use in behavioural medicine with people who have chronic health conditions. The seven areas for exploration were social life, family life, recreation, educational and intellectual development, activities of daily living, romantic experiences and expectations and hopes for the future. A multiple correlation design tested the relationship between the hypotheses suggested and perceived QoL and found that four of the seven factors suggested were related to QoL. Total number of symptoms and age were, indeed, negatively associated with QoL and educational level and employment status had positive associations. Therefore it was concluded that a lower subjective QoL was associated with a greater number of physical symptoms, a lower education level and forced lifestyle changes, such as, loss of employment (Koch et al, 2001).

Yorkston carried out a qualitative study of fourteen patients with MS and found that many of the participants did not want to give up work as they saw it as a valued activity. Hence, they did not appreciate well-intended advice such as, "take it easy" and "avoid work that is too stressful" but preferred to devise strategies that enabled them to continue in their employment. These strategies were identified as, firstly setting priorities as they were aware that both their time and energy were limited. Secondly they altered the way they carried tasks out, in order to accommodate the symptoms of MS or the problems within the working environment and also described being vigilant in anticipating potential problem (Yorkston et al, 2003).

### 2.8.6 Socioeconomic consequences of multiple sclerosis

During the first 10 years after diagnosis 50-80% of those with MS are out of work and it is only now that the socio-economic consequences of this are being addressed. MS is a costly disease due to its early onset, progressive nature and long survival time, the direct and indirect yearly costs amounting to 90,000 Euros per patient per year. (Kesslering et al, 2002) The combination of the long disease duration (mean 40 years) and the introduction of costly new disease modifying therapies have ensured that MS is one of the most expensive neurological diseases. Multiple sclerosis is more expensive than stroke or Alzheimer's disease, even though only a small proportion of the disease population will be eligible for disease modifying therapies (Battaglia et al, 2000).

Disease costs are absorbed both by the person with MS or their family and by society. They are classified into **tangible** and **intangible costs** by health economists. **Tangible** costs can be converted easily into financial terms and then further classified into **direct** and **indirect** terms. **Direct costs** are those attributable to medical care, such as, drug costs, medical visits, diagnostic tests, assistive devices, rehabilitation, home or nursing care or modifications to home or car. Historically, direct costs were largely comprised of hospitalisations (mainly long stay) and drug costs were much less, however this is no longer the case due to the introduction of costly disease modifying therapies.

**Indirect costs** are 70-80% of total disease costs and are basically loss of income or any additional costs to the employer, due to reduction or loss of work activity. They comprise the loss of earnings of those with MS, any lost earnings of the care-giver, lost taxation and any state benefits given. Informal assistance is thought to be second only to earning loss in terms of tangible expenses but often is not actually included in estimates when costing the disease.

**Intangible costs** are more difficult to quantify and are due to loss of a non-paid position, for example a student or a full-time parent, or are to do with health-related quality of life. A method known as "willingness to pay", that is, how much one would pay not to have the disease, has been proposed as a way of quantifying the intangible costs of a pathological condition, however there are no existing figures for MS at present (Battaglia et al, 2000).

## 2.9 Vocational Rehabilitation

Sickness and disability can result in loss of work which can result in exclusion from ordinary society (Frank et al, 2003). The workforce in Britain is decreasing in comparison to the numbers of old, ill or disabled people who no longer work and therefore need financial support from elsewhere. Nearly 2.7 million people in the UK are receiving incapacity benefits (Frank et al, 2003). There are approximately 3000 new claimants of incapacity benefit every week in Britain at a cost of £10 billion pounds a year (Disler et al, 2001).

It is clear from the studies discussed that being employed improves QoL in people with MS and maintains financial security; unfortunately high rates of unemployment are prevalent in this group (Bourdette et al, 1993). Early vocational rehabilitation intervention is the most effective measure against the dependence on benefits that is a consequence of long-term ill health (Frank, 2003).

Clear parallels can be drawn between employment in MS and employment in other chronic autoimmune conditions which can have a relapsing - remitting disease course, such as Rheumatoid arthritis (RA) but also with chronic diseases where there has been an initial insult such as Traumatic Brain Injury (TBI) (De Buck et al, 2002, Ponsford et al, 1995).

Rheumatic diseases are a major cause of work disability and place a significant economic burden on both the individual and society however it must be remembered that there are also significant intangible costs associated with this disease process too (Cooper, 2000, Young et al, 2000). Between 20-40% of individuals with RA will have stopped outside employment completely with the first 3 years of diagnosis and work disability is also of concern in patients with other rheumatic diseases, such as ankylosing spondylitis and systemic lupus erythematosus (De Buck et al, 2002).

Like those relating to MS, risk factors for work loss in patients with RA are both job and disease related and in fact are very similar across both diseases. Fatigue is also a common symptom in RA with up to 80% of sufferers attending hospital appointments reporting significant levels of fatigue (Belza BL, 1995, Wolfe et al, 1996). Like in MS, studies have shown that several factors may influence fatigue in RA including pain and depression, how people perceive their illness and poor social support (Pollard et al, 2006).

Job related risk factors leading to work loss in RA were how physically demanding the job was, job autonomy and control of the pace of work activities (Allaire et al, 1996). The main disease related risk factor was the severity of the disease but the demographic characteristics of increasing age and lower education level were also related to increased risk of work loss (Fex E, 1998).

In terms of employment in TBI, a multidimensional assessment was carried out on 131 male head-injury patients who suffered a range of severities of insult with specific emphasis on vocational outcome. It was estimated that those who returned to or remained in work after a mild brain injury was 42% with 30% of these having had to introduce work modifications (Stambrook et al, 1990). Condition related risk factors for work disability in TBI are known to be the severity of the injury, cognitive deficits, personality changes (including socially inappropriate behaviour), lack of awareness of injury and physical impairment. The main demographic risk factor was poor educational status (Dikmen et al, 1994). The search strategy for this section is described in Appendix 3.

### 2.9.1 Definition of vocational rehabilitation

The term "vocational rehabilitation" describes the strategies used to facilitate working for those who are willing and potentially able (Frank et al, 2003). Vocational rehabilitation (VR) has been shown to expedite return to meaningful employment, minimize workdays lost, reduce premature retirement and increases the productivity of injured workers (Disler et al, 2001).

### 2.9.2 Vocational rehabilitation interventions

Vocational rehabilitation interventions should focus on training and educational programs that broaden the person's knowledge and skills making work more accessible to them. Results point to the need for intensive and frequent monitoring of on-the-job-barriers and accommodation needs, along with long term follow up on the job, to address these barriers before they result in decreased productivity, job dissatisfaction and voluntary or involuntary termination of employment (Koch et al, 2001). It has been reported that it is more difficult for people who have previously lost work to return to work than for newly disabled people to be retained in their customary work (Lehmann et al, 1993).

The aim of vocational rehabilitation is to facilitate working for those who are willing and potentially able and can be described as top-down, which relates to government policy or bottom-up which employs more "person centered" methods (Frank et al, 2003). The Organization for Economic Co-operation and Development (OECD) reviewed top down approaches in various different countries. In Austria, Denmark, Spain, Sweden and Switzerland a request for benefits equals a request for vocational rehabilitation, this is enforced to a lesser extent in Germany, Norway and Poland. In contrast, in Australia, Canada, France, Italy, Korea, Mexico, Portugal, the

UK and the USA, vocational rehabilitation is at the request of the individual and may be difficult to access due to poor service provision. However all these countries have the same goal, which is to enable people with disabilities to carry out meaningful employment and fully participate in society whilst ensuring those who are deemed unfit for work still have an income (OECD, 2003).

In 2003 the Organisation for Economic Co-operation and Development issued a report on policies to promote work for disabled people which stressed the importance of early intervention (OECD, 2003). Initially patients perceive themselves as "sick" but able to return to work when "better". This phase probably lasts for 2-4 months, and is thought to be the period in which the psychological and multi-disciplinary process of vocational rehabilitation has the greatest impact, beyond that, patients believe themselves to be "disabled" and therefore unable to work (Frank et al, 2003). "Bottom up" approaches rely heavily on rehabilitation teams to work at grass roots level with employers, the local job centre and often the voluntary sector to ensure that the "sick individual" is back at work within the appropriate time frame (Frank et al, 2003).

The management of work disability has not been viewed as an important part of medical practice outside the specialties of rehabilitation and occupational medicine (Frank et al, 2003). Government top-down policies are as important as the bottom-up efforts by health professionals, neither approach will work independently of the other and both strategies must be enforced as vocational rehabilitation is of great importance, both economically and in terms of life satisfaction (Frank et al, 2003).

Marnetoft and Selander's findings in their 2002 work supported the hypotheses that early, rather than later vocational rehabilitation was most effective for younger women (i.e. < 47 years old) compared with other groups of different ages and sexes. The participants were males or females registered at social security offices as being on long-term sick leave (90 days or more) and a wide variety of illnesses were included in the study. Two outcome measures were used, firstly individuals not receiving any income support 48 months after rehabilitation. The second was that the level of benefit was lower at 48 months after rehabilitation compared with the level received when they started rehabilitation. The finding that vocational rehabilitation is most effective for younger women is pivotal, as the majority of incidence cases of MS occur in that very age group and other work has shown that the female sex is a risk factor for job loss in MS. Therefore this group could be targeted for vocational rehabilitation with a good chance of success (Marnetof et al, 2002).

In his 1991 work Rao stated that cognitive impairment was an important factor in determining the work status of MS patients and so it was imperative that early and accurate assessments of cognitive function would be carried out. The results of these neuropsychological tests could be made available to the employer, allowing appropriate workplace adaptations and possibly job alterations to be implemented which would enable the patient to stay in employment for longer. Once the problem had been identified these patients should also be candidates for retraining programs that could help enhance their existing level of cognitive function (Rao et al, 1991).

Rumrill et al studied the link between workplace barriers and employed people with multiple sclerosis in the United States of America. 59 adults with MS who self – initiated a telephone call to an employment hotline for assistance in managing their MS at work agreed to take part in interviews regarding their job. The study participants were people with MS in full or part – time employment and the findings revealed them to

1. Encounter few barriers related to the accessibility of their worksites

2. Experience moderate difficulties in performing the essential functions of their jobs
3. Have few problems regarding job mastery, that is, perceived satisfactoriness
4. Be generally satisfied with their jobs

Those who experienced a greater number of barriers reported lower levels of job satisfaction than those who perceived fewer barriers. The study findings provide a strong rationale for early intervention to reduce or remove job – related barriers before they undermine job satisfaction and, eventually, threaten job retention for employees with MS. Vocational rehabilitation professionals should be able to help people with MS remove or reduce workplace barriers so that they can continue to have productive, satisfying relationships to their work. That help should highlight the importance of early intervention strategies, such as, accommodation planning, self – advocacy training, job analysis and employer – employee negotiation as a means of resolving disability – related barriers before they threaten job satisfaction, job competency and job retention (Rumrill et al, 2004). Employees with MS who are satisfied with their jobs are less likely to leave the workforce, either voluntarily or involuntarily and vocational rehabilitation professionals should be aware of this and make concerted efforts to help clients maintain high levels of job satisfaction (Roessler et al, 1995).

The individual must be receptive to the idea of vocational rehabilitation or else all efforts will fail. Fraser et al looked at the profile of people with MS seeking vocational rehabilitation services in the US. In their 2003 work Fraser et al assessed a group of 79 people with MS who were concerned about current employment or had a desire to return to gainful employment. The study participants completed a demographic questionnaire, including age, gender, education, mental health diagnoses, insurance, marital and independent living status, sources and amounts of income, employment background and employment accommodation history. The next stage was to complete a battery of psychosocial outcomes, measuring fatigue, social support, depression, style of coping with health problems, their levels of optimism, their ADL's (activities of daily living – a measure of level of independence), anxiety levels and their readiness for employment. They were then all enrolled on a job placement assistance program, which was free and carried out by trained rehabilitation counselling staff. The study sample consisted mainly of well educated females who had relapsing-remitting MS, one third of whom were trying to maintain their jobs. 41% were receiving social security payments, indicating a large group of people who left their chosen career due to disability. Their psychosocial tests showed high levels of optimism, motivation to work and discomfort in accepting public subsidy; however, they also reported significant anxiety, depression and fatigue which could explain the job placement assistance programs high dropout rate (39%). The main reason for dropout was due to level of disability from MS. Other reasons stated for dropout included increased stress from job-hunting, which was thought by the participants to have precipitated relapses, and competing family interests. Conversely some dropped out as they had enrolled in full time education or had decided to resolve their employment problems independently. The high dropout rate could also be explained by the fact that 41% of those who embarked on the program were receiving social security and enrolling on a vocational rehabilitation course could have adversely affected the payments (Fraser et al, 2003).

La Rocca et al also found the attitude of the patient to be important. They studied 312 patients attending a major university based MS clinic and found level of disability and demographic variables counted for less than 14% of the variance in explaining the high rate of unemployment. They explored the psychosocial and environmental factors which could also influence employment status. The findings were that the patient's premorbid personalities and coping styles, as well as their ability to maintain an image of themselves as productive, working individuals played a significant role in vocational adaptation (La Rocca et al, 1985) which further emphasises the role of mediators in affecting participation.

Recently much of the evidence for the efficacy for vocational rehabilitation in MS has been drawn together by Khan, Ng and Turner- Stokes carried out a systematic review to look at the effectiveness of vocational rehabilitation (VR) on the return to work and employment of persons with multiple sclerosis. Two reviewers selected trials and rated them on their methodological quality. Two trials with a total of eighty participants met the review criteria – a randomised controlled trial and a case control trial. However both trials scored poorly on the methodological quality assessment and the conclusion was drawn that there was insufficient evidence for VR programs to be of use in altering rates of job retention, to support changes in employment or improvement in rates of re-entry into the labour force, these entities are known as "competitive employment". Nor was there any string evidence that VR changed levels of "work ability" in terms of improving participant's confidence in the accommodation request process or job seeking activity. The researchers were also unable to find any evidence for cost-effectiveness of the VR programs however they did conclude that their review identified some of the challenges in providing VR programs for those with MS. The authors recommended that clinicians be more aware of vocational issues in order to address the issues that can cause work issues earlier. More recommendations were that VR programs should be timely, proactive and practical in terms of managing work disability and should also focus on educating the wider community as well as employers. Lastly it was felt that future research in VR should include the development of appropriate and valid outcome measurement tools to be used for assessment of this area (Khan et al, 2009).

Systematic reviews of VR interventions in both RA and TBI also concluded that the knowledge regarding the effectiveness of VR programs in these areas is also insufficient (De Buck et al, 2002, Carney et al, 1999). A systematic literature review of vocational rehabilitation in patients with chronic rheumatic diseases was carried out. The VR programs had to be clearly defined interventions with the outcome of patients with rheumatic diseases re-entering or remaining in the workforce and the outcome was discussed in terms of specific vocational status; work disability, sick leave, job modification, paid occupation or re-training. Six trials met all the review criteria but none of these were controlled trials, five were retrospective follow up studies and the other was a prospective uncontrolled trial. Although five of the six showed a positive benefit of VR on work status, the evidence could not established due to the flaws of the intervention studies, in terms of methodological differences and shortcomings. Again these authors concluded that more research was needed to fully evaluate the clinical and cost effectiveness of VR programs and that any further studies must have robust baseline and endpoint measures (De Buck et al, 2002).

Although there were no published systematic reviews of VR in TBI there has been previous evidence of positive outcomes for specialist vocational rehabilitation programs (Gilworth et al, 2006). A prospective case series was used by Buffington et al to demonstrate the positive effects of a specialised brain injury vocational service delivery model on vocational outcome for eighty individuals with traumatic or other acquired brain injury. The results obtained were that 40% of the participants were placed in a job within three months of starting the project and 70% within twelve months with a ninety day job retention rate of 100%. The vocational intervention was early, beginning during the in-patient stay in the Rehabilitation Medicine department thus providing a smooth transition from medical to community-based out patient services (Buffington et al, 1997). In a 1999 systematic review of the effect of Cognitive Rehabilitation on outcomes for those with TBI the authors concluded that any future research must include relevant, durable outcome measures that would reflect the health and function of the individual (Carney et al, 1999). This was the same conclusion that had been drawn in the systematic reviews of vocational rehabilitation in multiple sclerosis and rheumatic diseases.

In summary job retention and job loss are substantive issues for those of working age who are living with chronic diseases. The evidence base for the efficacy of vocational rehabilitation across chronic disease, including multiple sclerosis, is currently weak yet strategies to continue in employment were seen as important for those with MS. One crucial requirement to facilitate such strategies would be early identification of the problems which put the job at risk. This project seeks to develop an instrument for that purpose, using a mixture of qualitative and quantitative methods. The methodology used had already produced a validated scale for work instability in Rheumatoid Arthritis (Gilworth et al, 2003) and Traumatic Brain injury (Gilworth et al, 2006).

# Chapter 3: Qualitative Research Methodology

## 3.1 Introduction to the Theoretical Approach

The initial part of this chapter covers the theoretical approach to qualitative research. A qualitative approach was chosen as the basis of understanding issues concerning job retention in MS and to identify potential items for a scale to measure the risk of job loss and the extent of work instability. A working knowledge of the theory behind qualitative research was necessary in order to successfully complete the qualitative methodology component of the research. It was important to explore the different types of sampling methods available to ensure that the sample used was of the correct size and was comprised of suitable participants. A detailed review of the different methods of data collection and analysis used in qualitative research was carried out so that the most appropriate types could be employed for the purposes of this study.

## 3.2 Purposive Sampling

The sampling for a qualitative study should be theoretical and not random and be designed to provide a close up, detailed or meticulous view of particular units which may constitute relevant cases (Mason, 1996)

Purposive sampling allows a case to be chosen because it illustrates a feature or process of interest to the research question. Purposive sampling demands critical thought regarding the parameters of the population of interest and choosing the sample carefully on that basis (Silverman, 2000). Many qualitative researchers employ purposive, rather than random, sampling methods by seeking out groups, settings and individuals where the processes being studied are most likely to occur (Denzin and Lincoln, 1994).

Setting up a sampling frame or *typology* to establish a matrix of, for example, employed people with multiple sclerosis is a methodology recognised in qualitative research (Stake, 1994). There are generally two main constraints, firstly there may not be examples to fit every cell and second, as resources often do not allow research into every single unit, a practical decision has to be made as to what should be studied (Silverman, 2000).

## 3.3 Data Collection

Qualitative research can provide a systematic way to explore certain issues once the study sample has been clearly defined. There are four main methodologies of data collection to choose from:

**Table 3.1 Methods of Qualitative Research**
**(Adapted from Silverman, 2000)**

| Method | Features | Claim |
|---|---|---|
| **1. Observation** | **Extended periods of contact** | **Understanding of "subcultures"** |
| **2. Texts and documents** | **Attention to organisation and use of such material** | **Understanding of language and other sign systems** |
| **3. Interviews** | **Relatively unstructured and "open – ended"** | **Understanding "experience"** |
| **4. Audio and video recording** | **Precise transcripts of naturally occurring interactions** | **Understanding how interaction is organized** |

### 3.3.1 Different uses for the four methods

These methods can be used in different ways and can be used in both qualitative and quantitative research. Observation is preliminary work in quantitative research e.g. prior to forming a questionnaire and in qualitative work is fundamental to understanding another culture. The interview technique employed in quantitative work is "Survey research", which is fixed choice questions to random samples. However qualitative interviews use "open – ended" questions to small samples and provide the text and transcripts necessary for further analysis. Textual analysis and transcripts are of some use in quantitative work for content analysis (counting in terms of researchers' categories) and to check the accuracy of interview records but are used in qualitative research to understand how participants talk in terms of categories and how these categories are organized (Silverman, 2000).

## 3. 4 Interviewing

Carrying out qualitative interviews was the methodology employed in the current study to develop the MS – WIS so providing the transcripts necessary for the first stage of the development of the scale.

### 3.4.1 Qualitative interviews

Qualitative interviewing uses semi – structured interviews with "open – ended" questions, as it is in this setting that the interviewee tends to share the information they feel is important (Silverman, 2000). With semi – structured interviews, the investigator will have a set of questions on an interview schedule, but the interview will be guided by the schedule rather than be dictated by it. The advantages of using the semi – structured interview methodology are that it allows a rapport to develop between interviewer and interviewee. It also allows a greater flexibility of areas to be covered during the interview and allows the interview to go into novel areas thus producing richer data for analysis which will examine the "lived experience" of the participant in their own words (Smith, 2003).

### 3.4.2 Sample Size for interviews

The qualitative researcher commonly asks "How many interview subjects do I need?" The answer is simply, "Interview as many subjects as necessary to find out what you need to know" (Kvale, 1996). Data should stop being gathered when categories are "saturated" and this happens when new data no longer sparks new insights. The kinds of research questions and the analytic level of the subsequent categories matter. Mundane questions may quickly lead to saturated but common categories, whereas novel questions may demand more complex categories and more sustained inquiry (Smith, 2003).

Bowling recognised that a common problem facing the researcher undertaking qualitative, in - depth interviewing is the question of sample size. Generally this is small due to a combination of the complexity of the data, which is costly to analyse, in terms of both time and money and because the data aims to provide insight into social phenomena (Bowling, 1997). There is no clear guideline regarding an appropriate "cut – off" point. There is, however, a rule of thumb which can be applied and that is when the same stories, themes, issues and topics are emerging from the study subjects i.e. saturation, then a sufficient sample size has been reached (Silverman, 2000).

The number of subjects necessary depends on the purpose of the qualitative study. If the number is too large, then it is not possible to assess the interviews in the depth required to make penetrating observations, this problem is usually due to a resource issue. However, if the number is too small, it will not lend itself to generalisations or to test hypotheses of difference between groups. For example, a representative population of approximately one thousand people is necessary in order to predict the outcome of a general election and so is not conducive to a qualitative interviewing method. However, if looking at the experience of a particular person, then only that person needs to be interviewed at length but this interview could not yield any generalizations (Kvale, 1996).

A combination of the limited time and resources available for research and the law of diminishing returns once saturation is reached mean, in current interview studies, the number of semi – structured interviews tends to be around 15 +/- 10 (Kvale,1996). Despite all the caveats about the choice of sample size a recent authoritative paper has suggested that in most cases, saturation is reached by twelve interviews (Guest et al, 2006). In the current study this has been balanced by a desire to be representative as indicated by the theoretical sample frame chosen (Table 3.2).

### 3.4.3 Interview Bias

This is the main problem encountered when carrying out qualitative interviews. "Outsiders" to a group influence "insiders", and vice versa (Miles and Huberman, 1994). The researcher can create behaviour and opinions in others that would not have occurred ordinarily which can lead to biased observations and inferences. This is known as "confounding", that is, when the "natural" characteristics of the setting are influenced by the artificial effects of the researcher – native relationship (Miles and Huberman, 1994). Therefore there are two possible sources of bias:

*The effects of the researcher on the case*

This occurs when the researcher threatens or disrupts ongoing social or institutional relationships. The interviewee considers why the researcher is there and what might be done with the collected information and they will change into an "on – stage role" or different persona – a presentation of oneself to the outsider. They may change their responses to be agreeable to the interviewer or to protect their own interests.

*Avoiding biases stemming from researcher effects*

- Stay as long on site as possible; spend some time simply hanging around, fitting into the landscape, taking a lower profile
- Use unobtrusive measures where possible
- Make sure the intentions are clear for participants; the reason for the researchers presence, what is being studied, the information being collected and how this will be done and how it will be used
- Co – opt an informant – asking that person to be attentive to the researcher's influence on the site and it's inhabitants
- Carry out some interviews "off – site", in a congenial social environment (café, restaurant, interviewee's home). This reduces the risk of appearing as a threat or as "exotic"

- Don't inflate the problem; the researcher is not such an important presence in people's lives as may be thought (Miles and Huberman, 1994).

*The effects of the case on the researcher*

During the course of the interviews or period of observation and, depending upon the relationship with the informants, the researcher's behaviour can also change. They can become more reassuring or, alternatively, can move into the investigative – adversarial mode. Either one will affect the data being collected.

*Avoiding biases stemming from the effects of the case on the researcher*

- Avoid the "elite" bias by spreading out the interviewees; include those from different backgrounds who can provide different information for the study and those on the periphery
- Spend time away from the study, spread out the interviews or bias from recent ones may creep in
- Include "deviant" cases
- Think conceptually; translate sentimental or interpersonal thoughts into more theoretical ones
- Do not display prior knowledge of subject as that can predispose to bias
- Show field notes/ transcripts to a colleague. Another researcher can be much quicker to see where and how biases or being misled is occurring

- Keep research questions firmly in mind; do not deviate too far from them by following alluring leads or drop them due to a more dramatic event (Miles and Huberman, 1994).

### 3.5 Data Analysis

There are diverse approaches to qualitative analysis, but there are features that are common to all approaches and so, some analytic practices may be used across different qualitative research types. Miles and Huberman (1994) defined a classical set of analytic moves arranged in sequence:

1. Affixing codes to a set of field notes drawn from observation or interviews
2. Noting reflections or other remarks in the margin
3. Sorting and sifting through the materials to identify similar phrases, relationships between variables, patterns, themes, distinct differences between subgroups and common sequences
4. Isolating theses patterns and processes, commonalities and differences and taking them out to the field when next collecting data
5. Gradually elaborating on a small set of generalisations that cover the consistencies discerned in the database
6. Confronting those generalisations with a formalized body of knowledge in the form of constructs or theories

Miles and Huberman described Qualitative data analysis as consisting of three concurrent flows of activity

1. Data reduction
2. Data display
3. Conclusion drawing/ verification

### 3.5.1 Data Reduction

This refers to the process of selecting, focusing, simplifying, abstracting and transforming the data that appear in written up field note or transcriptions. This starts prior to actual data collection as the researcher decides upon the conceptual framework to use, which cases to use, the research questions and the type of data collection chosen. During data collection, further data reduction occurs, for example, writing summaries, coding, identifying themes and writing memos. Data reduction is a continuous process from the beginning until data analysis is complete. It is a form of analysis that can be seen as "data condensation" (Tesch, 1990) as it sorts, focuses, discards and organises data in such a way that "final" conclusions can be made (Miles and Huberman, 1994).

#### *Coding the data*

Coding is the process of defining "what the data is about". In quantitative research *preconceived* categories or codes are applied to the data. In qualitative research, such as grounded theory, the researcher *creates* their codes by defining what they see in the data. Coding is the definitive link between collecting data and developing an emergent theory to explain these data. There are two stages:

1. An initial phase in which each line of data is named, followed by

2. A focused, selective phase that uses the most significant or frequent initial codes to sort, synthesize and organise large amounts of data (Smith, 2003).

   *Constant comparative methods* are used to establish analytic distinctions when coding, thus analytic comparisons are made at each level of analysis (Glaser and Strauss, 1967).

### 3.5.2 Data Display

A *Display* is an organised, compressed assembly of information that permits conclusion drawing and action (Miles and Huberman, 1994). Examples are newspapers and the gauges on petrol pumps in daily life, at work computer screens and handouts containing information on grounded theory. Historically, the most frequent form of display for qualitative data was *extended text.* This poses problems as it is cumbersome, bulky, it can be poorly structured and the important information it contains will be dispersed throughout making it easy for the researcher to jump to hasty, unfounded conclusions. As with data reduction, the creation and use of displays is not separate from the analysis, it is part of the analysis (Miles and Huberman, 1994).

### 3.5.3 Conclusion Drawing and Verification

This process should start from the beginning of data collection. Initially, the researcher should maintain an open mind, however the conclusions that may be vague at first may then become more explicit and “grounded” (Glaser and Strauss, 1967). The final, definite conclusions may not even appear until data collection has finished depending on how the data has been collected or stored and on the experience of the researcher.

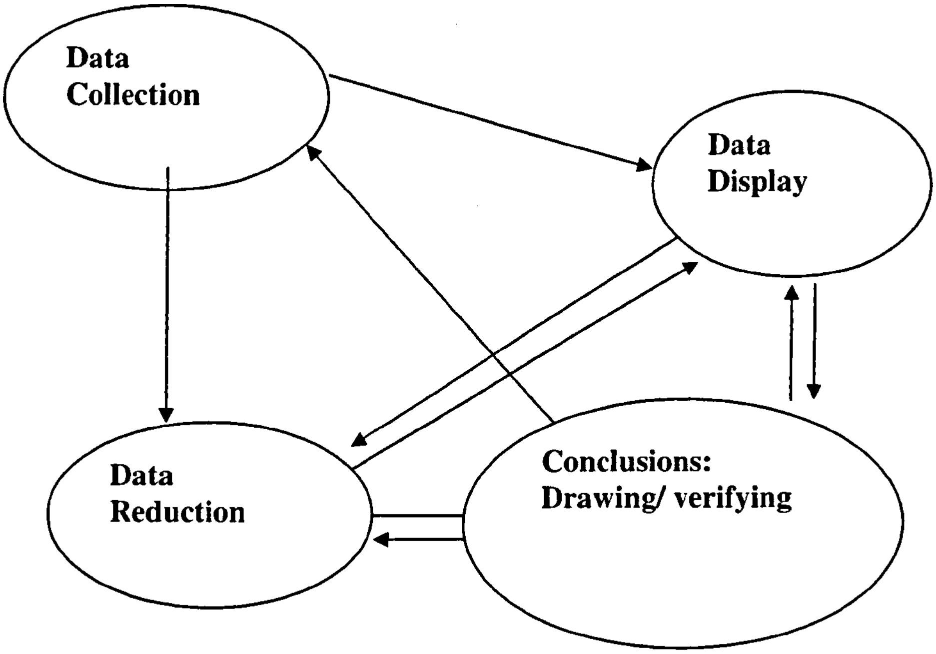
Verification consists of testing the meanings emerging from the data for their plausibility, their sturdiness and their *validity* or confirmability. If this does not happen the data the researcher is left with is an interesting story with unconfirmed truthfulness and therefore is of questionable use. There are certain instances when it cannot be said the claims of a research study are valid:

- When only a few exemplary instances are reported
- The criteria or grounds for including certain instances and not others were not provided
- The original forms of the material are unavailable

*Reliability* refers to the degree of consistency with which instances are assigned to the same category by different researchers or by the same researcher on different occasions. In order to assess this, the primary researcher must clearly document the procedure used and demonstrate that categories have been used consistently (Silverman, 2000).

*Verification* can range from being a brief thought crossing the analyst's mind leading to a review of the collected data to a lengthy discussion with colleagues to develop a "general consensus" on the subject (Miles and Huberman, 1994).

**Figure 3.1 Components of Data Analysis: Interactive model**



(Adapted from Miles and Huberman, 1994)

Qualitative data analysis is a continuous activity: The coding of data (data reduction) decides upon the content of a matrix (data display) and entering this data leads to further data reduction (Figure 3.1). As the matrix fills up, initial conclusions are drawn and the data is coded further in the data display, testing the conclusion. This process can be likened to that of collecting quantitative data; **data reduction** equals *computing means, standard deviations* and *indices.* **Data display** is likened to *correlation tables* and *regression printouts.* **Conclusion drawing/ verification** is akin to *significance levels* and *experimental/control differences.* Both qualitative and quantitative data analysis have well – defined methodologies, however the qualitative researcher has a more fluid position as there is overlap between the qualitative processes used (Miles and Huberman, 1994).

## 3.6 Analytic Methods for Qualitative Data

There are various methodologies used for the analysis of qualitative data. A thorough review of these revealed content analysis and grounded theory to be the methodologies best suited to the purposes of the project. A sophisticated form of content analysis allowed for the development of items to be used in the scale, a process known as item selection and the issues of working with multiple sclerosis were explored by using a grounded theory approach to examine the transcripts.

### 3.6.1 Content Analysis

Content analysis is a commonly used methodology when analysing qualitative data. The open – ended dialogue from the interviews is coded into closed categories, which summarise the data in a more systematic fashion. The categories are derived from the data itself or from the pre – existing theoretical framework. When the categories are derived from the data, usually using grounded theory, it is known as a "bottom – up" approach. The aim of the analysis is to illustrate each category with a representative quotation or "item" from the data (Smith, 2003).

An advantage of content analysis is that it allows the conversion of qualitative data into a quantitative form by counting the number of times a response falls into each category. The main advantage of content analysis is that it can provide a useful summary of opinions or beliefs that the study participants have about the topic. The main disadvantages are that a great deal of detail is lost and it can be difficult to select quotations which are representative of the categories yet easy and interesting to read, as Smith said in 2003 "Naturalistic talk doesn't come in sound bites". Lastly, the sense of individual participants is lost in the isolated quotations that result from this type of analysis (Smith, 2003).

### 3.6.2 Grounded Theory

A simplified model of Glaser and Strauss's famous account of grounded theory (1967) is:

- An initial attempt to develop categories which may illuminate the data
- An attempt to "saturate" these categories with many appropriate cases in order to demonstrate their relevance
- Developing these categories into more general analytical frameworks with relevance outside the setting (Silverman, 2000).

They challenged pre – existing ideas around qualitative research, such as:

- The arbitrary division of theory and research
- Prevailing views of qualitative research as a precursor to more "rigorous" quantitative methods
- Beliefs that qualitative methods were impressionistic and unsystematic
- Separation of data collection and analysis phases of research
- Assumptions that qualitative research could not generate theory (Smith, 2003)

The grounded theory process begins by exploring general questions about a research topic of interest. Data is collected from people who have experience of the topic, in what they say and do. The researcher using grounded theory will have some background assumptions that will lead them to look for particular issues or processes in their data. Grounded theorists often begin their studies with certain research interests and a set of general topics which allows for ideas to be pursued and particular questions asked about the study topic, however, every grounded theory researcher should remain open to new views during the research (Smith, 2003).

The core components of grounded theory studies are analytic categories developed while studying the data rather than preconceived concepts or hypotheses. These categories move the study towards abstract analysis using theories developed in the empirical world. Grounded theorists evaluate fit between their initial research interests and the emerging data, however they do agree on starting analysis with the data (Smith, 2000). Data collection is increasingly focused during a grounded theory research project, this is because the analytic work being carried out leads to the gathering of more specific data. Grounded theorists follow leads to develop their emerging theoretical categories (Glaser, 1978).

The inductive nature of grounded theory methods assumes an open flexible approach. The methodological strategies are shaped during the research rather than having them planned before beginning the data collection which can be altered in pursuit of the most interesting and relevant material (Smith, 2000). The purpose of grounded theory is to develop a theoretical analysis of the data that fits the data and has relevance to the area of study. The systematic procedures of grounded theory enable qualitative researchers to generate ideas which, later, may be verified through more traditional quantitative methods (Smith, 2000).

## 3.7 Qualitative Methodology: An introduction to the Practical Approach

The aim of the first part of the project was to gather and then analyse the qualitative data that both gave an understanding of working with MS, as well as items for the scale. This involved a range of tasks, including, the process of recruitment to the study, the sampling strategy, interviews as the main method of data collection and the role of the researchers involved in the data analysis. While the focus of the work was item generation, the wealth of information contained within the interviews allowed for a more detailed exploration of the issues of working with multiple sclerosis.

## 3.8 Settings

Subjects for the study were recruited from multiple sclerosis clinics carried out within the Leeds Teaching Hospitals NHS Trust, from the existing MS database and via St James's MS specialist nurses. All subjects had a definite diagnosis of MS and were aware of their diagnosis. Guidelines from the international panel on the diagnosis of MS were used (McDonald et al, 2001). Ethical approval was obtained from the Local research ethics committee (LREC) of the Leeds Teaching Hospitals NHS trust.

All participants were given an information leaflet regarding the study and their signed consent was obtained prior to the interview. They were informed the interview would last approximately one hour and be taped, but were assured the interview would be halted immediately at their request.

### 3.9 Study Participants

Employment issues in multiple sclerosis were being studied and so it was important to access people of both sexes carrying out different jobs, for example, "blue" or "white collar" work. Inclusion criteria were those with MS who were still working, either full or part time or had been off work on sick leave for less than 6 months and were aged between 18 – 65 years of age. Exclusion criteria were those with a co-existing serious illness and those who had been on sick leave for over six months as their interviews could be influenced by recall bias (Reisine et al, 1995).

### 3.9.1 Sampling

Purposive sampling of the study subjects was used as this best fitted the demands of the project. It was used to ensure that a wide spread of occupational groups across both sexes were included and that their experiences were represented accurately. This was important as the scale was to be representative of, and generalisable to the working MS population. Purposive sampling as construed in this study was actually introduced to decrease bias and ensure that the sample was representative.

Most autoimmune diseases show a female preponderance and MS is no exception with a 2:1 female to male ratio and therefore it was planned to recruit twice as many women than men (Ebers et al, 1997). Job categories were defined by "white-collar" or "blue-collar" work which indicate how physical the job is. "White-collar" work tends to be more sedentary than "blue-collar" work which leans towards manual labour. Disease type was defined in terms of progressive and non-progressive disease. It was thought to be important to categorise disease type as previous studies have shown that those with progressive disease are more likely to be unemployed than their relapsing-remitting counterparts (Bauer et al, 1965). The actual target for recruitment is shown in table 3.2.

**Table 3.2 The Theoretical Sampling Frame**

| | | Male | Female |
|---|---|---|---|
| White Collar Worker | Progressive Disease | | |
| | Non Progressive Disease | | |
| Blue Collar Worker | Progressive disease | | |
| | Non Progressive Disease | | |

## 3.10 Conducting the Qualitative interviews

Qualitative research provided a systematic way to explore the issue of working with MS. Carrying out interviews was the most appropriate method of data collection for this project as it was necessary to understand and quote the "experience". The interviewees were given a patient information sheet and were asked to sign a consent form prior to starting the interviews. The interviews were audio - recorded with the permission of the interviewees and transcribed verbatim for data exploration and analysis. The transcribing was carried out by the secretary of the Rehabilitation Medicine Research department who was experienced in transcribing having previously undertaken such work for other qualitative research projects in the department. The interviewer then checked the transcribed tape for accuracy and clarity.

The participants were asked to describe their experiences of work and the impact their diagnosis of MS had made on this. The interviews concentrated on factors relevant to the participants "struggle" to continue in work following the onset of MS. This approach was based on previous research methodology carried out by Gilworth et al in the Development of the Rheumatoid Arthritis Work Instability Scale (Gilworth et al, 2003). The participants chose where the interview was carried out, either in their home, their workplace, at Leeds University or an alternative location which was more appropriate for them. The interviewee also chose who was present during the interview. Only one person chose to have a spouse with them, all other participants felt they could speak more freely when being interviewed alone. On average, the interviews lasted approximately one hour before drawing to a natural close.

Semi – structured interview techniques ensured all relevant points pertaining to employment in multiple sclerosis were covered and enabled certain subject areas to be explored further. Therefore, in keeping with semi – structured techniques, the interview style employed open-ended questions guided by the purpose of the study which were raised if not already covered during the course of the interview (Appendix 4).

Approximately one third of the interviews were carried out by a co-researcher, with the rest being conducted by the primary researcher. This approach decreased the chance of interviewer bias as the co – researcher had no experience in multiple sclerosis and was previously unknown to the interviewees, whereas the primary researcher was employed as a medical doctor specialising in multiple sclerosis. Many of the interviewees were known to the primary researcher as patients, which may have influenced the interview content and so collaboration on the interviews was of great importance.

### 3.11 Data Analysis

Once the interviews were complete and had been transcribed verbatim the subsequent analysis was broken down into two different techniques: qualitative analysis of the transcripts to detect any themes that may be arising (section 3.11.1) and to identify items suitable for consideration of inclusion in the Work Instability Scale (3.11.2).

#### 3.11.1 Qualitative analysis of transcripts

This was used to identify emerging themes regarding employment and MS. *Grounded theory* was used to analyse the transcripts further and identify emerging themes relating to employment issues in MS. This methodology was strengthened by the fact that one of the researchers was very experienced in qualitative research but did not have any prior knowledge of MS. They were not familiar with the literature prior to the initial analysis and therefore did not have any pre-conceived ideas regarding work in MS which minimised biases stemming from the effects of the case on the researcher (Miles and Huberman, 1994).

The grounded theory approach used in this study was taken from Glaser (Glaser et al, 1967) which involves a systematic approach to the analysis, the theoretical basis of which has already been extensively discussed (Section 3.6.2).

Firstly, the aforementioned experienced qualitative researcher and the candidate both read the extended text of the transcript to ensure familiarisation with the data prior to coding it. All transcripts were read at least twice by the candidates prior to data analysis and then coded using a scheme developed from the issues identified in the interviews. Once this was completed the categories were reviewed and collapsed into headings, the process of data reduction. This ensured that themes were not duplicated and allowed for cross referencing of them. This process was repeated until the point was reached when the researchers could not identify any new information that was emerging. The results of this process are presented in chapter 5.

### 3.11.2 Item Selection

The primary objective of the study was to produce a screening questionnaire for work instability. Consequently the objective of Item Selection was to produce a draft questionnaire. The transcribed interviews were read several times by three researchers, two of whom had previous experience in the production of other disease specific work instability scales. A content analysis approach identified potential items for the scale, a process known as item selection (see section 3.6.1 for the theoretical approach and the results of this are shown in chapter 5).

The items were phrased in lay terms and, wherever possible, the exact wording from the interviews was used. This meant that actual quotations from the interviewees were used to form potential items for the measure wherever possible. Items were selected if they were common issues relevant to work instability in the MS population, were expressive of a single idea, applicable to all respondents i.e. not age or gender based ("it is getting harder to work as I get older" or "things are worse around the time of my period"). They also had to be able to be expressed in the first person. This process which had been validated when used in other similar studies (Gilworth et al, 2003, Gilworth et al, 2006) generated an item pool for the questionnaire and yielded the first draft of the questionnaire.

This initial list of items was then reviewed by the same three researchers and duplicated or unusable (for reasons as discussed above, gender etc) items were removed.

### 3.12 Face Validity

Once the draft questionnaire was available it was subjected to a process known as *Cognitive Debriefing*. The objective of this was to assess the face validity of the draft WIS. Field tests were carried out on ten employed people with MS who were asked to complete a draft of the scale and discuss how easy it was to understand and complete. Seven women and three men who were still in work were used as this is roughly representative of the female:male ratio seen in MS. Ten candidates was felt to be a very reasonable number for face validity and was consistent with the evidence from previous published work that had indicated that smaller numbers were appropriate for face validity testing. For example, five patients were used for the cognitive debrief in the development of a work instability scale for traumatic brain injury (Gilworth et al, 2006). However it was acknowledged that more than ten people may be needed to be involved in the cognitive debrief if the majority of the items proved to be troublesome. The participants were asked to complete the questionnaire with a view to highlighting items that were difficult to understand, difficult to interpret or quite simply did not make sense. It was also assessed whether or not the items in the scale conveyed the same meaning to everyone. The cohort of patients used at this stage had not been previously engaged with the project and the same methods of recruitment were used as for the qualitative interviews.

The cognitive debrief was carried out in a clinic setting as this is where the scale would be mostly be used. The primary researcher carried out the Cognitive Debriefing and therefore was present and available to answer questions whenever necessary.

The aim of this stage of the study was to identify and discard items that the participants felt were unclear, not relevant or were difficult to comprehend. Following this a second draft of the scale was produced, the results of which are discussed in section 5.3.3.

# Chapter 4: Quantitative Research Methodology

## 4.1 Introduction to the Theoretical Approach

Once the draft questionnaire was complete, the project switched to a qualitative methodology in order to test the psychometric properties of the scale. The initial part of this chapter outlines the theoretical approach to quantitative research. A working knowledge of the theory behind quantitative research was necessary in order to successfully complete the quantitative methodology component of the research. Initially it was important to understand the different types of rating scales available and how this might influence the analytical strategy. A detailed review of single and multi – item outcome measures did reveal a multi – item scale to be the most suitable way of measuring the problem of work instability in multiple sclerosis. It was also important to ensure a single construct was being measured, and to allow for the validity and the reliability, i.e. *the Internal Construct Validity* of the scale to be tested. Rasch analysis uses a mix of traditional and modern psychometric approaches is fully discussed in this chapter.

## 4.2 Rating scales

Rating scales are of great importance because they are a method of measurement and measurement is crucial because inferences are then based on it (Bond, 2001). In clinical trials variables are measured, subjected to statistical analysis and conclusions about a treatment or intervention are based on this. These conclusions influence patient care, prescribing, policy making and the expenditure of public funds, thus the validity of inferences from clinical trials is directly dependant on the quality of the measurements used. An example of a clear cut, easy measurement is mortality rates where death is the outcome measure. Some variables are more difficult to measure as they have less defined outcomes, such as, "impact of disease", level of "work instability" or "quality of life". However such variables are being increasingly used to evaluate clinical practice and therefore they must be measured as rigorously as possible (Hobart, 2003).

Large scale trials have been carried out for interferons and glatiramer acetate in multiple sclerosis (MS). They showed a reduction in relapse rate and reduction in magnetic resonance image (MRI) lesions over time, but a less conclusive reduction in the progression of physical disability (Paty et al, 1993). These findings have had a huge impact; calls for further research to explore the relationship between neuropathology and level of disability, calls for definitive studies that are free from pharmaceutical influence, a controversial review by the National Institute for Clinical Excellence (NICE)and the UK Risk Sharing Scheme for prescribing disease modifying therapies (DMTs). However the choice of rating scale used in these trials has never been questioned. In these trials, progression of disability was measured using Kurtzke's Expanded Disability Status Scale (EDSS) (Kurtzke 1983, appendix 2). There are potentially profound and inherent weaknesses within this scale; firstly, it mixes the measurement of different health domains. It measures impairment in the early part of the scale, mobility in the middle part and bulbar function in the upper part and therefore it is not a pure disability measure. Secondly, all such items generate ordinal scores rather than interval scores (Hobart, 2003) and lastly, the EDSS is poor at detecting change over time (Sharrack et al, 1996). These facts question the strength of any clinical conclusions made on the basis of the analysis of EDSS scores. The use of different statistical methods to analyse results acquired from rating scales cannot overcome flaws within the rating scale itself and maximising trial design will not overcome the problems caused by weak rating scales and vice versa. The same level of attention should be applied in both arenas (Hobart, 2003).

### 4.2.1 Types of scales

There are two main types of rating scales in use at present:

1. **Single item scales** e.g. Ashworth Spasticity scale
   EDSS
2. **Multi – item scales** e.g. The Leeds MSQoL

Single and multi – item scales contrast in their interpretability and scientific rigor. It is now more recognised that single item scales can be weak measures as each level of the scale is open to random error, they also have poor validity and reliability. Random error is less of a problem in multi – item scales as each item contributes unique information but does not act as an individual rating scale. Reliability is high as combining across items cancels out the unavoidable random error associated with each single item (Wright et al, 1989).

### 4.2.2 The Development of Rating Scales

Hobart argued that total scores generated by summing item scores are really good measures providing all of the items address the same underlying construct. This is the basic theory underlying multi – item scales and that multi – item development has four stages:

1. The construct, that is, the concept that is being measured needs to be defined
2. A pool of items is generated so that all important issues are considered for inclusion in the final scale

3. The item pool is administered to a sample of patients and the scale is developed from the analysis of the resulting data. The scale should be a reliable and valid representation of the construct
4. Examine the full properties of the scale in independent samples

### 4.2.3 Attributes and Criteria for reviewing Instruments

The Scientific Advisory Committee (SAC) of the Medical Outcomes Trust issued guidelines for evaluating rating scales in 2002 and identified eight attributes that serve as the principal foci for SAC instrument review

1. Conceptual and measurement model

Definition – A conceptual model is the description of the concepts and the populations that the measure is intended to assess and the relationship between those concepts.

2. Reliability

Definition – Test reliability is the degree to which an instrument is free from random error.

Classical approaches for examining this are

a) Internal consistency reliability using Cronbach's coefficient alpha or item response theory
b) Reproducibility using test – retest or inter – observer reliability. This can be done by judging the reproducibility or stability or an instrument over time (test – retest) and inter – rater agreement at one point in time.

3. Validity

Definition – The validity of an instrument is the degree to which the instrument measures what it is supposed to. The process of Internal Construct Validity provides information about how the health measurement outcome scale is performing. It is a more detailed examination of the structure of the scale, particularly related to unidimensionality and scaling properties (Kucukdeveci et al, 2004).

4. Responsiveness

Definition – Refers to an instrument's ability to detect change, that is, whether the measure can detect, even small, differences in outcomes. Sensitivity to change or responsiveness is an important part of the construct validation process

5. Interpretability

Definition – This is the degree to which one can assign easily understood meaning to an instrument's quantitative score which, in the case of questionnaires, is ordinal data

6. Respondent and administrative burden

Definition – Respondent burden is the time and effort placed on those to whom the instrument is administered. Administrative burden is the demands placed on those who administer the instrument.

7. Alternative forms

Definition – Alternative forms of administration that can be used for the development and application of instruments can include self – report, trained observer rating or versions of the original source instrument that are to be completed by proxy respondents such as parents, spouses or other substitute respondents

8. Cultural and language adaptations (translations)

Definition – Instruments can be adapted or translated for applications across populations, for example an instrument developed in English may be translated for use in other languages. Culturally specific approaches to some activities, bathing for example, can make adaptation of the outcome measure complicated (Tennant et al, 2004, Kucukdeveci et al, 2004). Therefore the SAC states that measurement properties of each cultural or language adaptation should be judged separately for evidence of reliability, validity, responsiveness, interpretability and burden (The Scientific Advisory Committee of the Medical Outcomes Trust, 2002).

## 4.3 Internal Construct Validity; Rasch Analysis (The Theoretical Approach)

In the current study, the conceptual model is provided by Internal Construct Validity; Rasch Analysis, the International Classification of Functioning (ICF) and the prior notion of Work Instability. The pool of items was based upon qualitative interviews and a strict measurement model has been chosen as the basis of testing reliability and the (internal) construct validity of the scale. Rasch analysis is the principal modern psychometric approach used in health outcome measurement to ascertain the reliability and internal construct validity of the scale (Rasch, 1980, Andrich, 1988).

Fitting data to the Rasch measurement model allows for a comprehensive analysis of various measurement properties in a unified framework. It also confers the added benefit of allowing the transformation of the raw ordinal score into an interval measurement. This is the principal advantage of adopting Rasch analysis over and above more classical psychometric measurement approaches.

### 4.3.1 The Rasch Model

The Rasch model is a definition of measurement, a mathematical derivation from the requirement that stable linear measures be constructed from the ordered qualities of rating scale data. Therefore the aim of Rasch analysis is to determine the extent to which observed rating scale data satisfy this stringent definition. Stable linear measures can be constructed only when the data satisfy the model and so data which satisfies the model is sought (Massof, 2002).

The Rasch model is a unidimensional model which asserts that the easier the item the more likely it will be passed, and the more able the person, the more likely they will pass an item compared to a less able person. It assumes that the probability of a given respondent to give a ‘correct’ answer to a particular item is a logistic function of the relative distance between the item location and the respondent location on a linear scale.

In other words the probability that a person will affirm an item is a logistic function of the difference between the person’s ability [ $\theta$ ] and the difficulty of the item [b] (i.e. the ability required to affirm item i), and only a function of that difference. (The equation of the Rasch model is shown on the right, Figure 4.1)

**Figure 4.1 The Rasch Model**

$$p_{ni} = \frac{e^{(\theta n - b_i)}}{1 + e^{(\theta n - b_i)}}$$

where $p_{ni}$ is the probability that person $n$ □will answer item $i$ correctly [or be able to do the task specified by that item], $\theta$ is person ability, and $b$ is the item difficulty parameter.

From this, the expected pattern of responses to an item set is determined given the estimated $\theta$ □ and b. When the observed response pattern coincides with or does not deviate too much from the expected response pattern then the items constitute a true Rasch scale (Van Alphen et al, 1994). Taken with confirmation of local independence of items, that is, no residual associations in the data after the Rasch trait has been removed, this confirms unidimensionality (Banerjee et al, 1997, Smith, 2000).Thus Rasch analysis provides a linear estimate of both person ability (i.e. their level of work disability) and item difficulty (i.e. the impact on work disability represented by the item).

### *Thresholds and Response categories*

The items in a scale can have simple "yes/no" responses or a range of responses for each item, such as in a Likert scale. A Likert scale asks questions to which people respond on an agreement scale, that is, strongly agree, agree, neutral, disagree, or strongly disagree. These categories should reflect an increasing amount of the trait being measured. Thus an "ordered category" implies that the categories have been numbered so that a higher numbered category is thought to imply more of the latent variable under investigation. Numerically ordering of categories as qualitative advances along the variable is a prerequisite to Rasch measurement. In the course of Rasch analysis, one may discover that the imagined category ordering is not supported by the scale data. Remedies include renumbering the categories, collapsing adjacent categories or dropping items. 'Categories with low frequencies are problematic as they do not provide enough observations for an estimation of stable threshold values. Such infrequently used categories often indicate unnecessary or redundant categories. Hence these are the categories that should be collapsed into adjacent categories. The recommended minimal number of responses per category is 10. Linacre thresholds are the boundaries between these categories and it is possible to observe "disordered thresholds" in the data (Bond and Fox, 2001).

### *Individual Item Fit*

The data collected from questionnaires should meet the expectations of the Rasch model. To test this the Rasch model uses chi square fit statistics. These look at the difference between what was observed and what is expected. This difference can be expressed as a residual. The residual values represent the difference between the Rasch model's expected and the actual observed score.

Misfit of an item indicates a lack of the expected probabilistic relationship between the item and other items on the scale. This may derive from disordered thresholds, from Differential Item Functioning (DIF, see below) or may indicate that the item does not contribute to the trait under consideration. Bonferroni corrections are applied to fit statistics due to the number of statistical tests undertaken for any given scale (Bond and Fox, 2001).

### *Differential Item Functioning*

Essentially the scale should work in the same way, irrespective of which group is assessed i.e. whether men or women, or younger and older age groups. Thus, the location of items along the measurement construct should remain the same between these groups. This type of analysis is given the name Differential Item Functioning (DIF). The basis of the DIF approach lies in the item response function, the S-shaped trace of the proportion of individuals at the same ability level who affirm an item. Under the requirement that the ability under consideration is unidimensional, if the item measures the same ability across groups then, except for random variations, the same curve is found irrespective of the nature of the group for whom a function is plotted. Items that do not yield the same item response function for two or more groups display DIF and are violating the requirement of unidimensionality. The statistical test of DIF is an ANOVA of the residuals. When some but not all individuals display DIF, it is possible to make an adjustment to allow items with DIF to vary by group by splitting the offending items across the subgroups to form separate items for each group (Van Alphen et al, 1994).

### *Person and Item Location*

Rasch analysis provides a linear estimate of both person ability and item difficulty. The logit scale, which is the measurement unit common to both person ability and item difficulty, is an interval scale; the equal distances anywhere up and down that scale have equal value. The mean difficulty of all the items has a location value of 0.0 logits. The items are situated along the interval scale according to their difficulty. The average person ability will indicate how well the scale is targeted at the sample with less well targeted scales having an average person ability some distance away from the central zero logit (Bond and Fox, 2001).

### *Overall Fit*

Three overall fit statistics are provided by the Rumm2010 package which determines how well each item contributes to defining a single dimension. Two of these statistics are item-person interaction statistics distributed as a Z statistic with mean of zero and standard deviation of one (this indicates perfect fit to the model). A third is an item-trait interaction statistic reported as a Chi-Square, reflecting the property of invariance across the trait (Andrich et al, 2000).

### 4.4 Introduction to the Practical Approach

This stage of the project had several practical aspects to ensure that the scale was reliable and valid, including subjecting the second draft of the scale to Rasch analysis. Finally comparison was made with the current "Gold Standard" means of assessing work instability to identify items which discriminated across expert defined levels of risk. The relevant results are presented in Chapter 6: Quantitative Research Results – Development of the Work Instability Scale

### 4.5 Sample Characteristics

The draft questionnaire was sent by post to people who fitted the recruitment criteria within the West Yorkshire area (inclusion and exclusion criteria has been identified in Chapter 3). There were seven hundred people on the Leeds MD database aged less than sixty – five. Previous postal questionnaires carried out using the Leeds MS population of yielded a good response rate of approximately 82 – 97% (Ford et al, 2001) based on this, it was expected that at least two thirds of the questionnaires would be returned. The literature review suggested that, at most, half of this population would be in employment (Busche et al, 2003, Bourdette et al, 1993, Rao et al, 1991). Therefore, initially, three hundred and fifty questionnaires were sent out as approximately one hundred replies were need for the initial statistical examination of the scale.

The patient sample was taken from the MS database and was randomly generated using a statistical software package (SPSS – statistical package for the social sciences). Unfortunately the response rate was poorer than anticipated and patients who were in work had to be recruited from clinics, in order to meet the one hundred responses needed for statistical analysis. The database had not been updated with new cases for approximately five years and did not include patients who had been diagnosed or had moved into the area more recently.

### 4.6 Internal Construct Validity; Rasch Analysis (The Practical Approach)

Rasch analysis was carried out on the responses from one hundred and twenty questionnaires using the Rumm 2020 computer software program. This statistical approach was used to define and quantify the construct of WI. It focused on the range of measurement of the scale, its unidimensionality and hierarchical ordering of the items (Smith, 2000, Banerji et al, 1997). Thus Rasch analysis provided a linear estimate of both person ability (i.e. their level of work instability) and item difficulty (i.e. the impact on occupational function represented by the item). Demographic details regarding the patient and their replies to the items on the scale were entered into SPSS to create a database. Some of the returned questionnaires were incomplete and therefore those responses were entered as missing. When completing the questionnaire the study participants were asked to fill the response that applied best to them *at that moment.* The choice of response to all the statements in the scale was "True" or "Not true". This response was entered into an SPSS (Statistical Package for the Social Sciences) computer program and then the data had to be transferred to the Rumm 2020 software package before analysis could be conducted.

Internal Construct Validity of the scale was carried out by subjecting the scale to Rasch Analysis. The aim of this was to formally test the properties of the scale and for item reduction. Rasch analysis was undertaken using the RUMM2020 computer software package (Andrich, 2001). After Rasch analysis the number of items in the scale was reduced, as redundant items were identified and discarded.

### 4.6.1 Criterion Validity

There is no instrument currently available to measure work instability in MS and thus to provide a defined gold standard. At present the only way to identify people who are at risk of work instability is through a comprehensive vocational assessment (VA). This type of assessment can be offered by the Employment Service Disability service in the United Kingdom and includes analysis of work tasks and postures as well as analysis of the medical history. Following such an assessment various interventions may be offered ranging from advice and relatively minor changes in work practice to provision of specialist tools and equipment and office furniture. The degree of intervention required is dependant on the degree of work instability present.

Consequently, the criterion validity of the scale can be assessed by comparing it with the only available present "gold standard" which is a comprehensive vocational assessment (Gilworth et al, 2003). Therefore comparison of the scale against a "Gold Standard" assessment was carried out to assess the reliability and sensitivity of the draft measurement against the pre-existing method of determining work instability which is vocational assessments (Gilworth et al, 2003).

A sample of thirteen subjects who had returned the questionnaire and were in work agreed to participate in a full vocational assessment. These were representative of the larger sample by age and sex. The vocational assessments were carried out by two professionals who were both chartered physiotherapists and registered ergonomists. Two assessors were used in order to minimise the risk of bias and they were also the assessors used in the development of the Rheumatoid Arthritis Work

Instability Scale (RA – WIS) so were already familiar with the process (Gilworth et al, 2003). The vocational assessments followed an agreed format (Table 4.1) and took approximately one hour to complete. The first two patients were assessed by both ergonomists to ensure consistency between the two assessors. Independent assessments were only carried once it was clear that they were achieving 100% agreement. The experts then carried out full vocational assessments on the remaining eleven subjects. They were blinded to the results of the draft questionnaire filled in by all thirteen participants immediately before their assessment. At the end of each assessment the expert indicated the level of intervention judged appropriate by allocating a Work Instability Score (WIS) between 0 - 4 representing increasing risk of work instability (Table 4.2). The scoring system was devised by the experts who regularly carry out this type of assessment and reflected their opinion following an assessment covering all of the areas of the agreed format (Table 4.1). The resultant grading was a clinical judgment made by the assessor/s in light of the evidence obtained from covering the topics in Table 4.1. and using a structured ergonomic assessment based around an evaluation of the participants working life. The assessors were highly experienced individuals from a company known as "Work Fit" which is based in Leeds. Work fit offers an occupational physiotherapy and ergonomics service with the aim of creating a health conscious work place, which over the years has worked with a variety of employers, across a wide range of sectors – from multi-national organisations, with their own health and safety officers to small local enterprises with a handful of employees ("Work Fit" website) and was involved in the development of the RA-WIS (Gilworth et al, 2003). The explanation beneath each level of work instability reflects the expert's view of the level of intervention recommended to assist the individual in terms of job retention and this "gold standard assessment" was also used and validated in the development of the RA-WIS, thus was felt to be the best pre - existing comparison measurement available (Gilworth et al, 2003).

The questionnaire responses of these subjects were validated against the result of the "gold standard assessment" using Multiple Response Analysis. Multiple Response Analysis is a SPSS procedure (SPSS - statistical package for the social sciences) which, in lay terms, looked at the frequency of affirmation of the items across the different levels of risk, that is across level 0 – 4 (Table 4.2). Items which discriminated across the levels of WI were retained. Rasch analysis was undertaken once again on the revised item set and the results of these analyses were used to produce a version of the questionnaire for final test – retest assessment.

**Table 4.1 Core elements in the full vocational assessment for work instability**

**Health Situation**

1. Medical history
2. Details of current condition
3. Current symptoms
    - Aggravating and easing factors
    - Limitations
    - Pattern of symptoms
4. Clinical management

**Work Situation**

1. Job description
    - Task analysis
    - Hours worked/ shift patterns/rest breaks
    - Works organization/ task variety/ control
    - Management culture/ style
    - Tools/ equipment
    - Environment
2. Postural analysis
3. Physical work factors
    - Repetition
    - Force
    - Duration
4. Getting to work and access in and around the workplace

**Hobbies**

**Other perceived stressors**

**Table 4.2 Levels of work instability (WI) used by experts completing gold standard assessments**

| Zero WI | Level 1 WI | Level 2 WI | Level 3 WI | Level 4 WI |
|---|---|---|---|---|
| No problems at work | Minor problems at work requiring advice only | Advice and intervention (for example modifications to work practices and/or use of coping strategies) are required for the individual to continue in this job and **complete all aspects of the job** | Some aspects of the job are unsuitable **even** with advice and intervention it is unlikely the individual will be able to complete **all** aspects of the job | Mis-match is such that **even** with modifications to work practices and advice and/or use of coping strategies **the majority** of the job is unsuitable and the individual is unlikely to cope. |

### 4.6.2 Reliability

The revised scale with the reduced number of items was then administered to a further sample of participants on two occasions, two weeks apart to test reliability of the questionnaire over time. Three hundred and fifty questionnaires were dispatched to a second patient sample which had also been randomly generated using SPSS. Once the replies from these were received a second identical questionnaire was sent to the respondents after a two week interval and the results entered into SPSS.

Then the scale was subjected to further Rasch Analysis to assess the test – retest reliability and internal consistency of the scale and to confirm the validity of the questionnaire.

## 4.7 Scale comparisons

The draft work instability questionnaire was also accompanied by other scales which were sent out in a booklet to complete. The aim of this was to specifically examine the relationship between the person's ability to continue to work and the effect this has on other areas of their life, for example, on their quality of life and social function and vice versa. It was also to explore what effect MS symptoms and fatigue levels have on employment. The review of the literature had suggested correlations between the MW-WIS and the other constructs would be found. The other scales sent out were:

1. The Leeds Multiple Sclerosis Quality of Life Scale ( LMSQoL) (Ford et al, 2001) (Appendix 5)
2. The Leeds Social Function Scale (SFS) (© *University of Leeds)* (Appendix 6)
3. The Multiple Sclerosis Impact Scale (MSIS – 29) (Hobart et al, 2001) (Appendix 7)
4. The Modified Fatigue Impact Scale (MFIS) (Fisk et al, 1994) (Appendix 8)

The scores from each of the scales were calculated and the results were compared to see if there was any correlation between them. Spearman rank correlation was used throughout as all of the above scales were ordinal scales and therefore needed non – parametric statistics. The Spearman's correlation coefficient was known as rho for these calculations.

# Chapter 5: Qualitative Research Results – Working with Multiple Sclerosis

## 5.1 Introduction

The findings after analysing the qualitative data that provided the basis for the scale are discussed here. There were two separate and definite types of results obtained. The first being the themes that arose during the grounded theory approach to analysing the typed manuscripts, exploring the issues of working with multiple sclerosis in great detail. The second was that the items for the scale were identified and pooled to be used in the scale and this was done by using a form of content analysis.

The actual sample of participants used is discussed and how the reality of this differed from the proposed sampling frame.

## 5.2 Study Participants

The resultant size and composition of the sample differed from the planned sampling frame. Twenty seven people were interviewed for the study, fifteen men and twelve women. The reason this number of people were interviewed was that saturation was actually reached at that point, that is, no new themes were from the interviews. Saturation has previously been discussed at length in chapter three however it is worth mentioning at this point that Kuzel actually tied his recommendations to sample heterogeneity and research objectives, recommending six to eight interviews for a homogeneous sample and twelve to twenty data sources "when looking for disconfirming evidence or trying to achieve maximum variation". Twenty seven interviewees is well in excess of his recommended number and confirms that saturation is likely to have been reached (Kuzel, 1992).

Multiple sclerosis occurs more commonly in women than men however the sampling frame was not representative of that. It was difficult to recruit more females from the pool of patients available, as many of them tended not to have paid employment outside the home. It was extremely difficult to find a female with progressive disease in a "blue-collar" job as these are the most significant demographic features cited for job loss and this is reflected by the fact that only one person was recruited to that particular cell.

**Table 5.1 The Sampling Frame**

| | | Male | Female |
|---|---|---|---|
| White Collar Worker | Progressive Disease | 4 | 3 |
| | Non Progressive Disease | 5 | 5 |
| Blue Collar Worker | Progressive disease | 3 | 1 |
| | Non Progressive Disease | 3 | 3 |

## 5.3 Analysis of the Qualitative Interview Data

A grounded theory approach was used to identify the main "themes" that arose when studying the manuscripts. Content analysis was employed for identifying the common recurring items that formulated the scale. Both approaches were constrained to the experience of work.

### 5.3.1 Qualitative analysis of the transcripts

Four main areas emerged for discussion as factors contributing to work instability on qualitative analysis of the data. Quotes have been taken from the transcripts to illustrate the individual problems that combine to form the aforementioned areas.

**1. Physical and Cognitive aspects of the disease and their impact upon work**

People with MS felt that their physical symptoms did adversely affect their working life. Particular symptoms arose frequently specifically tiredness, fatigue, pain, weakness, visual problems, cognitive deficits, walking and balance:

Fatigue

*"And I'll drop her (his daughter) off at nursery, go to work, work not constantly but pretty intensely for about three and a half hours up until I've had lunch because I know once I've had lunch, I'll, my, it'll deteriorate will my quality of work for the simple reason that fatigue will set in and I seem to get really tired after I've eaten"*

(White collar worker, non - progressive disease, male)

*"Because, they (his managers) feel that I'm - I get very tired. You see the hardest part is, ok I get up at five o'clock in the morning and like anyone I'm tired at five in the morning but you, you're used to, I'm used to that, but it's after being at work for six, hours, I really am physically drained, whether I'm sat down all day I'm still drained because that's the nature of the illness isn't it with the MS"* (Blue collar worker, progressive disease, male)

*"So roundabout this time of the day, you know, after one o'clock and up to four o'clock occasionally I will drop off while I'm working at my computer, you know actually go to sleep without realizing it and wake up five minutes later slumped over you know"* (Blue collar worker, progressive disease, male)

*"I mean I've only been diagnosed since February but I knew for a year before that, that something was going on and I suppose for a couple of years the fatigue has affected me an awful lot without realising what it was"* (White collar worker, non – progressive disease, female)

*"I just start feeling a bit, I don't know if it's tired or fatigue. I'm alright in a morning it's just as the day goes on I start feeling it. Normally eight till one o' clock and I'm fine, one o' clock onwards I'm starting to get where, "Oh god! Is it time for home yet?"* (Blue collar worker, progressive disease, male)

Pain

*"I actually had a major problem with my hands and I still have it now, I can't feel them and I have a lot of pain, neuropathic and nerve pain in my hands so much that*

*holding anything for any length of time ...is actually excruciatingly painful"* (White collar worker, non – progressive disease, female)

*"The clumsiness in my hands I've learnt to deal with but the extreme discomfort while walking and standing is something that is very difficult to stand and you know well cope with"* (Blue collar worker, progressive disease, male)

*"I do try to keep up with most things myself but because the lack of use of my left hand and pain – I suffer with a great deal of pain in my left arm and my knees and I suppose in some ways you get to the point where you actually have an ignorance of the pain and try to ignore it, so you know that it is there but it's become part of your life and you just have to accept it"* (White collar worker, progressive disease, male)

### Hand Function

*"My hands don't work very well, some days my writing comes out like a line. Things like typing I find quite hard because my left hand doesn't work properly, the keys are so little my fingers miss"* (White collar worker, non – progressive disease, female)

### Balance and walking

*"My legs go, it's weakness and extreme fatigue and I feel as though I'm walking like a duck, wobbling around, I mean if I can sit down between, I can do short distances and then sit down and recover for five/ten minutes, I can get up, I can walk again as long as I can sit down again"* (White collar worker, progressive disease, female)

*"..I tend to be unsteady therefore I can trip over things very easily, that's my biggest problem. That is where it has effected my job, because of my lack of balance I quite often bang into things when I'm unsteady, it's, its obviously not too bad if I'm sat at a computer"* (White collar worker, non – progressive disease, female)

*"When he gets very tired his walking goes, he stumbles a lot and his legs give"* (Partner of male white collar worker, progressive disease)

*"Yesterday was my long day, I am very, very tired and it does affect my legs, my legs feel a lot heavier, they feel numb, more numb than they normally do and walking can be a problem, I look as though I've had a few! I'm not as steady as I am when I am not tired"* (Blue collar worker, non - progressive disease, female)

Vision

*"If I seem to get hot, it affects my balance and my eyesight, and then my right, my right eye ends up sort of getting, I get blurred vision in my right eye and my left eye tries to correct for it so I end up with a sort of big wooly picture (laugh) I can still see things but I can't focus on them so I have to sit down for a few minutes and sort of come round a bit"* (Blue collar worker, non -progressive disease, male)

*"They took my licence away because I had lost eyesight, I had a big relapse where I couldn't see things at all , I thought I was going blind, I was petrified"* (White collar worker, progressive disease, male)

*"If I come home and I have had a really stressful day I will get pain in my eyes, from the last attack of MS, and the only way I will get rid of it is to sleep"* (White collar worker, non -progressive disease, female)

Speech

*"I think slow, very slow, my speech is affected. I slur, it sounds like I'm drunk. It takes me, it takes me a long time to get worked out, I know what I want to say but it's getting it to come out. Sometimes customers wonder what's happening; most people just are fine, they've no problem. Yeah, yeah. I had one occasion, there was two or three days where I couldn't talk on the telephone and I would tell the customer to, what the problem was and that if they were quite happy about it then use the phone themselves. No, it was during a period of relapse in, in, in January 2002 and then after a few days of rest I was fine"* (White collar worker, progressive disease, male)

*"I'm aware that nobody has commented to me, but I'm bothered about my speech"* (White collar worker, non - progressive disease, female)

Concentration / memory

Areas specifically discussed were the reduced ability to remember details and the slower pace of thinking since the onset of disease.

*"I use to have a great head for numbers could remember any phone number anyone put at me ...yeah I used to have, I mean I was very, very, I don't know I just had a good head for remembering things and things like that but I do get a bit forgetful I*

*mean I've forgotten I've turned up to the hospital for appointments the day after it should have been and I've convinced myself it was on this particular day and I do get a bit stressed with it sometimes"* (White collar worker, progressive disease, female)

*"I've gone off to one meeting and forgotten I should be somewhere else which, which does happen you know"* (White collar worker, progressive disease, male)

*"When I'm on the phone sometimes my head doesn't quite get round what they've – I mean I know what they're saying but then writing it down doesn't come out right especially with things like dates and numbers, telephone numbers, e-mails and things I find I really have to concentrate on those and again sometimes my, when I'm saying things the wrong word will come out or whatever occasionally, and I'll suddenly forget what I'm saying so you know it is a big pause or whatever"* (White collar worker, non - progressive disease, female)

### 2. Working environment and Organisational aspects

Working life in general was included under the heading "Working Environment". Interesting themes emerged from the analysis relating to how it was felt that stress and pace of work could aggravate the illness and worsen pre – existing symptoms. Participants felt they needed more time to complete tasks due to the direct effects of the disease and could no longer multi-task as this led to feelings of being "overloaded". The importance of working at one's own pace was highlighted by one of the interviewees as being a positive factor in job retention.

### Stress and pace of working practices aggravating symptoms

*"I mean I suppose it's more of a balancing act having something like MS that, because you know that if, if you do you know start to get that snowed under feeling then you know that's the beginning of you know knock on effects for your condition I think you know"* (White collar worker, non - progressive disease, male)

*"If I've had a bad day at work ...and I'm working trying to get you know and it will start to stress me at some point because my body starts to fail you know 'Oh God come on get it sorted' I've got to get out of here because I know that I'm going to like, seize and I'm not going to be able to do owt and I'm like frustrated and it's going to make me even worse...Strange it's the pressure that it puts on your body ; you've no control over it you know"* (Blue collar worker, progressive disease, male)

*"Some days, I mean like this week I've felt really, really well, I've felt sort of normal and you know been quite on the ball and not, you know, felt okay. But other days its just like my whole head is confusion really at times and you put the phone down and you think, or even on the phone particularly "Don't cry" you know just don't, don't know what's happening and don't know what to do and other people talking and you know people on the phone and you can't take it all in at once, your brain's sort of trying to sort out what you're trying to listen to. So that's quite hard. But sort of on the other side of the office there's a music department and they sometimes have music on which sometimes is fine, other times if its like really the wrong sort of music for the wrong day I do ask to turn it off and they're quite, quite okay with that, so and they*

*don't often have it on now anyway"* (White collar worker, non - progressive disease, female)

Importance of working at ones own pace

*"But I'm able to take it (work) at my own steam, I don't think I'd have lasted if they weren't so, they're very team orientated, excellent to work for. Being able to work with people who have patience because you do slow down in everything you do. And you, I feel, I feel you have to slow down and so you can do things and do things sort of firmly, be determined, do things slowly and don't do as much as you used to and not, don't do things as much but do them better even though you're probably not doing them better"* (White collar worker, progressive disease, male)

*"You've got to be able to sort of be able to spread work out, not just sort of say "Right, well I'll squash this all into one day and do nothing for the next" you can't do it like that, you've got to be able to spread the work and actually, rather than actually try and condense everything as, and cause yourself more problems"* (White collar worker, progressive disease, male)

The extent of managerial support and flexibility regarding reduction of hours and place of work emerged as themes as did practical aspects, such as, access to buildings, parking, the temperature and layout of the workplace. They interviewees felt they needed larger workspaces to accommodate their impairments.

### Stairs

*"No I must admit it is getting a bit of a problem now, I tend to spend more time downstairs than upstairs now because it can be quite tiring and it can be quite frustrating – if I'm taking somebody up to my office I'll be dragging my leg upstairs you know, but thinking 'but I'll get there' you know"* (Blue collar worker, progressive disease, *male.*

*"I'm struggling, I've struggled here with my steps, had to really like lift my right leg you see, it just felt it was not really doing what it should be doing"* (Blue collar worker, non - progressive disease, female)

### The Building

*"Just things like you know, the physical building itself you know, that's [pauses, thinking] things like that are quite, are quite hard and it makes you feel more different to other people you know like I've had, they actually now changed the door knobs to handles for me which is good because I couldn't do the door knobs and "Oh we should have done this if you're disability or whatever, you know should have done that quite a few years ago, yeah thanks for telling us" and everybody said "Oh, gosh they've re-done the door knobs, isn't that wonderful" and I haven't said, 'well you know its because of me.'*

*"So things like that, you know when and when the lifts not working you have to go up the stairs and stuff and it just, it just makes you feel different, it makes you feel, remember that you've got the MS you know and that doesn't seem a big thing to people who haven't got any disability or haven't, you know, who are, who are quite active and whatever, it doesn't you know, matter to them but I notice, you know a few*

*little things do – I mean like the hand rails aren't in the right places and stuff and you know, its like coming down the steps out of the town hall I feel I'm going to fall all the time, you know, you can hold on to a certain extent and then you have to let go and its sort of "Oh my God!".*

*"So little things, just little things I feel could, could be improved. Things like having a proper rest area would really, really help, [pauses, thinking] somewhere to get away because like a lot of the time I spend lunch times at my desk because I can only go out for so long without getting tired out walking round, I don't want to go to the shops and things you know so, so you're sat at your desk a lot of the time and then the phone's always ringing and things"* (White collar worker, non - progressive disease, female).

*"It is, it is beginning to affect it now, more than, since I first incurred the illness, shall we say, I seemed to get by reasonably well, but over the last, perhaps, eight months or so, it is becoming noticeable, it's getting more difficult to do my job at work. It's, it's the [slightly laughs] the twenty five percent of the job that's the most difficult, because it means walk – walking it's like round the factory, I also used to have to go upstairs in the fac-[stops mid sentence] in the workplace, we don't have a lift to a certain building that we work in, but now the management have actually stopped me doing that because of my, I'm so unsteady on my feet and I've fallen a few times so they won't allow me to go up stairs because they're frightened if I fall down the stairs which is, which is fair – and also walking round the factory floor now, I've been told not to do so much of that because of the forklift trucks, the pallets, etc I tend to be unsteady therefore I can trip over things very easily, that is my biggest problem. That is where it has affected my job, because of my lack of balance, I quite often bang*

*into things when I'm unsteady, it's, it's not obviously too bad if I'm sat at a computer"* (Blue collar worker, progressive disease, male).

### Working from Home

*"...working from home there and that sort of thing which I think is an option that should be open more than it is as well .....you could you know fit it in when you felt like doing it, you could work in the evening if you, if it was you know more appropriate to do that and you could move things round to your own time scales rather than to fit in with work ones"* (White collar worker, progressive disease, male)

### Space to manoeuvre

*"Now at the moment it's so small, so cramped, we're bumping into each other; it's just, yuck, it's difficult. The workroom is the kitchen which is the smallest place you've ever seen and its full of red boxes and it just brought it all home to me because I had to go in there and deal with some books on cassette and there's boxes everywhere and I couldn't move and I was, because my balance isn't, I'm just not quite as good as I should be, and it was difficult because I was walking into everything and trying to turn so I wasn't banging into that then banging into something at the back of me [laughing] and I'm thinking "Aggh!"* (Blue collar worker, non - progressive disease, female)

### Computer as a valuable resource

*"It's hard to say, it, it, it's difficult to explain the type of work I do but, they try – I must – they do try and put more computer work my way, if they can, and they have talked to the other people because again it means that if they've, the err colleague of*

*mine who's on the opposite shift to me, if they take the computer work from him, that means he has to do more of the manual work because that's the flow of work. It doesn't come in so, it's difficult for them to assess how it's going to come in, the workload, but they do make an effort to put work, more computer work my way. So I suppose yes, as somebody who now is finally admitting to having some disability, which I've found very difficult to admit to, sitting down at a computers probably been a life saver really*" (Blue collar worker, progressive disease, male)

Managerial response to illness

*"The employers are understanding, as I say because they have stopped me walk- [stops mid sentence] going up stairs for instance. They're understanding that I always, I always feel a little bit under pressure because now I cannot, I'm having to ask other people, we talk about the dyes, that are heavy and need carrying, and the boss my, my immediate management have said to me that, "Get someone else to take them for you"* (Blue collar worker, progressive disease, male).

### 3. Social aspects of the working environment

The themes that were identified included positive relationships with colleagues and the level of support received from them but there were also feelings of guilt and embarrassment that arise from dependency on others. The emphasis placed on personal negation of tasks deterred people from asking for help or delegating tasks. This need for personal negotiation was mainly due to lack of wider structured managerial support.

There were concerns about disclosing their illness and "what others might think", however the need to retain contact with people outside the home was highlighted.

Asking for help/delegating tasks

*"My immediate management have said to me 'get someone else to take them for you' but that puts me under a little bit of pressure because I'm having to ask other people to do things. To be honest no-one minds doing it but psychologically you feel that you're having other people running about"* (Blue collar worker, progressive disease, male)

Concerns around disclosure of illness to colleagues/management (What others might think)

*"I had a nasty relapse and I had no alternative but I had to tell them but once you come out there's not a lot they can do. But it's a problem telling somebody before you start because and the prejudices and for the future I will continue to well, not tell anybody and cope with things if they happen during employment"* (Blue collar worker, progressive disease, male)

*"I sort of told them (employees) pretty much straight away so they didn't think that I was slacking really you knows even though I'm the manager really, it's my company (laughter) but .....if I didn't tell people they wouldn't know, they would probably think I was lazy, you know"* (Blue collar worker, non -progressive disease, male)

### 4. Psychological aspects of working

The importance of work identity was discussed. A formal work identity is related to feeling normal, having a focus and can provide valued social contacts. Some interviewees needed to work in order to fulfill their financial commitments.

The coping strategies used to enable the continuation of a working life were mentioned in every interview although the particular strategies used differed widely. Some implemented very practical measures for adapting to workplace pressures and environment, such as, sitting down for activities. Other coping strategies had a more psychological basis, for example, non - disclosure of the illness and just "getting on with it" or putting on a front but inwardly preparing for worst case scenarios.

Fear and uncertainty about future regarding work emerged as an issue for employed people with MS. This was mediated through the uncertainty of the relapsing and remitting nature of the disease. The fear of job loss, having to change jobs or the disease limiting future career options were of obvious concern to the interviewees.

Importance of a work identity

*"I don't know, I mean I think it's really difficult for people with MS not just to continue to work to make decisions, I m always being badgered by people to claim the disability allowance and I don't want it ...I think you're on the road to letting yourself go down when you start relying on money whats not your own, I've always had to work since being a kid and I think that keeps you going...I don't need anybody being sorry for me, I want to do my job to the best of my ability and it's better I'm working than sat here useless"* (White collar worker, progressive disease, female)

### Work fulfilling financial commitments

*"Interviewer: so in a way your choosing to continue to work has sort of affected your life hasn't it? Interviewee: Because I have no option, because I've no alternative that's why. If I could afford to retire I would, oh yeah there's no doubt about that but I can't you know like a lot of people I just can't afford to stop working"* (White collar worker, non - progressive disease, female)

### Coping Strategies

*"So I suppose I'm lucky with my job and I keep it quiet because I'm lucky with my job – you'll find me repeating myself. It's all I know now is this for the last six years and it's, I know I'm deceiving people but I've got to because I have a mortgage and bills to pay and you know and there's, as long as I can get up on a morning and go to work I will go but the minute somebody finds out I'll get penalized someway or another whether it be I lose my job or whatever you know so"* (Blue collar worker, non - progressive disease, male)

*"I'd like not to work at all really. I mean I seem bright and cheerful to people, they come up they'll say 'you're looking well' but I'm not really. I'm in pain most of the time, I feel exhausted but you've got to put on a front haven't you?"* (White collar worker, non - progressive disease, female)

### Fear and uncertainty regarding the future

*"..Having this as you well know is the extra thing that you don't need but because you've got you've got to adapt around it really and it's not going to get the better of*

*me you know. But I haven't I've often said to people you know I'm not going to wait, sit and find out if I'm going to be in a wheelchair when I'm fifty. If I am I am, I will deal with it you know and whatever way I will deal with it"* (White collar worker, non - progressive disease, male)

*"It's frightening to think about the future, and I do think about it and I don't think about it or I don't want to think about it but I still do and I do not know what I'll be doing in five years time. If I'm still here it'll be brilliant, if I'm not I haven't go a clue where I'll be living or what I'll be doing and that is the frightening bit"* (White collar worker, progressive disease, male)

*"A while ago I was the one who was running up and downstairs and I can't – I can't do it, you know I can't do it so I'm slightly worried that if I can only do a certain part of the job they may, maybe move me to another department but, I'm, I'm - how can I say? - I'm reluctant to move because again the shift pattern situation which we work, I, I enjoy that, I prefer it, if I'm moved into a more administrative role I'd be back on days and, I don't like that and that's longer and harder"* (Blue collar worker, progressive disease, male)

### 5.3.2 Item Selection

On completion of the participant interviews and in conjunction with the qualitative data analysis, the transcriptions were read and coded for item selection. Each transcript was scrutinised by three researchers, the primary researcher and two others who had been involved in the development of other validated work instability scales – the RA-WIS and the TBI- WIS (Gilworth et al, 2003, Gilworth et al, 2006) to identify potential statements for the draft Work Instability questionnaire. The statements identified had to relate only to the impact of MS upon the individuals working life. The exact wording in the form of quotations from the interviews was used for the items in the scale wherever possible.

Items were identified and included if they fulfilled the criteria of relating to working with MS, were expressive of a single idea and applicable to all potential respondents (as discussed in section 3.11.2)

<u>Examples of how individual blocks of text were converted into a statement with a clear dichotomous response when exact quotations were not used:</u>

*"My hands don't work very well. Things like typing I find quite hard because my left hand doesn't work properly"* = My hands don't seem to work properly

*"I have no option, because I've no alternative that's why. If I could afford to retire I would, oh yeah there's no doubt about that but I can't you know like a lot of people I just can't afford to stop working"* = Financially I can't afford to stop working

*"The clumsiness in my hands I've learnt to deal with"*= my hands are clumsy now

*"The extreme discomfort while walking and standing is something that is very difficult, to stand you know, and well cope with"* = standing is very painful

*"Just things like you know, the physical building itself you know, that's [pauses, thinking] things like that are quite, are quite hard"*= the building I work in makes things harder

*"I use to have a great head for numbers could remember any phone number anyone put at me ...yeah I used to have, I mean I was very, very, I don't know I just had a good head for remembering things and things like that but I do get a bit forgetful"* = I cant remember numbers like phone numbers

*"It was difficult because I was walking into everything and trying to turn so if I wasn't banging into that then banging into something at the back of me [laughing] and I'm thinking "Aggh!"* = I bump into things easily

*"If I seem to get too hot at work, it affects my balance and my eyesight"*= My condition is worse if I get too hot at work

*"You've got to be able to sort of be able to spread work out, not just sort of say "Right, well I'll squash this all into one day and do nothing for the next" you can't do it like that, you've got to be able to spread the work and actually, rather than actually try and condense everything as, and cause yourself more problems"* = I need to spread my work out

One hundred and twenty two statements related to working with multiple sclerosis emerged. These were all identified individually by each of the three separate researchers involved in this part of the study. This first draft of the questionnaire consisted of one hundred and twenty two items where were agreed on by the primary researcher and the two other researchers who were familiar with the process of item selection having worked on two previous published studies regarding work instability (Gilworth et al, 2003, Gilworth et al, 2006) (Appendix 9).

### 5.3.3 Cognitive debriefing

These one hundred and twenty two potential items that formulated a draft questionnaire were then read by ten people with multiple sclerosis who were in work. This was to assess how difficult the items were to read and understand and whether or not they conveyed the same meaning to everyone. Exclusion of the items on the basis of a few patient's responses has been discussed in section 3.12, was a qualitative research judgment made by the team, most of whom were highly experienced in this area and examples are outlined below.

Seven women and three men took part in the cognitive debriefing which was representative of the sex ratio of those affected by MS. One man and four women felt that the items read clearly and were easily understood.

However two men and three of the women felt that there were problems with some of the items:

PG (male) felt that item 23 "I flag very quickly" was ambiguous and rather than meaning to lose energy quickly would take the term "to flag" as "to flag up potential problems" and actually be more alert at work which was exactly the opposite of the context it was being used in, in the interview. However the other 9 people involved in the cognitive debrief felt that its meaning was clear and relevant. Therefore it was kept in the scale.

2 women, RG and LT highlighted the same item; item 4 "I need to spread my work out". This statement was felt to be ambiguous in its meaning. They were unsure if it was a physical need, that is, increased space to work or if it was to do with pacing oneself. When attention was then drawn to this item two other participants agreed that the meaning could be unclear and so this item was discarded from the scale.

Two men and three women felt that item 93 "It has reduced my effectiveness to do my job" was long – winded and the meaning of it was not easily interpreted. One person participating in the cognitive debriefs singled this item out as "reading very badly". This item was also discarded from the scale.

These items were not revised to make them more understandable as the participants and research team felt any efforts to do so may have changed the meaning of the item completely. This would defeat the object of item selection which was to try to keep as close to the words and meanings of items derived from the interviews wherever possible.

Therefore, the second draft of the scale contained one hundred and twenty items (Appendix 10). The ambiguous or difficult to understand items had all been identified by the first six people involved in the cognitive debrief and no new concerns arose when the rest of the participants were asked for their opinion confirming that ten participants was an adequate number for this part of the study.

# Chapter 6: Quantitative Research Results –

# Development of the Work Instability Scale

## 6.1 Introduction

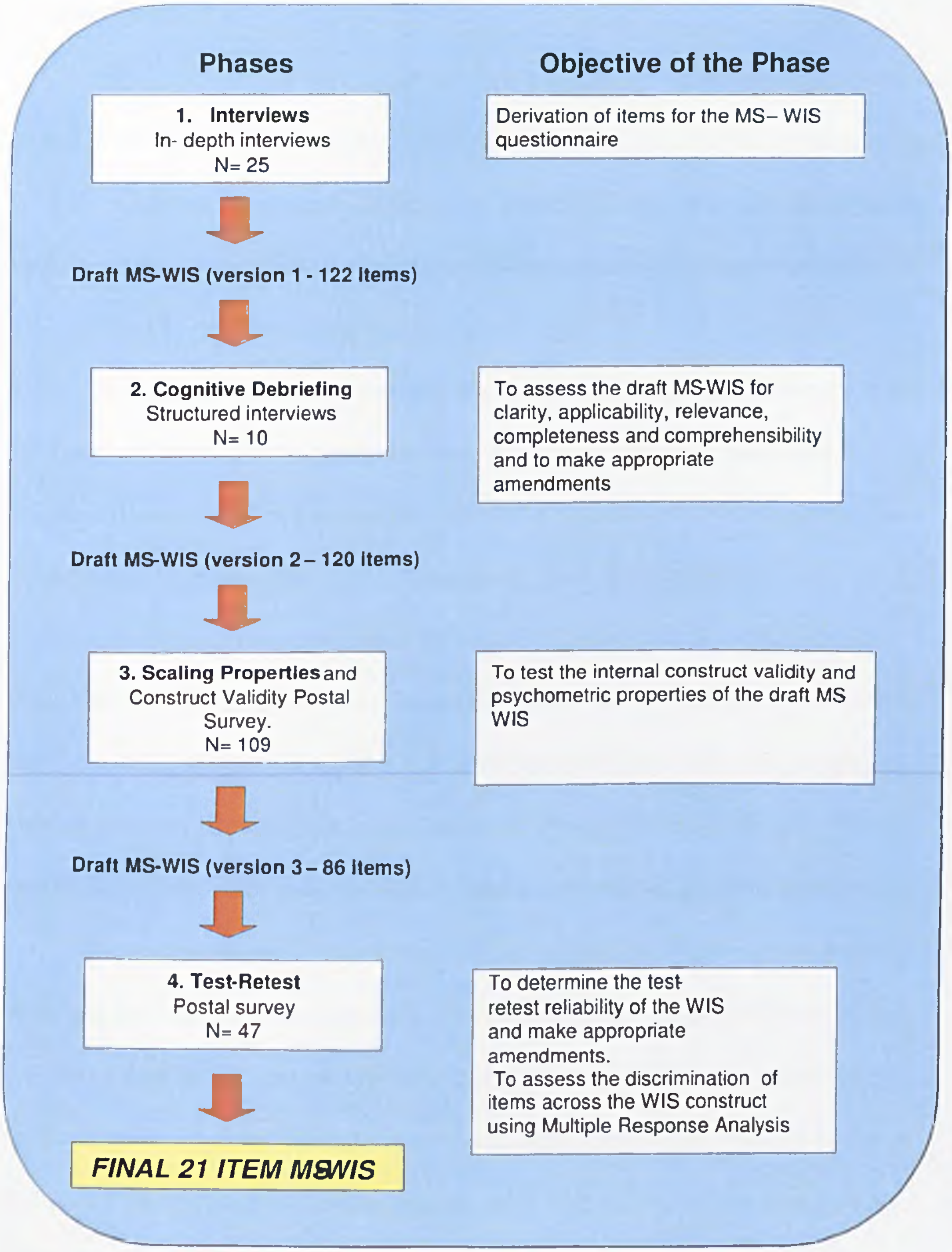
This chapter provides the results of the development of the MS – WIS, the scale that measures work instability in multiple sclerosis. The steps taken to reach this are set out in chronological order. The reasons for discarding items when obeying the rules of the Rasch model and Criterion Validity are clearly displayed and prove that the resulting scale is robust, reliable and valid. It was important to identify the specificity and sensitivity of the scale prior to it being used as this helps clarify any limitations of the scale, the results of which were excellent as the sensitivity of the scale reached 100% at the high – risk threshold with regards to the need for work place intervention.

### 6.1.1 Summary of Scale development.

Qualitative interviews led to identification of themes and potential items which could indicate that a person with multiple sclerosis was a risk of job loss, that is, displaying instability in their work. 120 potential items were then formed into a draft questionnaire which was sent out and the data resulting from this first postal questionnaire and recruitment from hospital clinics was subjected to a number of developmental stages. Using Rasch analysis, items which displayed DIF were identified and discarded, as well as those which displayed misfit to the Rasch model. The initial item reduction phase led to an 86-item WIS. This 86 item questionnaire was then administered to more participants via a postal survey for the test re-test analysis to ensure the scale would behave consistently over time. This scale also underwent further Rasch analysis.

At the same time a small set of vocational assessments were underway which allowed for criterion validity to be carried out. Then based upon the vocational assessment, items which discriminated across expert levels of risk were identified and retained. This reduced item set which consisted of 24 items was subjected to further Rasch analysis, resulting in a 21 item MS-WIS. Rasch analysis was applied to the 21 item WIS using both the initial dataset and the test re-test dataset and the MS-WIS was shown to have good individual level reliability and adequate sensitivity for an initial screen for work instability, and excellent sensitivity and specificity for identifying those at high risk of job loss. A summary of the developmental process is given in Figure 6.1.

Figure 6.1 Summary of the Project Phases for the Development of the MS-WIS



## 6.2 Rasch analysis applied to the draft 120 item MS-WIS in the first postal questionnaire

Internal Construct Validity of the scale was carried out by subjecting the scale to Rasch Analysis. The aim of this was to formally test the properties of the scale and for item reduction. The process of Internal Construct Validity was adhered to which provided detailed information about how the Work Instability scale for multiple sclerosis was performing during its construction.

A dichotomous scale (answering yes or no) was used for the purposes of this study rather than a Likert scale (answering, for example, always/ sometimes / occasionally/ never). The possible limitation of using a dichotomous model is that some important information may be lost through lack of responses along the distribution of the measurement scale or "ruler". However the Item Map (fig 6.7) clearly shows that there is reasonable distribution of the person-item threshold. This may have been enhanced if a Likert scale had been used but it was felt that for this type of questionnaire a definite response was needed by the participants in order to make the result relevant in terms of being able to offer some clinical intervention.

Measurements were obtained on the basis of two class intervals as all subjects were grouped into two "class intervals" or "ability groups". Everyone in the sample was listed in order in terms of their overall ability location estimates which was based on the sum score and then were split up into approximately equal size groups by RUMM. Two class intervals were necessary as it is desirable for Rasch modelling to have approximately fifty subjects in each group for the tests of fit and for groups to be of equal size or as near in size as possible. The number of class intervals is purely a function of sample size and is geared towards providing the correct number of cases in each cell for a chi squared analysis. It does not affect the interpretation of the items within the scale.

### 6.2.1 Response rate and sample characteristics

Initially 350 questionnaires were sent out in the first round. The response rate was 34% as 120 people returned them. Of these 120 people, 41 were working (33.3%). These numbers were too small to conduct significant analysis upon and so, after four weeks, a second round of questionnaires was posted as a reminder to the 230 people who did not return them initially. There were 83 replies (a response rate of 36%) of these 25 people were in work (30%). The total response rate after a reminder was 58% (120 + 83/ 350 x 100) and the percentage of people in work was 33% (41 + 25/ 203 x 100). These figures were completely in keeping with the findings from the literature as previously discussed.

There were still not enough questionnaires from those in work to allow for statistical analysis, therefore people under the care of the St James University Hospital MS team, not yet on the database but, known to be working, were recruited. The limitations of this are discussed in chapter seven. Another 43 questionnaires were completed by people who were in employment when they attended clinic bringing the total to 109 which was then deemed a suitable number to allow statistical analysis to begin as it satisfied the basic sample size requirement for Rasch analysis

40% of the patient sample were in the age group 18 – 40 years, the other 60% were aged between 41 – 65 and it was made up of 70 females (64%) and 39 men (46%). There was missing data for type of job but the available data showed 19% were in blue collar work, and 55% worked full time. 85% were not reliant on a wheelchair at all. Those out of work were significantly older than those in work. The mean age of the 'in work' sample was 42.9 ± 8.31 years, compared to 52.53 ± 8.74 for the 'out of work' sample (t = -8.42, df = 221, p = 0.00).

Data from these 109 responses from those in work (i.e. those who were able to respond to the draft work instability items) were then fitted to the Rasch measurement model. The aim of this was to formally test the measurement properties of the scale and for item reduction. However only one hundred and eight responses could be analysed as one of the respondents was considered "extreme". That is, the individual answered in the affirmative to all items and so were at the ceiling of the scale, thus exceeding the range of the measurement scale which precluded their inclusion in the Rasch analysis. Thus this indicates that in practice the draft scale actually was well targeted, as less than one-percent were at either the floor or ceiling of the scale.

Although that excluded participant's data could not be included in the estimation basis of the item difficulties of the scale, an estimate of their work instability could still be obtained through an extrapolation procedure within the Rasch analysis software.

### 6.2.2 Internal Construct Validity

*Summary Fit for the 120 – Item Scale*

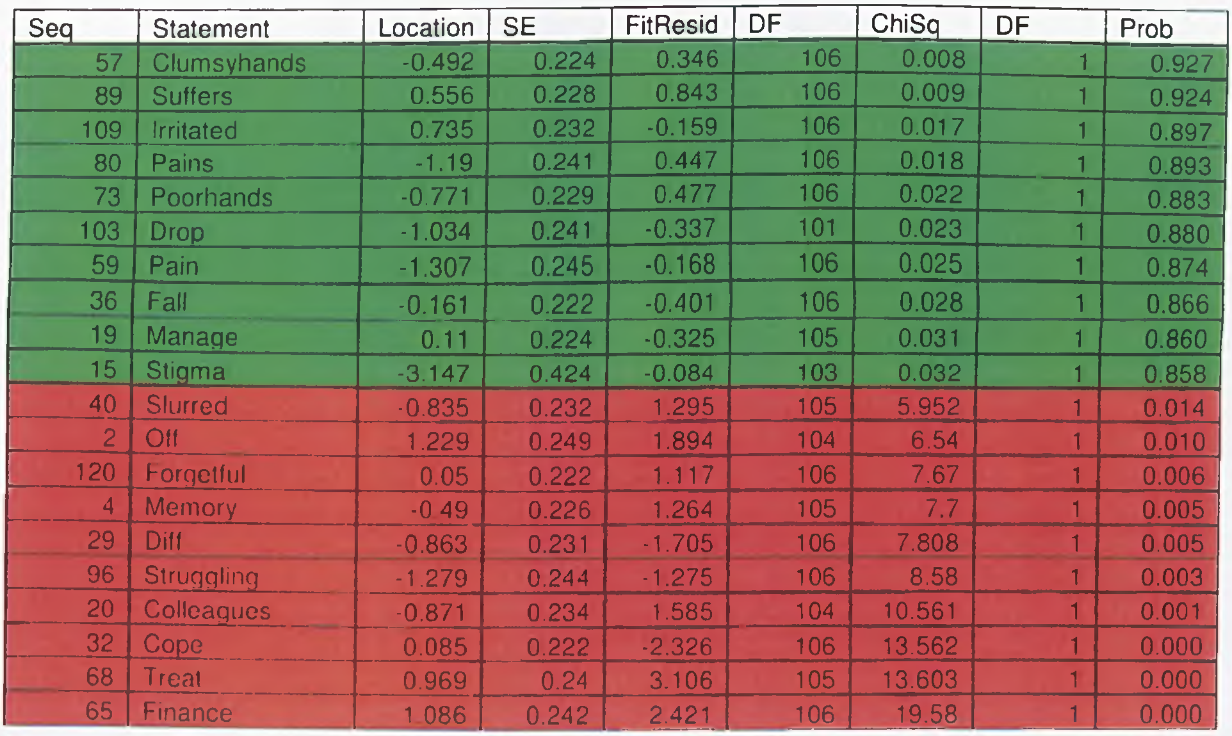
The overall fit statistics provided by the Rumm2020 package determined how well each item contributed to defining the single dimension of work instability for the 120 item scale. Overall fit to the model was poor (Item Fit Mean 0, SD 1) and the Item – Trait Interaction Statistic was significant ( $\chi2$ = 270.815, P = 0.000). However, the Person Separation Index (PSI) which is a measure of reliability and indicates how well the items are differentiating between persons was satisfactory at 0.972. The

Cronbach alpha, which is also a test of reliability, was not applicable as there was missing data.

*Individual Item Fit for the 120 Items*

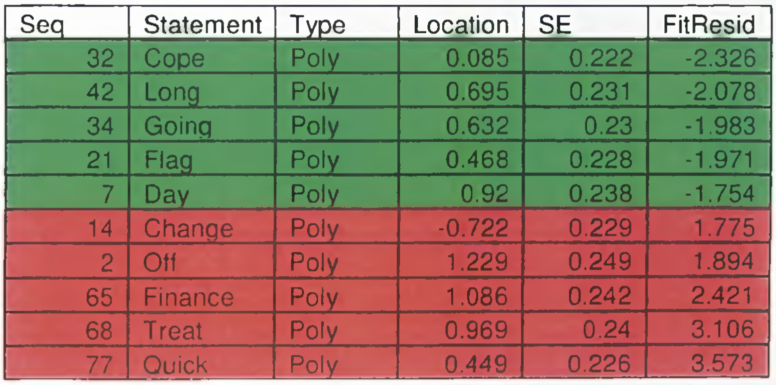
A χ2 probability level of 0.01 was used; this stringent level was applied to ensure the best fit of the items to the scale. Ten of the best and ten of the worst fitting items by χ2 probability are shown in Table 6.1. Overall only two items were displaying a misfitting residual of greater than 2.5, shown in Table 6.2.

**Table 6.1 Individual Item Fit for 120 items by χ2 – The 10 best (green) and worst (red) fitting items out of the 120 item scale**



| Seq | Statement | Location | SE | FitResid | DF | ChiSq | DF | Prob |
|---|---|---|---|---|---|---|---|---|
| 57 | Clumsyhands | -0.492 | 0.224 | 0.346 | 106 | 0.008 | 1 | 0.927 |
| 89 | Suffers | 0.556 | 0.228 | 0.843 | 106 | 0.009 | 1 | 0.924 |
| 109 | Irritated | 0.735 | 0.232 | -0.159 | 106 | 0.017 | 1 | 0.897 |
| 80 | Pains | -1.19 | 0.241 | 0.447 | 106 | 0.018 | 1 | 0.893 |
| 73 | Poorhands | -0.771 | 0.229 | 0.477 | 106 | 0.022 | 1 | 0.883 |
| 103 | Drop | -1.034 | 0.241 | -0.337 | 101 | 0.023 | 1 | 0.880 |
| 59 | Pain | -1.307 | 0.245 | -0.168 | 106 | 0.025 | 1 | 0.874 |
| 36 | Fall | -0.161 | 0.222 | -0.401 | 106 | 0.028 | 1 | 0.866 |
| 19 | Manage | 0.11 | 0.224 | -0.325 | 105 | 0.031 | 1 | 0.860 |
| 15 | Stigma | -3.147 | 0.424 | -0.084 | 103 | 0.032 | 1 | 0.858 |
| 40 | Slurred | -0.835 | 0.232 | 1.295 | 105 | 5.952 | 1 | 0.014 |
| 2 | Off | 1.229 | 0.249 | 1.894 | 104 | 6.54 | 1 | 0.010 |
| 120 | Forgetful | 0.05 | 0.222 | 1.117 | 106 | 7.67 | 1 | 0.006 |
| 4 | Memory | -0.49 | 0.226 | 1.264 | 105 | 7.7 | 1 | 0.005 |
| 29 | Diff | -0.863 | 0.231 | -1.705 | 106 | 7.808 | 1 | 0.005 |
| 96 | Struggling | -1.279 | 0.244 | -1.275 | 106 | 8.58 | 1 | 0.003 |
| 20 | Colleagues | -0.871 | 0.234 | 1.585 | 104 | 10.561 | 1 | 0.001 |
| 32 | Cope | 0.085 | 0.222 | -2.326 | 106 | 13.562 | 1 | 0.000 |
| 68 | Treat | 0.969 | 0.24 | 3.106 | 105 | 13.603 | 1 | 0.000 |
| 65 | Finance | 1.086 | 0.242 | 2.421 | 106 | 19.58 | 1 | 0.000 |

**Table 6.2 Individual Item Fit for 120 items by Fit Residual - The 5 highest (red) and lowest (green) fit residuals out of the 120 item scale**



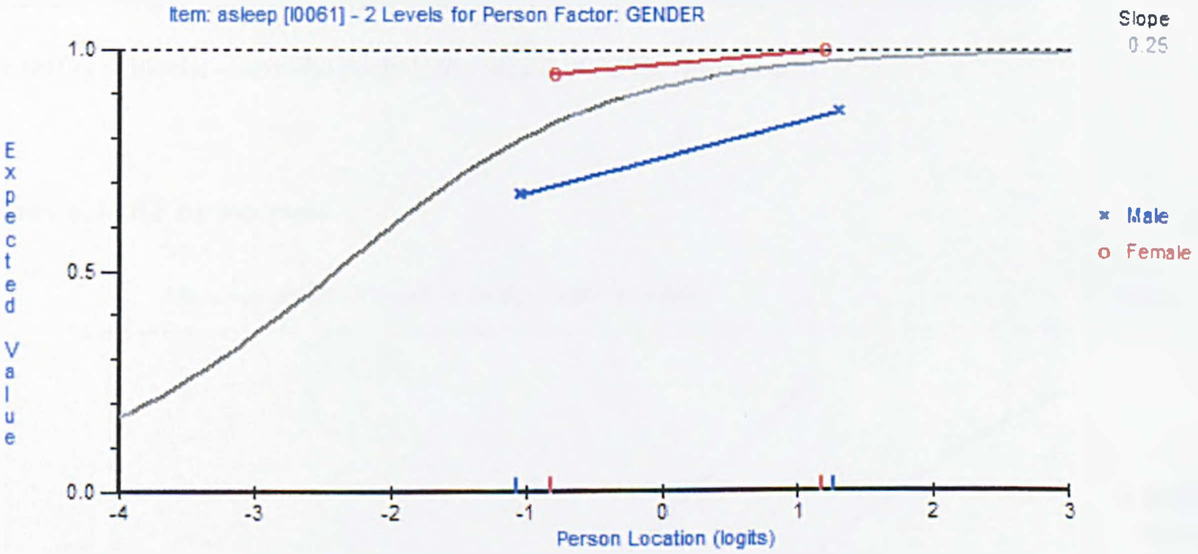
| Seq | Statement | Type | Location | SE | FitResid |
|---|---|---|---|---|---|
| 32 | Cope | Poly | 0.085 | 0.222 | -2.326 |
| 42 | Long | Poly | 0.695 | 0.231 | -2.078 |
| 34 | Going | Poly | 0.632 | 0.23 | -1.983 |
| 21 | Flag | Poly | 0.468 | 0.228 | -1.971 |
| 7 | Day | Poly | 0.92 | 0.238 | -1.754 |
| 14 | Change | Poly | -0.722 | 0.229 | 1.775 |
| 2 | Off | Poly | 1.229 | 0.249 | 1.894 |
| 65 | Finance | Poly | 1.086 | 0.242 | 2.421 |
| 68 | Treat | Poly | 0.969 | 0.24 | 3.106 |
| 77 | Quick | Poly | 0.449 | 0.226 | 3.573 |

*Differential Item Functioning (DIF)*

One possible cause of misfit to model expectations is DIF. Consequently initial item reduction was based on DIF and items were removed until all the remaining items were DIF free at the p=0.005level. This level was used to allow for multiple testing but was still stringent enough to ensure the best fit of items to the scale. All items which displayed a DIF by person factors were removed as the item showed a bias to that distinct group of subjects e.g. age group. In total twenty seven items were removed due to DIF.

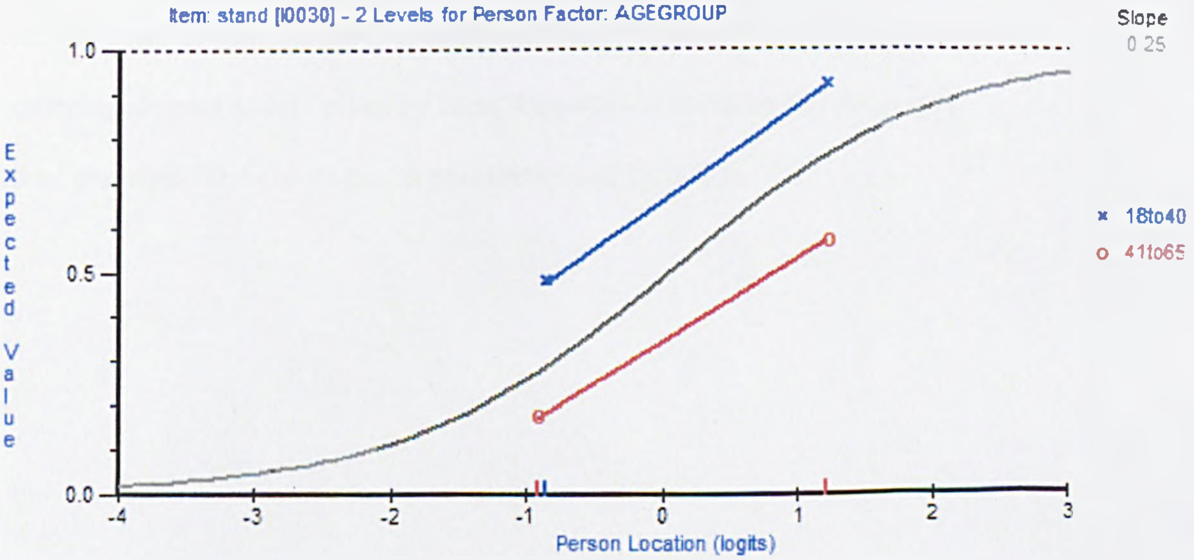
Essentially the scale should work in the same way, irrespective of which group is assessed i.e. whether men or women, or younger and older age groups, however upon analysis items had to be removed due to (DIF). For example, item 61 "I fall asleep at work" displayed DIF by gender meaning men and women were not answering the questions in the same way (Figure 6.1). Females were more likely to affirm this item at any given level of work instability. The chi – squared probability was 0.0029.

Figure 6.2 DIF by gender



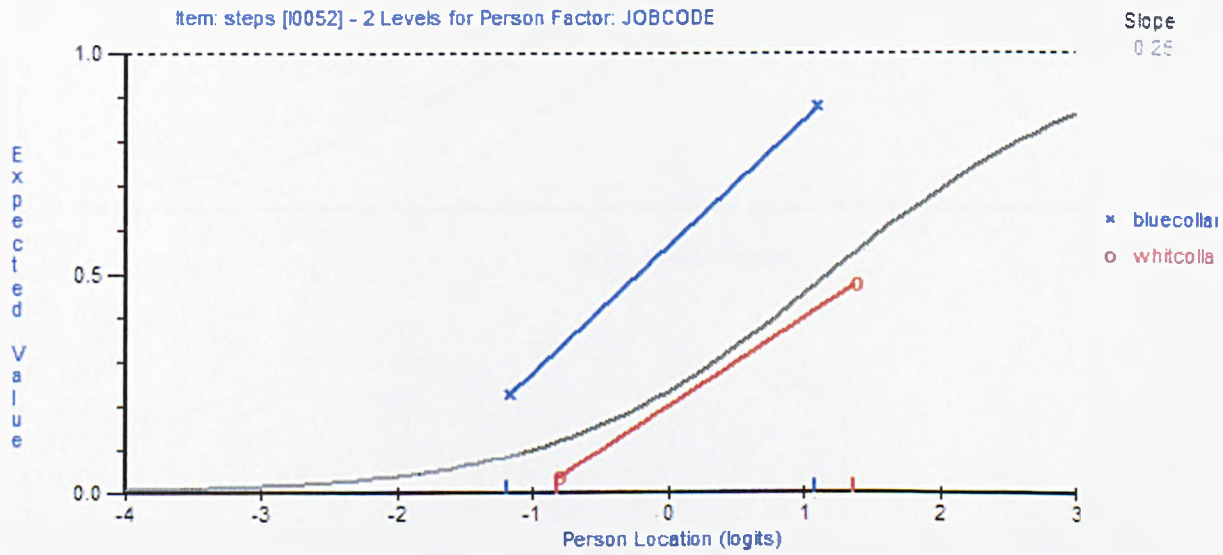
Item 30 “I have trouble standing upright sometimes” displayed DIF by age group, with younger respondents more likely to affirm this item at any given level of work instability (Figure 6.2).

Figure 6.3 DIF by age



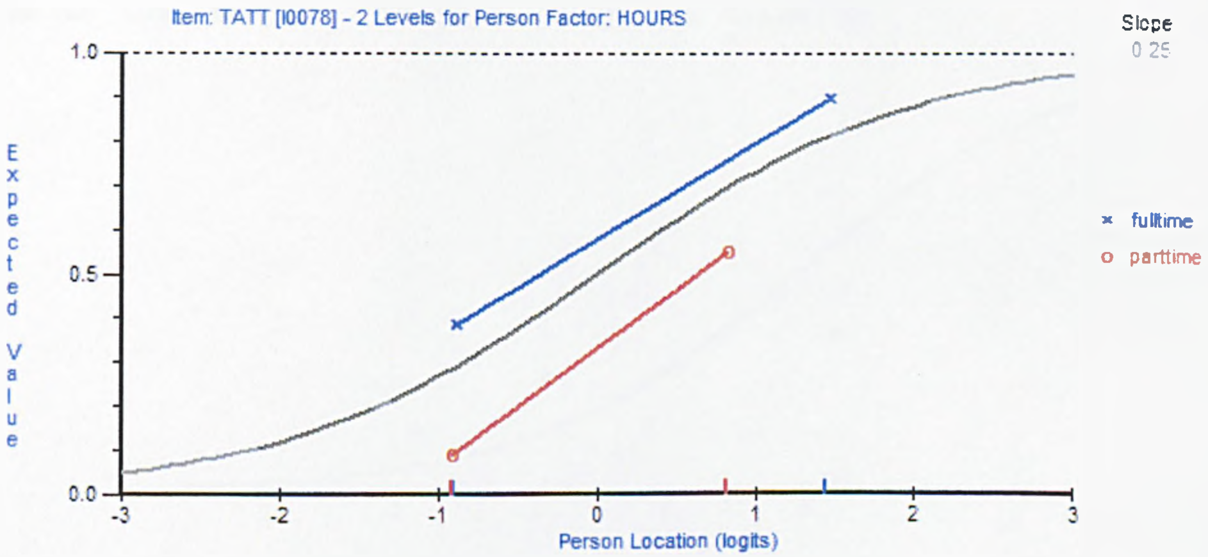
Item 52 "Steps and stairs are a struggle for me" displayed DIF by type of job, with blue collar workers reporting more difficulty with this at any given level of work instability. The chi – squared probability was 0.000713. (Figure 6.3)

**Figure 6.4 DIF by job code**



Item 78 "I am tired all the time" displayed DIF by the amount of hours worked, that is, differing responses were given by those who worked full time and those who worked part time. The chi – squared probability was 0.000268.

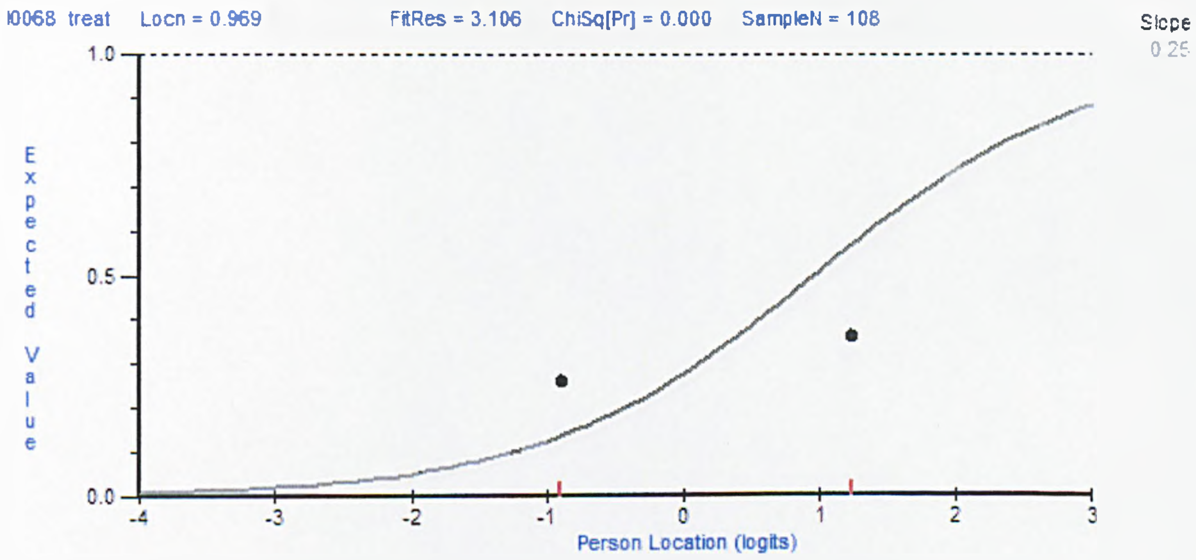
**Figure 6.5 DIF by Hours**



*Misfiting Items*

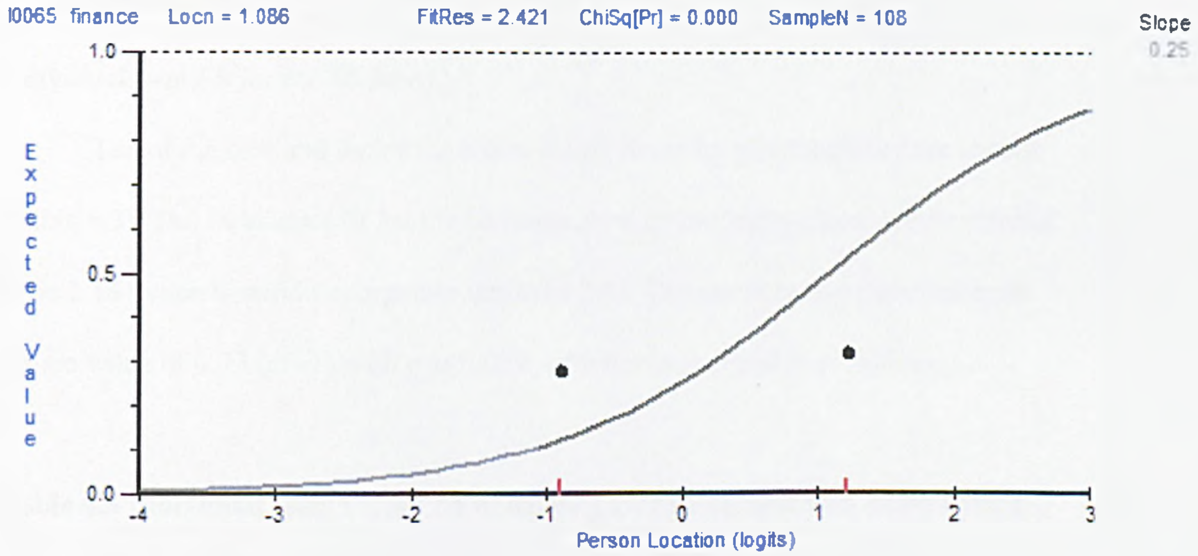
After removal of items displaying DIF, those showing continuing misfit were examined. Item 68 "I don't like it when people treat me differently" revealed a chi – squared probability of 0.000227 and a fit residual of 3.166 (-2.5 to + 2.5). The Item Characteristic Curve showed a lack of discrimination between different levels of work instability (Figure 6.5). Misfit of this item indicated a lack of the expected probabilistic relationship between the item and other items on the scale. In other words, this item was likely to be affirmed irrespective of the level of work instability and thus it displayed a lack of the expected probabilistic relationship between the item and other items on the scale (summed to provide the latent estimate of work instability), and was therefore removed from the scale.

**Figure 6.6 Misfit of Item 68**



Item 65 “Financially, I can’t afford to stop working” was also removed from the scale due to misfit. The chi – squared probability was 0.000011 and the fit residual was 2.421 and the flat nature of the points across the person location on the Item Characteristic Curve showed little change in relative difficulty of the item across the sample (Figure 6.6). This misfit indicted that the item did not contribute to the trait under consideration.

Figure 6.7 Misfit of Item 65



Consequently, after these procedures, twenty five items were removed on the basis of DIF factors and nine items showed misfit to the model expectations, making 34 item deletions in total at this stage. Therefore the Multiple Sclerosis Work Instability Scale was reduced from 120 items to 86 items as a result of the initial Rasch Analysis. The overall fit statistics provided by the Rumm2020 package determined how well each item contributed to defining the single dimension of work instability for the new 86 item scale. The summary fit for the 86 – item scale at this stage the overall Chi – square item trait interaction statistic showed good fit to the Rasch model with a non – significant Chi – square probability ( $\chi2$ = 104.513, $\chi2$ probability = 0.085). Person Separation Index reliability remained good at 0.966. This 86 item scale was then carried forward to the test-retest questionnaire, and the criterion related discrimination analysis.

## 6.3 Rasch analysis applied to the draft 86 item MS-WIS in the second postal Questionnaire

*Individual Item Fit for the 86 Items*

Ten of the best and ten of the worst fitting items by χ2 probability are shown (Table 6.3). The individual fit for the 86 items showed the highest positive fit residual to be 2.18 which is within acceptable limits (± 2.5). The worst fitting item had a chi square value of 4.73 (df=1) with p at 0.029, which was acceptable at this stage. .

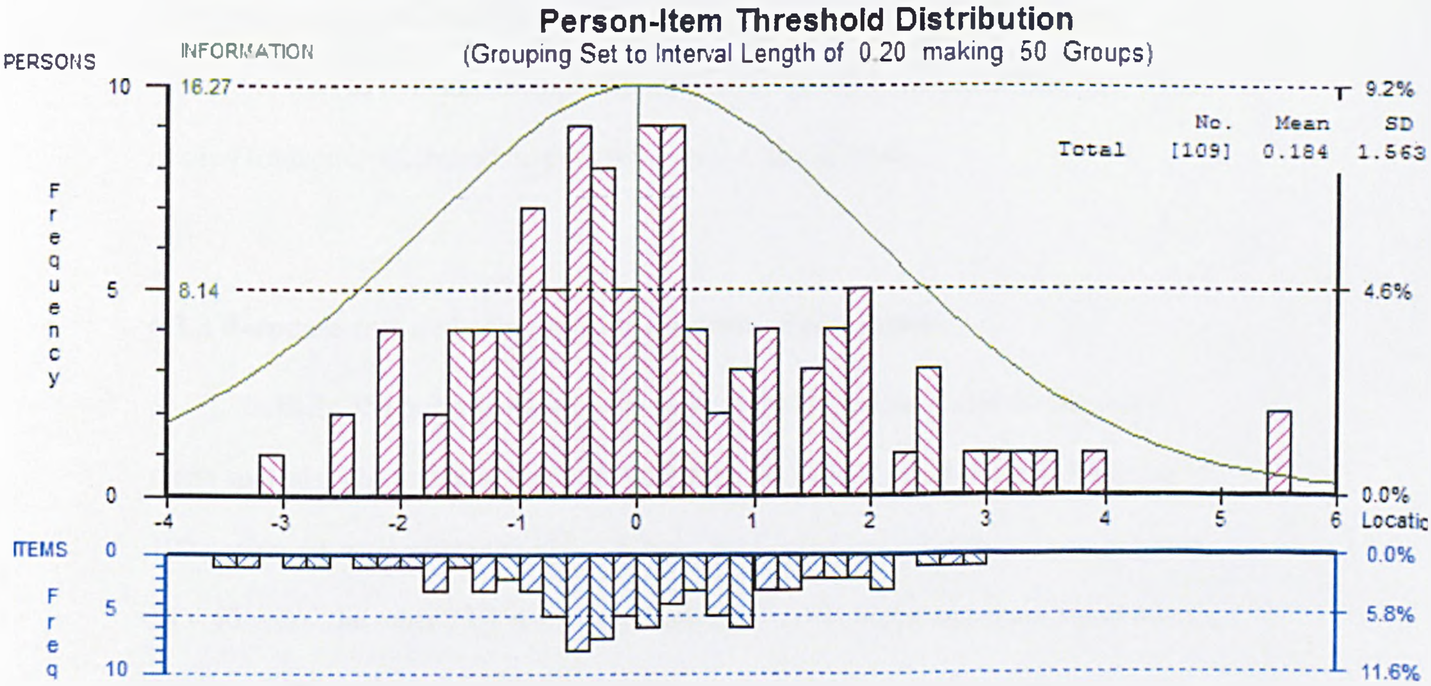
**Table 6.3 Individual Item Fit for 86 items by χ2 - The 10 best and worst fitting items out of the 86 item scale**

| Seq | Statement | Location | SE | FitResid | DF | ChiSq | DF | Prob |
|---|---|---|---|---|---|---|---|---|
| 104 | reach | -0.976 | 0.235 | 0.279 | 104.75 | 0.003 | 1 | 0.956 |
| 84 | temper | 0.431 | 0.228 | -0.241 | 104.75 | 0.014 | 1 | 0.907 |
| 115 | writing | -0.246 | 0.224 | 0.32 | 104.75 | 0.014 | 1 | 0.906 |
| 27 | restrict | -0.436 | 0.225 | -1.345 | 104.75 | 0.02 | 1 | 0.888 |
| 15 | stigma | -3.21 | 0.428 | -0.207 | 101.82 | 0.03 | 1 | 0.862 |
| 25 | back | 0.372 | 0.228 | -0.815 | 103.77 | 0.031 | 1 | 0.860 |
| 86 | mentally | 1.196 | 0.248 | -0.835 | 104.75 | 0.034 | 1 | 0.853 |
| 13 | fut | 0.364 | 0.228 | 1.014 | 103.77 | 0.039 | 1 | 0.843 |
| 55 | trouble | -0.127 | 0.224 | 0.174 | 104.75 | 0.054 | 1 | 0.816 |
| 106 | strong | 1.684 | 0.269 | -1.052 | 104.75 | 0.081 | 1 | 0.775 |
| 26 | earns | -0.615 | 0.227 | 0.778 | 104.75 | 3.38 | 1 | 0.066 |
| 37 | eyes | -0.47 | 0.228 | 1.346 | 102.8 | 3.52 | 1 | 0.060 |
| 6 | energ | 0.672 | 0.234 | -1.082 | 103.77 | 3.715 | 1 | 0.053 |
| 31 | cant | -0.208 | 0.224 | 0.334 | 104.75 | 3.834 | 1 | 0.050 |
| 114 | reliant | -0.39 | 0.225 | 0.965 | 104.75 | 4.121 | 1 | 0.042 |
| 40 | slurred | -0.879 | 0.234 | 1.765 | 103.77 | 4.511 | 1 | 0.034 |
| 44 | bladder | 0.091 | 0.226 | 2.18 | 103.77 | 4.534 | 1 | 0.033 |
| 116 | moretired | 2.842 | 0.349 | -1.35 | 104.75 | 4.538 | 1 | 0.033 |
| 96 | struggling | -1.304 | 0.245 | -1.397 | 104.75 | 4.671 | 1 | 0.030 |
| 110 | unpred | 2.425 | 0.315 | -1.251 | 104.75 | 4.736 | 1 | 0.029 |

*Person and Item Location*

The item map below (figure 6.7) shows how well the 86 work instability items are targeting the persons. Generally the items seem to be targeting the sample well. The average item and person difficulty estimates show a difference of 0.184; this means that the instrument is well targeted, with only 2 patients (~ 2%) at the ceiling of the scale. These "extreme" patients are in the "danger zone" for work instability as they are scoring so highly on the scale.

**Figure 6.8 Item Map**



### 6.3.1 Reliability: Test – retest analysis

The revised scale with the reduced number of items (86) was administered to a further sample of participants on two occasions, two weeks apart to test reliability of the questionnaire over time. The replies to the questionnaire given two weeks apart were subjected to Rasch analysis to ensure that the scale was consistent over time. Statistical analysis began when forty - nine people had completed two questionnaires two weeks apart giving a sample size of 98. The Test-Retest analysis was carried out on 49 persons, repeated over 2 time points, meaning that 98 records were entered into the analysis. 2 cases were removed from the analysis as data was missing meaning N= 47 or 94 cases for analysis. The number of Class Intervals within RUMM was reduced to 2 so as to maintain approximately equal group sizes and to keep the desired frequency of around 50 persons in each Class Interval.

### 6.3.2 Response rate and sample characteristics: Test - retest

Initially 355 questionnaires were sent out in the second round for the test – retest analysis. The response rate was 53.5% as 190 people returned them. Of these 190 people, 61 were working (32%). 27% of the patient sample were in the age group 18 – 40 years, the other 73% were aged between 41 – 65 and it was made up of 43 females (70%) and 18 men (30%). There was missing data for type of job in 9 of the 61 responses but the available data showed 19% held down blue collar work and 48% worked full time. A repeat questionnaire was distributed to the 61 people who were in work two weeks later and of these 49 returned a second completed questionnaire, a response rate of 80%.None of the 49 people who completed the scale twice had relapsed or undergone a significant change in their condition over the two week period, this was specifically enquired about in the covering letter.

### 6.4 Discrimination of Items across the Work Instability Construct

After completion of the second postal questionnaire (the 86 item test – retest survey) further analysis was undertaken of the small set of questionnaires administered at the same time as the vocational assessment. The experts who completed the gold standard assessments identified five levels of work instability, each representing progressively increasing levels of work disability and it was important to identify which item sets discriminated across the gold standards.

The five categories were compressed to three categories for the purposes of analysis and subjected to a multiple response analysis to identify the degree of discrimination of items across each level of risk. The three categories were low, medium and high risk of work instability. More categories would have given more information regarding the discrimination of items. However the collapse to three categories was necessary due to sample size and was also more acceptable as this number of categories number had been used successfully in other work instability scales, such as the RA-WIS and the TBI – WIS (Gilworth et al, 2003, Gilworth et al, 2006). Cross tabulations were created and the frequency of affirmations was assessed (the % of persons saying "true" to an item). Within each level of risk, initially it was proposed that the percentage of affirmations of risk would increase by 15% for each category. When the 15% rule was applied thirteen items from the potential 86 item scale were found to discriminate across the three categories:

1. *I push myself to keep working*
2. *I don't have as much energy to do my job as I used to*
3. *As the day goes on my condition gets worse*
4. *There are some things I can't do any longer at work*
5. *It takes me longer to do some things at work*
6. *I don't like to ask for help*
7. *Its affecting the feeling in my hands*
8. *My hands are clumsy now*
9. *Sometimes in the afternoon I can get really, really tired*
10. *I push myself to go to work because I don't want to give in to my condition*
11. *I have to be careful not to overdo it at work*
12. *I have to rely on other people for some parts of my job*
13. *My writing is affected*

A further five items were found to discriminate across the three categories when a 12.5% cut off was applied:

14. *I have thought about changing my job*
15. *People treat me differently*
16. *I'd like another job but I am restricted to what I can do*
17. *Its painful walking*
18. *I get double vision*

Two further items discriminated between the 2 higher levels of risk:

19. *I feel guilty about getting others to help me*
20. *My hands don't seem to work properly*

This 20 item scale was fitted to the Rasch model with data derived from the second postal questionnaire. All items fitted the model well except for item 14 "I have thought about changing my job" was removed as it was found not to be differentiating due to a high fit residual. When this was removed all other 19 items were found to be well fitting, the person separation index (PSI) was 0.873 and the chi – squared probability was 0.31 with an excellent power – of – test – fit. The item map for the 86 item scale was plotted in order to see where the 19 items lay within it (Figure 6.8). These items were highlighted in red. It was noted that there were no polar items in the 19 item scale. Therefore 5 polar items, highlighted in green, were added to the scale in order to ensure full coverage of items for the scale and so that the scale had potential to be used as an outcome measure.

**Figure 6.9 Location Map for 86 Item Scale**

```
----------------------------------------------------------------------------------------
LOCATION PERSONS ITEMS [locations]
------------------------------------------------------------------------------------------
6.0                      |
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                      oo |
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5.0                      |
                         |
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                         |
4.0                      |
                       o |
                         |
                       o |
                       o |
3.0                    o |
                       o | I0116
                         | I0054
                     ooo | I0110
                       o |
2.0                      | I0028 I003  I0107
                   ooooo | I0098 I0074
                    oooo | I0106 I0001
                     ooo | I0087 I0118
                         | I0064 I0085 I0099
1.0                 oooo | I0017 I0046 I0086
                     ooo | I0081 I0094 I0049 I0053 I0075 I0007
                      oo | I0117 I0042 I0006 I0109 I0009
                    oooo | I0084 I0089 I0034
               ooooooooo | I0095 I0013 I0025 I0022
0.0            ooooooooo | I0120 I0019 I0044 I0023 I0051 I0041
                   ooooo | I0012 I0055 I0024 I0033 I0108
                oooooooo | I0114 I0090 I0011 I0100 I0101 I0115 I0031
               ooooooooo | I0068 I0004 I0057 I0066 I0037 I0050 I0027 I0112
                   ooooo | I0014 I0111 I0076 I0058 I0026
-1.0             ooooooo | I0104 I0040 I0073
                    oooo | I0088 I0045
                    oooo | I0105 I0096 I0080
                    oooo | I0010
                      oo | I0016 I0072 I009
-2.0                     | I0067
                    oooo | I0119
                         | I009
                      oo |
                         | I0062
-3.0                     | I006
                       o |
                         | I0015
                         | I000
                         |
-4.0                     |
-------------------------------------------------------------------------------
o = 1 Person
```

There were twelve items that could have been considered polar items as they occupied the upper and lower items of the scale, of these, five were chosen as they best fitted the Rasch model. The selection of the five additional items is detailed below and resulted in a 24 item scale

1. I am more tired than I used to be (Item 116). This was chosen as it is a polar item and so ensured that the full ability range of the sample could be measured.
2. I have to pace myself (Item 39). This item was chosen rather than items 28 or 107 which had the same location on the map as it provided the best fit to the model out of the three items in terms of fit residual and chi – square values.
3. If I don't reduce my hours I may have to give up work (Item 97). This was included as it provided a distinct marker on the "ruler" that is the scale and provided a better fit to the model than item 119.
4. My employers are not supportive (Item 63). Again, this particular item was chosen as it provided a distinct marker on the "ruler" that is the scale and provided a better fit to the model than item 62.
5. My job is physically impossible (Item 8). This item was included for its polarity.

## 6.5 Further item reduction after the initial discrimination analysis: Rasch analysis applied to the 24 item scale

The overall fit of this revised 24 item scale to the Rasch model improved (from the 19 item version) with a PSI was 0.897 but the chi – square probability was still marginally low at p= 0.0424. Most individual items showed acceptable fit to the model (Table 6.4). The internal construct validity of this proposed 24 item scale was then tested by fit to the Rasch model

### *Individual Item Fit for the 24 Items*

The 24 items worked well overall and were contributing to the trait under consideration however the table below containing summary statistics show the scale could be improved upon.

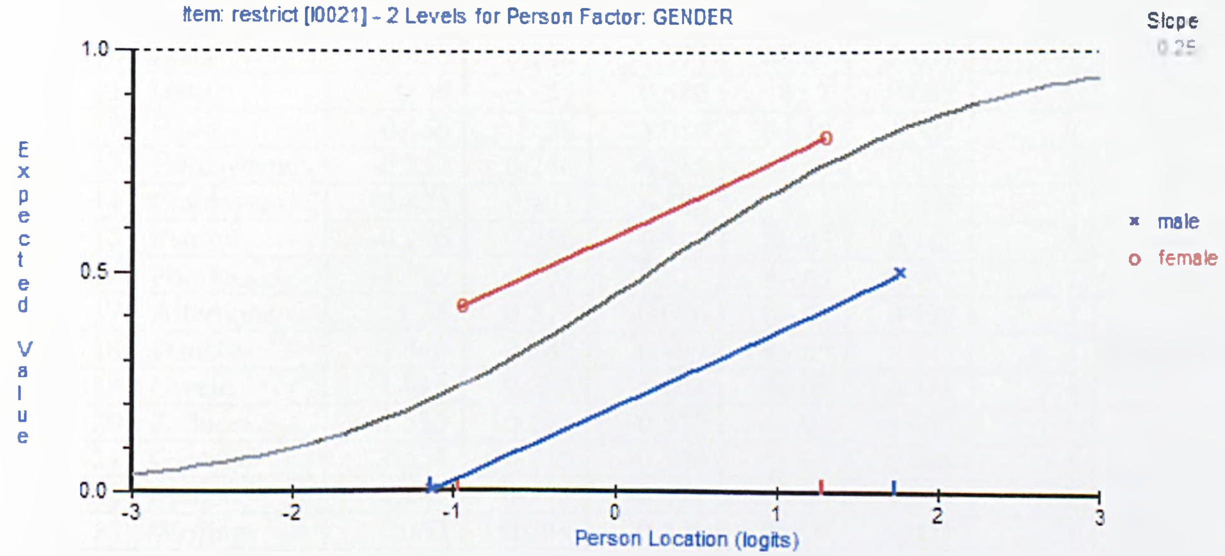
**Table 6.4 Individual Item fit of 24 Item scale**

| Item | Statement | Location | SE | FitResid | DF | ChiSq | DF | Prob |
|---|---|---|---|---|---|---|---|---|
| 1 | Push | 2.073 | 0.301 | -1.33 | 87.16 | 4.58 | 1 | 0.032 |
| 2 | Energy | 0.641 | 0.255 | -1.45 | 87.16 | 6.13 | 1 | 0.013 |
| 3 | Day | 0.798 | 0.257 | 0.544 | 87.16 | 2.38 | 1 | 0.123 |
| 4 | Myjob | -4.14 | 0.597 | -0.153 | 87.16 | 0.30 | 1 | 0.583 |
| 5 | People | -1.371 | 0.286 | -0.164 | 86.21 | 0.13 | 1 | 0.712 |
| 6 | Restrict | 0.188 | 0.251 | 1.38 | 87.16 | 2.96 | 1 | 0.085 |
| 7 | Cant | -0.166 | 0.253 | -0.275 | 86.21 | 0.02 | 1 | 0.887 |
| 8 | Paces | 2.192 | 0.312 | -0.497 | 86.21 | 0.87 | 1 | 0.352 |
| 9 | Guilt | -0.163 | 0.253 | -0.529 | 86.21 | 0.18 | 1 | 0.673 |
| 10 | Long | 0.458 | 0.254 | -1.796 | 86.21 | 3.45 | 1 | 0.063 |
| 11 | Help | 0.359 | 0.254 | 0.467 | 85.27 | 0.20 | 1 | 0.654 |
| 12 | Hand | 0.453 | 0.257 | 1.005 | 84.32 | 0.85 | 1 | 0.355 |
| 13 | clumsyhands | -0.239 | 0.253 | -0.138 | 86.21 | 0.01 | 1 | 0.939 |
| 14 | Employers | -2.845 | 0.408 | 0.187 | 85.27 | 0.78 | 1 | 0.375 |
| 15 | Painful | -0.292 | 0.254 | 0.87 | 86.21 | 0.32 | 1 | 0.568 |
| 16 | Poorhands | -1.068 | 0.272 | -0.78 | 86.21 | 1.47 | 1 | 0.225 |
| 17 | Afternoon | 1.35 | 0.273 | -0.028 | 86.21 | 1.06 | 1 | 0.303 |
| 18 | Double | -1.454 | 0.288 | 0.386 | 86.21 | 4.82 | 1 | 0.028 |
| 19 | Givein | 1.778 | 0.29 | -0.985 | 86.21 | 0.14 | 1 | 0.705 |
| 20 | Reduce | -1.564 | 0.294 | -0.668 | 86.21 | 0.58 | 1 | 0.446 |
| 21 | Overdo | 1.549 | 0.282 | -0.739 | 85.27 | 0.48 | 1 | 0.489 |
| 22 | Reliant | -1.143 | 0.274 | -0.61 | 87.16 | 1.15 | 1 | 0.282 |
| 23 | Writing | -0.057 | 0.251 | 0.855 | 87.16 | 3.69 | 1 | 0.054 |
| 24 | Moretired | 2.662 | 0.338 | -0.123 | 87.16 | 0.51 | 1 | 0.476 |

*Differential Item Functioning (DIF)*

A DIF analysis was then performed and it became apparent that Item 6 (restrict) was displaying significant Uniform DIF by Gender (Figure 6.9).

**Figure 6.10 DIF by Gender**



Thus, the ‘restrict’ item was removed from the analysis.

This left 23 Items in the scale for further analysis. The Individual Item Fit of the 23 items was as follows, the 3 Items displaying the most misfit were highlighted:

### Table 6.5 Individual Item fit of 23 Item scale

| Item | Statement | Location | SE | FitResid | DF | ChiSq | DF | Prob |
|---|---|---|---|---|---|---|---|---|
| 1 | Push | 2.108 | 0.305 | -1.325 | 86.99 | 5.521 | 1 | 0.018 |
| 2 | Energy | 0.644 | 0.257 | -1.283 | 86.99 | 7.159 | 1 | 0.007 |
| 3 | Day | 0.807 | 0.26 | 0.592 | 86.99 | 0.973 | 1 | 0.324 |
| 4 | Myjob | -4.102 | 0.585 | -0.119 | 86.99 | 0.315 | 1 | 0.574 |
| 5 | People | -1.389 | 0.287 | -0.193 | 86.05 | 0.239 | 1 | 0.624 |
| 7 | Cant | -0.166 | 0.255 | 0.016 | 86.05 | 0.069 | 1 | 0.792 |
| 8 | Paces | 2.216 | 0.314 | -0.489 | 86.05 | 1.256 | 1 | 0.262 |
| 9 | Guilt | -0.171 | 0.255 | -0.529 | 86.05 | 0.253 | 1 | 0.615 |
| 10 | Long | 0.455 | 0.256 | -1.771 | 86.05 | 2.635 | 1 | 0.104 |
| 11 | Help | 0.36 | 0.257 | 0.586 | 85.1 | 0.089 | 1 | 0.765 |
| 12 | Hand | 0.466 | 0.26 | 1.019 | 84.16 | 1.331 | 1 | 0.248 |
| 13 | clumsyhands | -0.252 | 0.256 | -0.212 | 86.05 | 0.217 | 1 | 0.641 |
| 14 | Employers | -2.825 | 0.401 | 0.188 | 85.1 | 1.209 | 1 | 0.271 |
| 15 | Painful | -0.296 | 0.256 | 0.843 | 86.05 | 0.185 | 1 | 0.667 |
| 16 | Poorhands | -1.095 | 0.273 | -0.817 | 86.05 | 0.462 | 1 | 0.496 |
| 17 | Afternoon | 1.38 | 0.277 | -0.076 | 86.05 | 0.729 | 1 | 0.393 |
| 18 | Double | -1.446 | 0.288 | 0.465 | 86.05 | 8.047 | 1 | 0.004 |
| 19 | Givein | 1.812 | 0.294 | -1.008 | 86.05 | 0.488 | 1 | 0.484 |
| 20 | Reduce | -1.555 | 0.293 | -0.577 | 86.05 | 1.097 | 1 | 0.294 |
| 21 | Overdo | 1.551 | 0.285 | -0.552 | 85.1 | 0.086 | 1 | 0.769 |
| 22 | Reliant | -1.145 | 0.275 | -0.399 | 86.99 | 1.277 | 1 | 0.258 |
| 23 | Writing | -0.063 | 0.254 | 0.776 | 86.99 | 6.027 | 1 | 0.014 |
| 24 | Moretired | 2.707 | 0.341 | -0.147 | 86.99 | 0.736 | 1 | 0.390 |

As can be seen, the removal of the ‘restrict’ Item altered the structure of the remaining Items and the overall Fit of the scale actually became worse when the ‘restrict’ item was removed from the analysis. No significant DIF was apparent on any of the Items, for any person factor, at this point.

Despite the overall fit and the individual item fit being acceptable if a Bonferroni correction was applied (p=0.05 / 23 Items = 0.00217), Item reduction was continued to manufacture a set of Items which displayed a better fit to the Rasch Model.

At this stage Item 18 (double) was removed from the analysis as it was displaying the biggest misfit to the model.

The Individual Item Fit for the remaining 22 Item Scale was as follows:

**Table 6.6 Individual Item fit of 22 Item scale**

| Item | Statement | Location | SE | FitResid | DF | ChiSq | DF | Prob |
|---|---|---|---|---|---|---|---|---|
| 1 | Push | 2.024 | 0.303 | -1.289 | 86.81 | 5.138 | 1 | 0.023 |
| 2 | Energy | 0.591 | 0.258 | -1.348 | 86.81 | 6.647 | 1 | 0.009 |
| 3 | Day | 0.756 | 0.261 | 0.506 | 86.81 | 1.144 | 1 | 0.284 |
| 4 | Myjob | -4.212 | 0.58 | -0.128 | 86.81 | 0.312 | 1 | 0.576 |
| 5 | People | -1.455 | 0.289 | -0.182 | 85.87 | 0.353 | 1 | 0.552 |
| 7 | Cant | -0.222 | 0.256 | 0.017 | 85.87 | 0.031 | 1 | 0.859 |
| 8 | Paces | 2.148 | 0.313 | -0.471 | 85.87 | 1.181 | 1 | 0.277 |
| 9 | Guilt | -0.234 | 0.256 | -0.568 | 85.87 | 0.151 | 1 | 0.697 |
| 10 | Long | 0.39 | 0.257 | -1.815 | 85.87 | 2.313 | 1 | 0.128 |
| 11 | Help | 0.295 | 0.258 | 0.582 | 84.93 | 0.033 | 1 | 0.855 |
| 12 | Hand | 0.416 | 0.261 | 0.947 | 83.98 | 1.637 | 1 | 0.200 |
| 13 | clumsyhands | -0.311 | 0.257 | -0.161 | 85.87 | 0.343 | 1 | 0.557 |
| 14 | Employers | -2.87 | 0.397 | 0.173 | 84.93 | 1.533 | 1 | 0.215 |
| 15 | Painful | -0.35 | 0.257 | 0.887 | 85.87 | 0.319 | 1 | 0.572 |
| 16 | Poorhands | -1.178 | 0.276 | -0.757 | 85.87 | 0.354 | 1 | 0.552 |
| 17 | Afternoon | 1.329 | 0.278 | -0.218 | 85.87 | 0.592 | 1 | 0.441 |
| 19 | Givein | 1.727 | 0.293 | -0.948 | 85.87 | 0.374 | 1 | 0.541 |
| 20 | Reduce | -1.634 | 0.295 | -0.582 | 85.87 | 0.927 | 1 | 0.335 |
| 21 | Overdo | 1.506 | 0.286 | -0.635 | 84.93 | 0.11 | 1 | 0.739 |
| 22 | Reliant | -1.221 | 0.277 | -0.435 | 86.81 | 1.062 | 1 | 0.302 |
| 23 | Writing | -0.121 | 0.255 | 0.796 | 86.81 | 6.685 | 1 | 0.009 |
| 24 | Moretired | 2.626 | 0.339 | -0.06 | 86.81 | 0.612 | 1 | 0.433 |

Again, there was no significant DIF on any of the Items, for any person factor, at this point. The overall Item-Trait Interaction improved a lot following the removal of the 'double' item but the Individual Item Fit was still identifying a couple of items that are displaying a certain amount of misfit to the Rasch model (see Table 6.8 below).

It was decided that the 'writing' item should be the next candidate for removal. The Chi-square fit statistics for both of the misfitting items (writing & energy) were approximately equivalent, but it was decided that the 'writing' item should be removed first due to the relative lack of discrimination between ability groups when compared with the 'energy' item. Following the removal of the 'writing' item, the Individual Item Fit for the remaining 21 Items was as follows:

**Table 6.7 Individual Item fit of 21 Item scale**

| Item | Statement | Location | SE | FitResid | DF | ChiSq | DF | Prob |
|---|---|---|---|---|---|---|---|---|
| 1 | Push | 2.039 | 0.305 | -1.251 | 86.61 | 3.708 | 1 | 0.054 |
| 2 | Energy | 0.592 | 0.262 | -1.281 | 86.61 | 3.224 | 1 | 0.072 |
| 3 | Day | 0.753 | 0.265 | 0.69 | 86.61 | 3.312 | 1 | 0.068 |
| 4 | Myjob | -4.252 | 0.583 | -0.166 | 86.61 | 0.306 | 1 | 0.580 |
| 5 | People | -1.458 | 0.288 | -0.026 | 85.67 | 1.097 | 1 | 0.294 |
| 7 | Cant | -0.233 | 0.259 | -0.082 | 85.67 | 0.025 | 1 | 0.874 |
| 8 | Paces | 2.176 | 0.315 | -0.491 | 85.67 | 0.682 | 1 | 0.408 |
| 9 | Guilt | -0.251 | 0.259 | -0.595 | 85.67 | 0.684 | 1 | 0.408 |
| 10 | Long | 0.38 | 0.26 | -1.848 | 85.67 | 2.974 | 1 | 0.084 |
| 11 | Help | 0.291 | 0.261 | 0.636 | 84.73 | 0.091 | 1 | 0.762 |
| 12 | Hand | 0.414 | 0.264 | 1.101 | 83.79 | 2.775 | 1 | 0.095 |
| 13 | clumsyhands | -0.323 | 0.259 | -0.03 | 85.67 | 0.457 | 1 | 0.499 |
| 14 | Employers | -2.879 | 0.393 | 0.238 | 84.73 | 1.488 | 1 | 0.223 |
| 15 | Painful | -0.36 | 0.26 | 0.918 | 85.67 | 0.187 | 1 | 0.665 |
| 16 | Poorhands | -1.184 | 0.276 | -0.577 | 85.67 | 0.084 | 1 | 0.771 |
| 17 | Afternoon | 1.334 | 0.28 | -0.232 | 85.67 | 0.703 | 1 | 0.401 |
| 19 | Givein | 1.746 | 0.295 | -0.975 | 85.67 | 0.673 | 1 | 0.412 |
| 20 | Reduce | -1.691 | 0.297 | -0.66 | 85.67 | 1.298 | 1 | 0.254 |
| 21 | Overdo | 1.522 | 0.289 | -0.516 | 84.73 | 0.772 | 1 | 0.379 |
| 22 | Reliant | -1.257 | 0.278 | -0.488 | 86.61 | 1.578 | 1 | 0.209 |
| 24 | Moretired | 2.639 | 0.337 | 0.172 | 86.61 | 0.201 | 1 | 0.653 |

The remaining 21 Items displayed a good overall individual fit to the Rasch model and all individual items fit at p=0.05 (Table 6.7). The scale was DIF-free for all person factors, with only one item displaying any DIF statistic of any note, but not so after adjusting for the number of DIF tests undertaken (No. of items (21)*No. of DIF types (2)*No. of Person Factors (4)), giving a Bonferroni adjusted alpha level of p=0.05/168 tests = 0.000298.

A summary of the overall fit statistics for the item reduction phase after the item discrimination analysis is given in Table 6.8.

**Table 6.8 Table summarising the different versions of the scale**

| Version | Item | | Person | | Chi squared Probability | PSI |
|---|---|---|---|---|---|---|
| | Mean | SD | Mean | SD | | |
| 24 | -0.19 | 0.806 | -0.231 | 0.688 | 0.043 | 0.907 |
| 23 | -0.218 | 0.734 | -0.254 | 0.715 | 0.013 | 0.905 |
| 22 | -0.259 | 0.734 | -0.271 | 0.705 | 0.079 | 0.902 |
| 21 | -0.26 | 0.746 | -0.281 | 0.696 | 0.194 | 0.899 |

*Test – re-test reliability of the 21 – Item Scale*

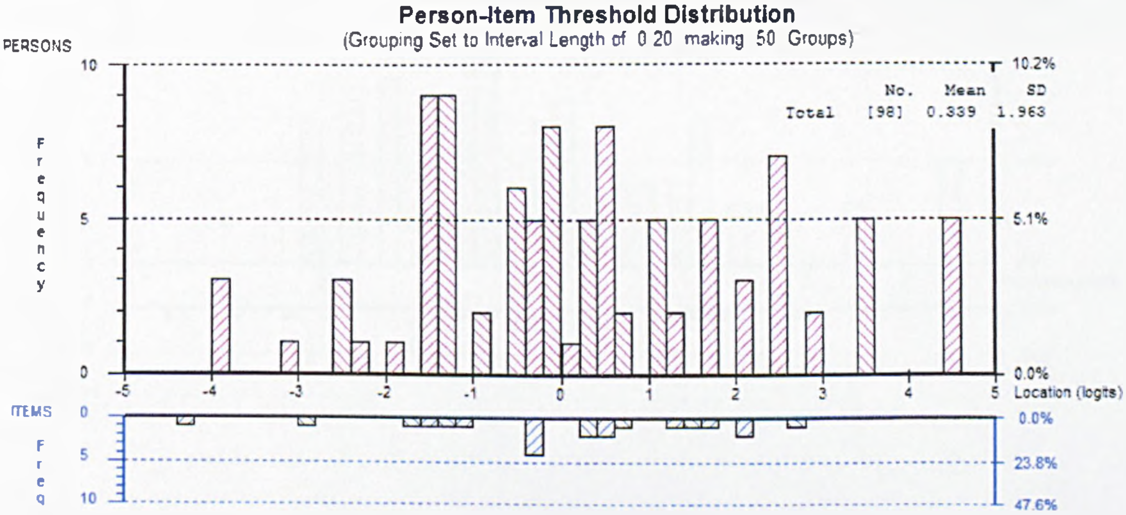
The 21 - item scale was applied to the datasets from the Test re-test postal survey for the 86 – item scale (n = 49) which were collected two weeks apart to test reliability of the questionnaire over time. Spearman's non parametric correlation was used and the test re-test correlation should be at least 0.8. Analysis of the returned scale revealed a spearman's (non-parametric) correlation of 0.861. This value suggests that the 21 – item scale has a good reliability over time. The test-retest questionnaire had 61 people at timepoint 1 and 47 at timepoint 2 and the correlation is based on the latter. 2 sets of data could not be used due to missing data which is why n=47 at timepoint 2.

The use of correlation coefficient in conjunction with median values for both timepoints was the appropriate statistical method for test re-test analysis in this case, because these are ordinal data. Including the median WIS values at time 1 = 11, and at time 2 = 10 shows that there was no systematic difference across the timepoints.

*Person and Item Location*

This item map below shows that the 21 work instability items are targeting the sample well.

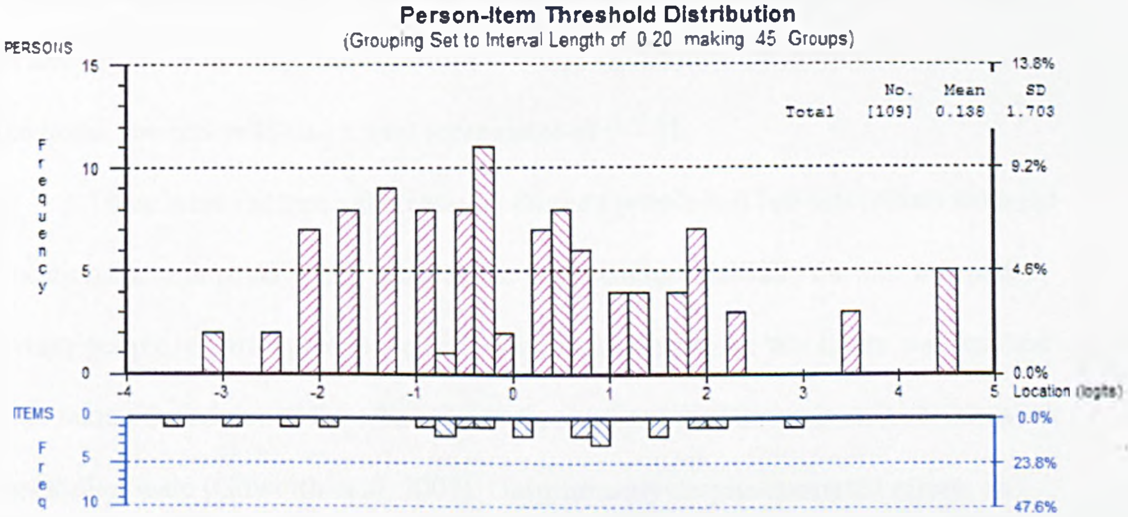
**Figure 6.11 Item Map for 21 Item Scale**



*Overall Fit of the 21 item scale to the initial dataset*

The 21-Item set was deemed a valid 'solution' and therefore the final 21 items from the test-retest data were then selected from the original postal questionnaire to see if the scale remained valid on the initial dataset. When the 21-Item set was reanalysed with data from the original postal questionnaire the targeting remained excellent, with a mean location of 0.138 logits, where the scale is centered on zero logits (Figure 6.12). The item trait interaction statistic (overall scale fit) showed good fit to the Rasch model with a Chi – square probability of 0.253. The person separation index (PSI) which is a measure of reliability and indicates how well the items are differentiating between persons and is consistent with individual use was 0.881. All items were DIF free.

**Figure 6.12 Item Map for 21 Item Scale when applied to initial dataset**



Therefore after the items were subjected to another round of Rasch analysis, the Multiple Sclerosis Work Instability Scale (MS - WIS) consisted of 21 items (Appendix 11)

### 6.6 Criterion Validity

The scale scores derived from the final 21 item version were then were examined across the three aggregated levels of work instability as defined by the vocational assessment, and values were examined to determine appropriate cut points to identify those needing modifications at work. Given the dichotomous responses to the items, the MS-WIS had a total score range of 0 – 21.

There were thirteen valid cases as thirteen people had full sets of data from the vocational assessments which acted as the gold standard. Initially the aim was to get twenty people to participate in the full vocational assessment; this figure was reached after taking the advice of the team who had completed the Rheumatoid Arthritis Work Instability Scale (Gilworth et al, 2003). Unfortunately despite concerted efforts on behalf of the researcher, thirteen was the maximum number of people who agreed to participate and then actually turn up on the day of the assessments; however these thirteen participants were largely representative of the larger sample in terms of age and sex. Therefore the primary researcher then made the decision to proceed with the analysis using these numbers after taking further advice from the authors of the RA – WIS and the professionals who carried out the assessments. This analysis was retrospectively applied to the test re-test (n= 47) dataset.

The five levels of work instability as determined by the experts were compressed to 3 levels of risk for the purpose of cross tabulation to determine sensitivities and specificities and the three aggregated levels of work instability used to determine sensitivity and specificity were :

Risk level 0 = none or little problem

Risk level 1 = advice or intervention needed

Risk level 2 = severe problem

Three levels of risk were applied to the scores obtained from the thirteen respondents who completed the MS WIS. Cut points were set at scores of 8 and 17 calculated from the 21 item scale. The lower cut point of 8 represented a shift from a low to medium level of risk and 17 the transition to the high risk level, so giving:

Risk level = 0 for scores of 0 – 7

Risk level = 1 for scores of 8 – 16

Risk level = 2 for scores of 17 and above

### Level of Risk (as per WIS) verses Gold Standard Assessment Cross tabulation

The level of risk given to the thirteen people during the gold standard assessments were compared to the level of risk they scored after completing the MS WIS on the same day as the vocational assessment.

The following tables examine the sensitivity and specificity of the proposed cut points for low, medium (cut off point =8) and high (cut off point =17) risk of work instability.

**Table 6.9 Level of Risk (as per WIS) verses Gold Standard Assessment Cross tabulation**

Count

| | | gold2 | | | |
|---|---|---|---|---|---|
| | | None or little problem | Advice intervention needed | Severe problem | Total |
| Risk | 0 | 3 | 1 | 0 | 4 |
| | 1 | 2 | 5 | 0 | 7 |
| | 2 | 0 | 0 | 2 | 2 |
| Total | | 5 | 6 | 2 | 13 |

**Table 6.10a Gold Standard Assessment of low risk of work instability**

Count

| | | gold2 | | | |
|---|---|---|---|---|---|
| | | None or little problem | Advice intervention needed | Severe problem | Total |
| Risk | 0 | 3 | 1 | 0 | 4 |
| | 1 | 2 | 5 | 0 | 7 |
| | 2 | 0 | 0 | 2 | 2 |
| Total | | 5 | 6 | 2 | 13 |

The gold standard assessments stated that 5 people did not have a problem at work and no risk of job loss (highlighted in green)

**Table 6.10b Screening tool assessment of low risk of work instability**

Count

| | | gold2 | | | |
|---|---|---|---|---|---|
| | | None or little problem | Advice intervention needed | Severe problem | Total |
| Risk | 0 | 3 | 1 | 0 | 4 |
| | 1 | 2 | 5 | 0 | 7 |
| | 2 | 0 | 0 | 2 | 2 |
| Total | | 5 | 6 | 2 | 13 |

The scale stated that 3 people did not have a problem at work (highlighted in orange)

The specificity of the scale is 60% as 3/5 people do not have any problems at work as determined by the scale (*They do not have a problem at work and the scale states there are no problems at work).*

Sensitivity and specificity at cut off point = 8

**Table 6.11a Gold Standard Assessment of medium risk of work instability**

Count

| | | gold2 | | | |
|---|---|---|---|---|---|
| | | None or little problem | Advice intervention needed | Severe problem | Total |
| Risk | 0 | 3 | 1 | 0 | 4 |
| | 1 | 2 | 5 | 0 | 7 |
| | 2 | 0 | 0 | 2 | 2 |
| Total | | 5 | 6 | 2 | 13 |

The gold standards stated that eight people (highlighted in red) were at a risk of job loss (i.e. risk greater than 0).

**Table 6.11b Screening Tool Assessment of medium risk of work instability**

Count

| | | gold2 | | | |
|---|---|---|---|---|---|
| | | None or little problem | Advice intervention needed | Severe problem | Total |
| Risk | 0 | 3 | 1 | 0 | 4 |
| | 1 | 2 | 5 | 0 | 7 |
| | 2 | 0 | 0 | 2 | 2 |
| Total | | 5 | 6 | 2 | 13 |

The scale stated that seven people (highlighted in blue) were at risk of job loss.

Therefore the sensitivity of the scale when compared with the gold standard assessment is 88% as the scale states that 7/8 have a problem (*They are having problems at work and the scale states they have a problem).*

Sensitivity and specificity at cut-off point = 17

At a score 17 and above the sensitivity and specificity of the scale reaches 100%.

**Table 6.12a Gold Standard Assessment of high risk of work instability**

Count

| | | Gold2 | | | |
|---|---|---|---|---|---|
| | | None or little problem | Advice intervention needed | Severe problem | Total |
| Risk | 0 | 3 | 1 | 0 | 4 |
| | 1 | 2 | 5 | 0 | 7 |
| | 2 | 0 | 0 | 2 | 2 |
| Total | | 5 | 6 | 2 | 13 |

The gold standards stated that two people (highlighted in red) were at a high risk of job loss (i.e. risk greater than 1).

**Table 6.12b Screening Tool Assessment of high risk of work instability**

Count

| | | Gold2 | | | |
|---|---|---|---|---|---|
| | | None or little problem | Advice intervention needed | Severe problem | Total |
| Risk | 0 | 3 | 1 | 0 | 4 |
| | 1 | 2 | 5 | 0 | 7 |
| | 2 | 0 | 0 | 2 | 2 |
| Total | | 5 | 6 | 2 | 13 |

The scale also stated that two people (highlighted in blue) were at high risk of job loss as seen in table

The sensitivity of the scale when compared with the gold standard assessment is 100% as the scale states that 2/2 are at high risk of job loss (*They have a problem and the scale states there is a problem).*

In summary, the MS Work Instability Scale is very quick and simple to complete. It is scored in 3 bands indicating low, medium and high risk of Work Disability. The medium – risk threshold has 88% sensitivity and 60% specificity and the sensitivity and specificity of the scale reaches 100% at the high – risk threshold with regards to the need for work place intervention.

## 6.7 Concurrent Validity

There were significant (P<0.005 correcting for multiple testing) and substantive (rho>0.3) associations between MSWIS and all of the other scales used in the postal questionnaires. As expected, positive correlations were found between MSWIS and MFIS (Modified Fatigue Impact Scale) (rho = 0.779, P<0.001) and MSIS (Multiple Sclerosis Impact Scale) (rho = 0.804, P<0.001), whereas negative correlations were found between MSWIS and LMSQoL (Leeds Multiple Sclerosis Quality of Life Scale) (rho = -0.389, P = 0.004) and SFS (Social Function Scale) (-0.544, P<0.001). The strongest associations were between MSWIS and MSIS/MFIS.

Positive correlations mean that those who had a high risk of work instability also had high levels of fatigue and their multiple sclerosis symptoms impacted greatly on their day – to – day life in the previous 2 weeks and vice - versa. Negative correlations revealed that those who had a low risk of work instability however had a good quality of life and social functioning and again the converse was true.

**Table 6.13 Scale Comparisons**

| Scale | N patients | Median (IQR) [Range] | Correlations | |
|---|---|---|---|---|
| MSWIS | 52 | 11.50 (6.00 to 15.00) [0 to 20) | | |
| LMSQoL | 57 | 21.00 (20.00 to 23.00) [14 to 27] | MSWIS<br>MSIS | ρ=-0.389 P=0.004<br>ρ=-0.454 P=0.001 |
| SFS | 56 | 21.00 (15.25 to 23.75) [10 to 28] | MSWIS<br>MSIS<br>MFIS | ρ=-0.544 P<0.001<br>ρ=-0.598 P<0.001<br>ρ=-0.665 P<0.001 |
| MSIS | 54 | 62.00 (44.25 to 81.25) [29 to 125] | MSWIS<br>MFIS | ρ=0.804 P<0.001<br>ρ=0.826 P<0.001 |
| MFIS | 55 | 35.00 (18.00 to 46.00) [0 to 76] | MSWIS | ρ=0.779 P<0.001 |

# Chapter 7: Discussion

The hypothesis that it is possible to identify people who are at risk of losing employment due to multiple sclerosis by developing a valid and reliable work instability scale (WIS) for Multiple Sclerosis has been proven. This thesis has fulfilled its aim which was to devise an outcome measure that could be used to assess the risk of job loss in those with multiple sclerosis, that is, a work instability scale (WIS) for multiple sclerosis (MS).

## 7.1 Strengths of the Study

The study has used a mixed methodology approach, combining qualitative and quantitative methods to provide a high quality patient centred scale for work instability. The methodology used had already produced a validated scale for work instability in Rheumatoid Arthritis (Gilworth et al, 2003) and Traumatic Brain injury (Gilworth et al, 2006). In the qualitative work the potential for bias was reduced by having more than one researcher conducting the interviews, and in item selection. It may be thought that purposive sampling introduced bias in terms having preconceived ideas about the patient characteristics relevant to the research question. However purposive sampling was actually employed in this study with the aim of decreasing bias by ensuring that the sample was representative of the MS population.

In the quantitative work the most rigorous measurement model, the Rasch Model, was adopted for scale development giving state- of- the- art psychometrics for scale construction. This included tests for item bias and a criterion validity test to identify the most discriminating items. Consequently, even though the final scale was only 21 dichotomous items, the level of reliability was high enough for individual patient use.

Expert help had been enlisted for both the qualitative and quantitative entities, in terms of both my training and reviewing the analysis. The resulting questionnaire was validated against the "Gold Standard" and was found to have high specificity and sensitivity.

Apart from detailed work placement assessments that are undertaken within current services there is no existing standardised questionnaire based assessment that can easily capture the risk of job loss in multiple sclerosis. Therefore this piece of work does have the potential for clinical benefit.

## 7.2 Limitations of the Study

There are a number of limitations to the study. Recruitment was a problem at each stage. In the qualitative work it was not possible to recruit the expected number in each cell, and there was a particular shortfall with female blue collar workers with progressive disease.

Reasons for the disparity in the numbers of people in each of the cells can, in part, be considered consistent with previous research findings about changing work and work disability in multiple sclerosis, bearing in mind inclusion into a particular cell was determined by sex and type of job. When reviewing the demographics of work in multiple sclerosis, the 1991 study of 210 registrants of the Vancouver Island Multiple Sclerosis Society showed that in relation to employment status over two-thirds (66.7%) reported a change in their job status since the onset of MS, and almost half (44.8%) reported having to make a forced change in their field of employment. The greatest change in employment included a 5.4% decrease in medicine / health professions, and a 5% decrease in the service industries. There was only a 1% decrease in those employed in clerical work (Jackson et al, 1991). It would have to be considered that the minimal decrease in persons with multiple sclerosis working in clerical areas is due to their ability to maintain their jobs longer in this area. Another reason for this could be that some individuals move to clerical work when they cannot function adequately in other areas of employment any longer hence our study sample had the requisite number of white collar workers.

A number of studies have found that higher education was associated with lower probability of unemployment (Bushe et al, 2003, Solari et al, 2001) and this is most likely to be because, generally those with a higher level of education and more qualifications tend to hold less manual jobs. This would also add weight to the finding that people are able to maintain clerical work for longer as it tends not to be manual in its nature.

In their 2001 work, Koch et al looked at quality of life (QoL) in those with multiple sclerosis and explored the areas of life which could affect this. One of the conclusions drawn was that a lower subjective QoL was associated with a greater number of physical symptoms, forced lifestyle changes, such as, loss of employment and a lower education level. There is an obvious correlation between physical disability due to severe symptoms, low education level and job loss. An example is the blue collar worker experiencing problems carrying out their physical job; lighter work may not be an option as a low level of education is the most likely reason for their doing a manual job.

The existing literature supports the fact that it was also difficult to recruit more females from the pool of patients available to participate in the research, as many of them tended not to have paid employment outside the home. It also goes some way to explaining why only one female with progressive disease in a "blue-collar" job was included. These are the most significant demographic features cited for job loss and this is reflected by the fact that there was only one person who fitted those criteria in the entire sample population. All other women in that situation who were attending hospital clinic appointments were not in paid employment.

The literature revealed that males with multiple sclerosis are more likely to be employed than females, the female sex increasing the risk of premature unemployment (Solari et al, 2001, Kornblith et al 1996). There could be various reasons for this. When the disease worsens in female patients, it may become increasingly difficult to work and attend to household responsibilities as well. Unable to do both, the female with MS may relinquish outside employment. In our society, the only legitimate reason for men not working or looking for work is physical or mental incapacity resulting from disease or injury (Weiss et al, 1971). It could be that it is more socially acceptable for disabled women not to work rather than disabled men due to the other roles women historically play, such as, mother and "housewife".

In the quantitative work, obtaining the sample size necessary for the Rasch analysis was also difficult. Initially, insufficient numbers of people returned the postal questionnaires and so others had to be recruited from the MS specialist hospital clinics. This gave a sufficient degree of precision to estimate item differences to about 0.5 logit (Bond and Fox, 2001). Fortunately, the nature of the Rasch model is such that it does not require a representative sample to estimate item difficulties, but it is always useful to have a sample which covers the full range of the variable being measured, in this case Work Instability.

In part, this goes to offset a possible disadvantage of using the additional recruitment from an MS clinic, which may have introduced unknown bias into the scale development. The sample frame based upon the Leeds MS register was out of date and newly diagnosed people, people who had changed address, either due to normal house moves or those whose disease had worsened and were then in residential or nursing care were excluded from the original sample.

Non response also introduces bias, Ford showed a significant difference in disease course in those who requested their details be removed from the MS register. The aim of her 2006 study was to determine rates of consent for a multiple sclerosis community based register and to investigate authorisation bias by examining the disease characteristics of those refusing consent (Ford, 2006). Out of a total of 820 people questioned 34 (4.1%) wished their details to be removed from the MS register. There was a significant difference in disease course in this group with more patients with benign relapsing remitting disease compared with the prevalent population. Therefore it was recognised that use of the MS register to recruit patients to the study did introduce bias as those refusing consent to be included on it had different disease characteristics than those who consented.

This however may or may not have been of significance in the development of the scale as those who were at extremes of the disease process were unlikely to have had significant information to add to the study. This is because those at the early stages of the disease were unlikely to be experiencing work instability as the evidence from the literature showed this tended to be an issue over longer lengths of time. Those in residential/ nursing home care were highly unlikely to be in employment and so unlikely to be able to fill in the MS-WIS postal questionnaire in any case.

It is possible that those attending hospital clinics may have had more severe disease than those who filled out the form in the community; conversely they could have been on Disease Modifying Therapy and have had more quiescent disease so it is difficult to ascertain the impact of this upon the scale development. Also if someone had received the postal questionnaire but did not return it out of apathy initially but then did so when they were attending a hospital clinic may not have been a very motivated individual which may have been reflected in their questionnaire responses. Recruitment was an issue at all stages of scale development and the decision had to be made to recruit from hospital clinics for this particular stage, fully recognising that a different profile of people were being used which may or may not have adversely affected scale development.

The scale was validated against the current "Gold Standard" assessment. This was to assess the reliability and sensitivity of the draft measurement against the pre-existing method of determining work instability which is vocational assessments. These assessments were full vocational assessments carried out by experienced ergonomic physiotherapists specialising in the area of work loss, who had carried out the same role in the development of the RA-WIS. However it must be recognised that these assessments, while considered to be a clinical gold standard in this field have never been independently validated. Also as these assessments were more focused on physical problems the cognitive problems that can occur in MS may have been missed. The "Gold Standard" assessment used in the development of the TBI-WIS was a detailed neuropsychological assessment (Gilworth et al, 2006) which may also have been useful in the development of the WIS.

These limitations were taken into account, but comparison against the current "Gold Standard" was the methodology used in both the development of the RA-WIS and the TBI WIS (Gilworth et al, 2003, Gilworth et al, 2006) and so was deemed as the most appropriate comparison for validation of the MS-WIS in the absence of a more robust method of measuring risk of work loss.

There is a need for further work to prospectively assess the predictive value of the scale. The obvious way to do so would be to follow up those who were shown to be at risk of job loss by the scale to determine the proportion of them that actually do lose their jobs and over what time scale, this figure could then be compared with the literature. The main limitation of this method is that it could potentially take a considerable length of time. The current literature suggests that during the first 10 years after diagnosis 50-80% of those with MS are out of work (Gronning et al, 1990, LaRocca et al, 1985, Kornblith et al, 1986) therefore a predictive validity study may need to run for approximately ten years which may not be sustaiable in terms of research study funding. As there are considerable time delays between the the onset of work instabilty to actual job loss it may be more useful just to observe those who are at medium to high risk of job loss at the outset as time to job loss is likely to be shorter. At present it must be acknowleged that the predictive validity of the MS-WIS is unknown and needs to be quantified however this is the same for the majority of newly developed scales used in clinical practice.

Finally it should be acknowledged that the study was undertaken on a relatively small population from Northern England. Future validation studies should be undertaken and, if the scale is to be used in other countries appropriate cultural adaptation studies should be undertaken.

## 7.3 Clinical implications of the study

### 7.3.1 Use as a screening tool

With its high level of reliability the MS WIS could be used as a screening tool in the clinical setting. It would be administered alongside other measurement tools used in the assessment of multiple sclerosis to address an area that is often not specifically asked about by clinicians. Once it has been identified that the individual is at risk of work loss, appropriate interventions can be instituted to reduce the level of risk as determined by the scale. These interventions are best carried out within the realms of a multidisciplinary team by health professionals with training in Vocational Rehabilitation who fully understand the complexities of job retention measures.

As the scale is self – completed it could be administered to those attending a hospital clinic by reception staff to be filled in as they were waiting to see the doctor or specialist nurse. The results of the scale could then be addressed during the consultation. If the WIS score has increased the clinician would then be alerted to identifying and addressing the symptoms that may be responsible for this wherever possible. A change in the WIS score may make the clinician more vigilant in terms of history taking so that new or worsening symptoms can be discussed, for example, have their fatigue or pain levels worsened or has continence become an issue? It could be determined at a follow up appointment whether or not addressing the problems has changed the level of work instability reported.

Obvious barriers to the use of the scale in clinical practice would be the unwillingness of the clinician to use it due to factors ranging from a personal opinion that history taking can provide all the information necessary to a perceived idea that they will not have enough time to use this within a clinical setting and it may add stress to an already busy clinic. However the clinician does not have to administer the scale as it is self completed and they would be using the consultation to address symptoms in any case, the normal running of the clinic would not be affected. In this setting the scale would be merely used as an adjunctive measure to good clinical practice. However if resistance to use remained widespread a qualitative study could be carried out among clinicians who had used to the scale to ascertain their views regarding the MS-WIS particularly how "user – friendly" it is in a clinical setting and how helpful the information yielded is in the management of the patient.

MS is a progressive disease and not all symptoms can be improved with moderators, such as medical intervention, and it may be that barriers to work persist despite treatment of the symptoms, for example, problems within the workplace itself. Therefore the impact of other moderators in the work environment should be taken into account by the clinician. This project has shown that persons with MS did not like to draw attention to themselves within the workplace and did not want to be treated any differently than their colleagues and that fear of job loss was a huge factor in this. However they and their employers may not be aware of the Disability Discrimination Act (DDA). The DDA is piece of legislation that promotes civil rights for disabled people and protects disabled people from discrimination. The DDA 1995 aimed to end the discrimination that many disabled people face. The Act has been significantly extended, including by the Disability Discrimination Act 2005. It now gives disabled people rights in the areas of:

- employment
- education
- access to goods, facilities and services, including larger private clubs and land-based transport services
- buying or renting land or property, including making it easier for disabled people to rent property and for tenants to make disability-related adaptations
- functions of public bodies, for example issuing of licences

The Act requires public bodies to promote equality of opportunity for disabled people. It also allows the government to set minimum standards so that disabled people can use public transport easily and to that end will help somewhat with actually getting to work. It aims to give disabled people the freedom in the workplace to ask for adjustments to their working life safe in the knowledge that they are protected by staute in doing so.

Another moderater has been the significant improvement in government policy regarding disability. In March 2005 The National Service Framework (NSF) was introduced. This is a ten-year programme to improve health and social care services in England for people with long-term conditions, including MS. It is the latest in a series of NSFs covering major challenges to health and social care, such as coronary heart disease and mental health. There is good practice in many areas of England, but the aim of the NSF is to make good practice standard practice. The NSF was developed by a panel of experts, known as the External Reference Group, including MS society volunteers (the MS society of Great Britain and Northern Ireland is a United Kingdom based multiple sclerosis charity) and the society's medical advisor Professor Alan Thompson who made recommendations about the NSF to ministers and produced early drafts of the standards that should be reached. People with MS were invited to take part in Neurological Alliance Focus groups which gave civil servants an indication of the kinds of issues that people with Neurological conditions would like the NSF to address. Eleven "quality requirements" were identified for the NSF:

**National Service Framework: Key Points**

1. A person-centred service
2. Early recognition, prompt diagnosis and treatment
3. Emergency and acute management

4. Early and specialist rehabilitation
5. Community rehabilitation and support
6. Vocational rehabilitation
7. Providing equipment and accommodation
8. Providing personal care and support
9. Palliative care
10. Supporting family and carers
11. Caring for people with neurological conditions in hospital or other health and social care settings

The NSF has further clarification on all the points but looking at point 6 – Vocational Rehabilitation – it states that people with long term neurological conditions are to have access to appropriate vocational assessment, rehabilitation and ongoing support, to enable them to find, regain or remain in work and access other occupational and educational opportunities. The MS society's opinion (stated on their website at the current time) regarding this clarification echoes the findings of this research study that many people with MS who would like to work need support to be able to continue to do so. It also reflects this research as it particularly welcomed the recognition that people with MS may need support to remain in work, something that is often lost in the focus of getting people into work.

### 7.3.2 Use as an outcome measure

The MS-WIS could also be used as an outcome measure to assess the effect of interventions expected to impact on the person's ability to do their job. Further evidence of the responsiveness of the scale would support its use in this context. An example of such an intervention could be a Vocational Rehabilitation programme and the scale is already being used in Britain in this context. It is one of the primary outcome measures being used to evaluate a model of occupational rehabilitation for people with Multiple Sclerosis which is being developed at the National Hospital for Neurology and Neurosurgery (NHNN), University College London Hospital NHS Foundation Trust, London, United Kingdom. This hospital delivers a comprehensive MS service covering all stages of the disease from diagnosis to the management of severe disability.

The aim of the study was to define the intervention in practice in terms of content and cost, and to identify which outcome measures captured the impact of the intervention. A description of the study and how the MS-WIS is to be used in it is found in the appendices, courtesy of Joanna Sweetland who is an Occupational Therapist and is the project lead (Sweetland, Personal Communication, Appendix 12).

Another advantage of an outcome scale concerned with job retention would be in studies using the Disease Modifying Therapies (DMTs) which are expensive medications and, whose cost effectiveness is still largely undetermined. It would be interesting to see if the DMTs lessened the risk of work loss or caused it to remain stable over long periods of time and therefore have an impact on the socioeconomic consequences of Multiple Sclerosis. Thus the scale can be of use in clinical trials, particularly looking at the effect of DMTs in terms of employment. As previously discussed in this project parallels can be drawn between Rheumatoid Arthritis (RA) and MS in terms of the nature of the disease process and its effect on employment and that significant job loss is seen in both illnesses. However results from a four-year study have brought positive news to people with Rheumatoid Arthritis (RA) who are trying to stay in work. The PROWD (Prevention of Work Disability) study has shown that an anti-TNF treatment (Humira) plus the standard treatment (methotrexate) can be twice as effective as methotrexate alone at preventing job loss in people with early RA, potentially helping to double the number of people with RA that remain in work (Bejarano et al, 2006). This is the first study to use work impairment as a primary endpoint, work being job loss of any cause and/or imminent job loss and defined as i) RA- WIS score deterioration from baseline or ii) A persistently high RA-WIS score (>17). The secondary endpoints included, among other measures, the number of working days lost which was assessed by using diaries. The PROWD study was a multi-centre, randomised controlled trial which ran for 56 weeks.

A study has already been carried out which looked at the effects of a DMT (Glatiramer Acetate) on fatigue and days of absence from work in 291 treatment naive patients with relapsing remitting multiple sclerosis. The proportion of patients absent from work at least once was reduced by a factor of two from 65.1% to 30.1% which is a marked reduction in absence from work (Ziemssen et al, 2008). The MS-WIS could be used in such as study in future as a primary endpoint and the number of days of work lost can be used as a secondary endpoint as they have been shown to perform well in these respective settings by the PROWD study.

An advantage of the use of the scale in clinical trials in comparison with measuring employment rates is that it will actually measure risk of job loss and conversely show if treatment with a DMT is improving the level of work disability. Employment rates are much less sensitive and only show if there has been actual job loss or not by which stage is may be too late to help the patient stay in employment. As discussed previously it is easier to maintain someone in work than get them back to work.

The obvious disadvantage of use of the scale in clinical trials usage is that the trial may need to run for a longer time and therefore may be more laborious and expensive than one that may have, for example, MRI changes as an outcome measure. The scale is likely to be of use in a trial where the majority of DMTs are directed at patients with relapsing remitting disease, because, although they are at a lower risk of job loss than those with progressive disease they are likely to be at a younger age and so would be losing more work years in total resulting in significant socioeconomic problems for them.

It has been suggested that quoted figures for cognitive deficits in early multiple sclerosis appear to be higher than seen in current clinical practice. There may be various reasons for this. Firstly, the research done to assess cognition employed complex neuropsychological testing that can pick up subtle cognitive changes, which is not usually available to the average clinician in the setting of a busy outpatient clinic and therefore mild cognitive deficit may not be detected by the clinician. Secondly, as public awareness of MS improves people may be more likely to attend their doctor for mild symptoms, such as, altered sensation than they may have done previously and so MS may be being diagnosed and treated with DMTs at an earlier stage than previously before cognition has been significantly affected.

## 7.4 Vocational Rehabilitation

In his 2003 work, Frank stated that the introduction of vocational rehabilitation at the correct time is of the utmost importance for a successful return to work and that this phase probably lasts for 2-4 months, as if a patient is off work beyond that, they believe themselves to be "disabled" and therefore unable to work (Frank, 2003). The UK's attitude to return to work reinforces this attitude by completing medical care before considering a return to work. The thinking of health professionals must change in order for vocational rehabilitation to be successful. Not only must they be responsible for "curing" the unwell individual, they must also accept that they have a responsibility to help the patient return to work wherever possible, and broach this subject before negative attitudes are formed. The management of work disability has not been viewed as an important part of medical practice outside the specialties of rehabilitation and occupational medicine (Frank, 2003). At present, in the UK, General Practitioners are the body mainly responsible for the provision of "sick notes" but have little or no formal training in this area. Consideration should be given to further training for GPs regarding the role and time frame of vocational rehabilitation. General Practitioners feel reluctant to take on the vocational rehabilitation role as it effectively means denying their patient a "sick note". The patient could then perceive that their family doctor has been directly responsible for sending them back to work before they felt ready and could greatly interfere with the future doctor/patient relationship. The majority of general practitioners in the UK do not feel is their role to be "gatekeeper" for the working population and currently feel lack confidence when broaching the issues around vocational rehabilitation as they have received no formal training in the area.

Ideally vocational rehabilitation and the provision of "sick notes" should be carried out by occupational health physicians in this 2 – 4 month window (as discussed above) who would then work closely with the GP in setting out rehabilitation goals. Employers maintaining contact with the ill employee and supporting the rehabilitation program is also crucial at this stage. Different issues exist around Vocational Rehabilitation programmes in the United States of America. In 2003 Fraser et al assessed a group of 79 people with MS who were concerned about current employment or had a desire to return to gainful employment. They were then all enrolled on a job placement assistance program, which was free and carried out by trained rehabilitation counselling staff. The study sample consisted mainly of well educated females who had relapsing-remitting MS, one third of whom were trying to maintain their jobs but there was a high dropout rate at 39%. The drop-out rate could also be explained in part by the fact that 41% were receiving social security and enrolling on a vocational rehabilitation course could affect this adversely.

U.S. national data indicates that those with MS do not have to wait as long to be subsidised as those with other neurological disorders, such as epilepsy. It may be that employers and health professionals were aware of this and by encouraging the MS population towards state assistance, were hindering the attempts of the vocational rehabilitation program. Conflicting themes have emerged from this particular piece of research, in that; highly motivated, well-educated, people are withdrawing from vocational rehabilitation programs. This could be due to the attitude of the individual or due to the external factors discussed above, however the clear message is that this population needs a more creative vocational rehabilitation approach due to the diverse symptoms of MS and the severity of the disability that can result.

A more flexible benefits system is also needed in the United Kingdom as it seems that the present system does not understand nor accommodate the relapsing remitting nature of the disease. Patient experience is that when benefits are put into place they are deemed to be there for a significant length of time. This means that when the person with MS recovers from the relapse and goes back to work, relinquishing their benefits at that stage, it would be very difficult to reclaim the benefits again without another lengthy assessment process if they were to relapse again and find themselves unable to work. If they do not get sick pay from their employer they can find themselves on statutory sick pay during this period which could cause marked financial stress. Understandably many people are unwilling to take this risk and so stay on benefits and do not attempt to go back to work. If the provision of benefits could be implemented and withdrawn with relapses and remissions the likelihood is that those people who wanted to work could do so knowing their financial security would be unaffected.

Many people with MS who were interviewed for the current study found it difficult to work full time due to their symptoms but would like to maintain some presence in the workplace but even as little as a few hours per week can adversely affect their benefits. People with MS who are in this position talk about work "not being worth my while" as the present inflexible system actively discourages part time, temporary work. The benefits system in the UK appears to view capacity to work with multiple sclerosis in black or white with no grey areas.

### 7.5 Directions for future research

There are several areas of future research following on from the current study. As mentioned above further validation studies on larger samples would be appropriate, including studies of responsiveness to support the scale's use as an outcome measure.

Adaptation of the scale into other languages would be appropriate; the RA-WIS is currently being adapted into several European languages. The adaptation of the scale may be made more complicated by the fact that some activities have marked cultural or literal aspects, "my job is physically impossible" for example. Therefore there is potential to look at Cross – Cultural Validity of the scale within the framework of the Rasch Model (Tennant et al, 2004).

There is also the potential to use the scale in English speaking countries, for example, the United States of America. Exploration of that area has already taken place. Collaborative work between the group at Leeds and academics based at the Centre for Disability Studies, Kent State University, USA and the Disability Research Institute, University of Illinois, USA, Rumrill and Roessler respectively is currently underway. Rumrill and Roessler have published extensively on the area of multiple sclerosis and employment and are highly regarded within the field. An experienced researcher from the University of Leeds travelled to the USA to carry out interviews in an American population of employed people with multiple sclerosis. This set of interviews will be analysed to see if the same themes and items arise regarding the issue of employment in multiple sclerosis. Depending upon the results, for example if the same items arise during item selection, then adaptation of the scale would not be necessary and the MS WIS could be administered to the American population.

In conclusion, apart from detailed work placement assessments that are undertaken within current services there is no existing standardised questionnaire based assessment that can easily capture the risk of job loss in multiple sclerosis. Therefore this piece of work does have significant potential for clinical benefit.

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# Appendix 1

## Fatigue Severity Scale (FSS)

### FSS Questionnaire

| **During the past week, I have found that:** | **Disagree <----> Agree** | | | | | | |
|---|---|---|---|---|---|---|---|
| My motivation is lower when I am fatigued. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| Exercise brings on my fatigue. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| I am easily fatigued. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| Fatigue interferes with my physical functioning. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| Fatigue causes frequent problems for me. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| My fatigue prevents sustained physical functioning. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| Fatigue interferes with carrying out certain duties and responsibilities. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| Fatigue is among my three most disabling symptoms. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| Fatigue interferes with my work, family, or social life. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| | | | **Total Score:** | | | | |

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### Scoring your results

Now that you have completed the questionnaire, it is time to score your results and evaluate your level of fatigue. It's simple: Add all the numbers you circled to get your total score.

### The Fatigue Severity Scale Key

A total score of less than 36 suggests that you may not be suffering from fatigue.

A total score of 36 or more suggests that you may need further evaluation by a physician.

# Appendix 2

## Kurtzke Expanded Disability Status Scale (EDSS)

☐ 0.0 - Normal neurological exam (all grade 0 in all Functional System (FS) scores*).

☐ 1.0 - No disability, minimal signs in one FS* (i.e., grade 1).

☐ 1.5 - No disability, minimal signs in more than one FS* (more than 1 FS grade 1).

☐ 2.0 - Minimal disability in one FS (one FS grade 2, others 0 or 1).

☐ 2.5 - Minimal disability in two FS (two FS grade 2, others 0 or 1).

☐ 3.0 - Moderate disability in one FS (one FS grade 3, others 0 or 1) or mild disability in three or four FS (three or four FS grade 2, others 0 or 1) though fully ambulatory.

☐ 3.5 - Fully ambulatory but with moderate disability in one FS (one grade 3) and one or two FS grade 2; or two FS grade 3 (others 0 or 1) or five grade 2 (others 0 or 1).

☐ 4.0 - Fully ambulatory without aid, self-sufficient, up and about some 12 hours a day despite relatively severe disability consisting of one FS grade 4 (others 0 or 1), or combination of lesser grades exceeding limits of previous steps; able to walk without aid or rest some 500 meters.

☐ 4.5 - Fully ambulatory without aid, up and about much of the day, able to work a full day, may otherwise have some limitation of full activity or require minimal assistance; characterized by relatively severe disability usually consisting of one FS grade 4 (others or 1) or combinations of lesser grades exceeding limits of previous steps; able to walk without aid or rest some 300 meters.

☐ 5.0 - Ambulatory without aid or rest for about 200 meters; disability severe enough to impair full daily activities (e.g., to work a full day without special provisions); (Usual FS equivalents are one grade 5 alone, others 0 or 1; or combinations of lesser grades usually exceeding specifications for step 4.0).

☐ 5.5 - Ambulatory without aid for about 100 meters; disability severe enough to preclude full daily activities; (Usual FS equivalents are one grade 5 alone, others 0 or 1; or combination of lesser grades usually exceeding those for step 4.0).

☐ 6.0 - Intermittent or unilateral constant assistance (cane, crutch, brace) required to walk about 100 meters with or without resting; (Usual FS equivalents are combinations with more than two FS grade 3+).

☐ 6.5 - Constant bilateral assistance (canes, crutches, braces) required to walk about 20 meters without resting; (Usual FS equivalents are combinations with more than two FS grade 3+).

☐ 7.0 - Unable to walk beyond approximately 5 meters even with aid, essentially restricted to wheelchair; wheels self in standard wheelchair and transfers alone; up and about in wheelchair some 12 hours a day; (Usual FS equivalents are combinations with more than one FS grade 4+; very rarely pyramidal grade 5 alone).

☐ 7.5 - Unable to take more than a few steps; restricted to wheelchair; may need aid in transfer; wheels self but cannot carry on in standard wheelchair a full day; May require motorized wheelchair; (Usual FS equivalents are combinations with more than one FS grade 4+).

☐ 8.0 - Essentially restricted to bed or chair or perambulated in wheelchair, but may be out of bed itself much of the day; retains many self-care functions; generally has effective use of arms; (Usual FS equivalents are combinations, generally grade 4+ in several systems).

☐ 8.5 - Essentially restricted to bed much of day; has some effective use of arm(s); retains some self-care functions; (Usual FS equivalents are combinations, generally 4+ in several systems).

☐ 9.0 - Helpless bed patient; can communicate and eat; (Usual FS equivalents are combinations, mostly grade 4+).

☐ 9.5 - Totally helpless bed patient; unable to communicate effectively or eat/swallow; (Usual FS equivalents are combinations, almost all grade 4+).

☐ 10.0 - Death due to MS.

*Excludes cerebral function grade 1.

Note 1: EDSS steps 1.0 to 4.5 refer to patients who are fully ambulatory and the precise step number is defined by the Functional System score(s). EDSS steps 5.0 to 9.5 are defined by the impairment to ambulation and usual equivalents in Functional Systems scores are provided.

Note 2: EDSS should not change by 1.0 step unless there is a change in the same direction of at least one step in at least one FS.

Sources: Kurtzke JF. Rating neurologic impairment in multiple sclerosis: an expanded disability status scale (EDSS). Neurology. 1983 Nov;33(11):1444-52.

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# Appendix 3

## Strategy for Pragmatic Literature Review

In order to explore relevant literature on work instability and its impact in MS, a literature review was undertaken. A pragmatic, iterative approach was adopted for reviewing the literature, in preference to a systematic review for two reasons:

- (i) there is limited literature on the impact of MS on work practices, with the exception of job-loss
- (ii) the concept of work instability is relatively new and a systematic search of specific terms was unlikely to result in a meaningful search of appropriate literature.

Pub Med and Medline were used to search English-language articles related to MS and work impact. Specifically, the following terms were used:

1. Multiple Sclerosis
2. Work in multiple sclerosis
3. Work loss in multiple sclerosis
4. Employment in multiple sclerosis
5. Unemployment in multiple sclerosis

6. Work in rheumatoid arthritis
7. Work loss in rheumatoid arthritis
8. Employment in rheumatoid arthritis
9. Unemployment in rheumatoid arthritis

10. Work in traumatic brain injury
11. Work loss in traumatic brain injury
12. Employment in traumatic brain injury
13. Unemployment in traumatic brain injury

From this search, relevant papers were identified, sourced and reviewed. From this process, the following additional search terms were identified:

1. Work instability in multiple sclerosis
2. Work disability in multiple sclerosis

3. Work instability in rheumatoid arthritis
4. Work disability in rheumatoid arthritis

5. Work instability in traumatic brain injury
6. Work disability in traumatic brain injury

In an iterative process, references used in these papers were also reviewed, with additional papers identified that would not have been included in the original search.

To ensure recent articles were included, this process was repeated once the data collection was completed.

The literature review was the combined into research themes, which formed the basis of Chapter 2.

# Appendix 4

## Topic List for "Work Instability in Multiple Sclerosis" interviews

- What job entails (take me through an average day)
- Does condition affect job?
- Do they use strategies to help this
- What are working conditions like – does working environment affect how they do their job?
- Could any improvements be made
- Has condition affected career progression or ambitions
- What is relationship with colleagues like, has this been affected by condition?
- Has continuing to work affected home life or relationships at home?
- Is there anything else that hasn't been discussed, that is important?

Appendix 5

# The Leeds MSQoL
# CONFIDENTIAL QUESTIONNAIRE

---

**Please tell us a little about yourself by answering the following questions.**

**Your day to day life may be affected in many different ways by your health. On the following pages you will find some questions asking about what you have been doing and how you have been feeling.**

**Please answer these questions as honestly as you can. It is important that your answers reflect how you have been feeling. There are no right or wrong answers - just say what you think best applies to you.**

**Please try to answer all the questions. Read the instructions for each section and just put a tick in the box which is closest to how you have been feeling.**

**If you have needed help in ticking the boxes please indicate this on the back page.**

## Please tick ONE answer for each statement

### THE FOLLOWING QUESTION IS FOR EXAMPLE ONLY.

*During the past month . . .*

1. **My health has affected my relationships with my family.**

*please tick √ one*

| | |
|---|---|
| not at all | |
| sometimes | √ |
| quite often | |
| most of the time | |

*During the past month . . .*

*please tick √ one*

1. **My health has affected my relationships with my family.**

| | |
|---|---|
| not at all | |
| a little | |
| quite a lot | |
| very much | |

2. **I have felt lonely.**

| | |
|---|---|
| most of the time | |
| very often | |
| occasionally | |
| not at all | |

3. **I have felt good about my appearance.**

| | |
|---|---|
| most of the time | |
| very often | |
| occasionally | |
| not at all | |

4. **I have worried about my health.**

| | |
|---|---|
| most of the time | |
| a lot of the time | |
| quite often | |
| occasionally | |

5. ***I have worried about other peoples attitudes***
**Towards me.**

| | |
|---|---|
| not at all | |
| occasionally | |
| often | |
| very often | |

***During the past month . . .***

*please tick √ one*

**6. I have felt tired.**

| | |
|---|---|
| most of the time | |
| a lot of the time | |
| quite often | |
| occasionally | |

**7. I have had as much energy as usual.**

| | |
|---|---|
| as much | |
| not quite as much | |
| definitely not as much | |
| not at all | |

**8. I have felt happy about the future.**

| | |
|---|---|
| most of the time | |
| quite often | |
| occasionally | |
| hardly at all | |

*LMSQoL © Dr Helen Ford*

**Thank you for taking the time to fill in this questionnaire**

# Appendix 6

## *The Social Function Scale*

*During the past month . . .*

1. **I have joined in family activities.**

| | |
|---|---|
| not at all | |
| sometimes | |
| quite often | |
| most of the time | |

2. **We have taken family holidays (weekends/day trips).**

| | |
|---|---|
| very often | |
| quite often | |
| occasionally | |
| not at all | |

3. **I have been out of the house.**

| | |
|---|---|
| very often | |
| quite often | |
| occasionally | |
| not at all | |

4. **I have taken part in sports or other outdoor activities.**

| | |
|---|---|
| not at all | |
| occasionally | |
| quite often | |
| very much | |

***During the past month . . .***

*please tick √ one*

**5.** **I have been out with friends.**

| | |
|---|---|
| very often | |
| quite often | |
| occasionally | |
| not at all | |

**6.** **I have spent evenings out with my partner or close friend.**

| | |
|---|---|
| frequently | |
| quite often | |
| occasionally | |
| not at all | |

**7.** **I have been able to make plans for the future (holidays/trips).**

| | |
|---|---|
| not at all | |
| occasionally | |
| quite often | |
| most of the time | |

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**Thank you for taking the time to fill in this questionnaire**

# Appendix 7

## MS IMPACT SCALE (MSIS-29)

- The following questions ask for your views about the impact of MS on your day-to-day life **during the past two weeks**
- For each statement, please **circle** the **one** number that **best** describes your situation
- Please answer **all** questions

| **In the past two weeks, how much has your MS limited your ability to ...** | Not at all | A little | Mod-erately | Quite a bit | Extreme-ly |
|---|---|---|---|---|---|
| **1. Do physically demanding tasks?** | 1 | 2 | 3 | 4 | 5 |
| **2. Grip things tightly (e.g. turning on taps)?** | 1 | 2 | 3 | 4 | 5 |
| **3. Carry things?** | 1 | 2 | 3 | 4 | 5 |

| **In the past two weeks, how much have you been bothered by ...** | Not at all | A little | Mod-erately | Quite a bit | Extreme-ly |
|---|---|---|---|---|---|
| **4. Problems with your balance?** | 1 | 2 | 3 | 4 | 5 |
| **5. Difficulties moving about indoors?** | 1 | 2 | 3 | 4 | 5 |
| **6. Being clumsy?** | 1 | 2 | 3 | 4 | 5 |
| **7. Stiffness?** | 1 | 2 | 3 | 4 | 5 |
| **8. Heavy arms and/or legs?** | 1 | 2 | 3 | 4 | 5 |
| **9. Tremor of your arms or legs?** | 1 | 2 | 3 | 4 | 5 |
| **10. Spasms in your limbs?** | 1 | 2 | 3 | 4 | 5 |
| **11. Your body not doing what you want it to do?** | 1 | 2 | 3 | 4 | 5 |
| **12. Having to depend on others to do things for you?** | 1 | 2 | 3 | 4 | 5 |

**Please check that you have answered all the questions before going on to the next page**

| In the **past two weeks**, how much have you been bothered by ... | Not at all | A little | Mod-erately | Quite a bit | Extreme-ly |
|---|---|---|---|---|---|
| 13. Limitations in your social and leisure activities at home? | 1 | 2 | 3 | 4 | 5 |
| 14. Being stuck at home more than you would like to be? | 1 | 2 | 3 | 4 | 5 |
| 15. Difficulties using your hands in everyday tasks? | 1 | 2 | 3 | 4 | 5 |
| 16. Having to cut down the amount of time you spent on work or other daily activities? | 1 | 2 | 3 | 4 | 5 |
| 17. Problems using transport (e.g. car, bus, train, taxi, etc.)? | 1 | 2 | 3 | 4 | 5 |
| 18. Taking longer to do things? | 1 | 2 | 3 | 4 | 5 |
| 19. Difficulty doing things spontaneously (e.g. going out on the spur of the moment)? | 1 | 2 | 3 | 4 | 5 |
| 20. Needing to go to the toilet urgently? | 1 | 2 | 3 | 4 | 5 |
| 21. Feeling unwell? | 1 | 2 | 3 | 4 | 5 |
| 22. Problems sleeping? | 1 | 2 | 3 | 4 | 5 |
| 23. Feeling mentally fatigued? | 1 | 2 | 3 | 4 | 5 |
| 24. Worries related to your MS? | 1 | 2 | 3 | 4 | 5 |
| 25. Feeling anxious or tense? | 1 | 2 | 3 | 4 | 5 |
| 26. Feeling irritable, impatient, or short tempered? | 1 | 2 | 3 | 4 | 5 |
| 27. Problems concentrating? | 1 | 2 | 3 | 4 | 5 |
| 28. Lack of confidence? | 1 | 2 | 3 | 4 | 5 |
| 29. Feeling depressed? | 1 | 2 | 3 | 4 | 5 |

**Please check that you have circled ONE number for EACH question**

# Appendix 8

## MODIFIED FATIGUE IMPACT SCALE (MFIS)

Following is a list of statements that describe how fatigue may affect a person. Fatigue is a feeling of physical tiredness and lack of energy that many people experience from time to time. In medical conditions like MS, feelings of fatigue can occur more often and have a greater impact than usual. Please read each statement carefully, and then circle the one number that best indicates how often fatigue has affected you in this way during the past 4 weeks. Please answer every question. If you are not sure which answer to select, please choose the one answer that comes closest to describing you.

**Because of my fatigue during the past 4 weeks....**

| | **Never** | **Rarely** | **Sometimes** | **Often** | **Almost always** |
|---|---|---|---|---|---|
| **1. I have been less alert** | **0** | **1** | **2** | **3** | **4** |
| **2. I have had difficulty paying attention for long periods of time** | **0** | **1** | **2** | **3** | **4** |
| **3. I have been unable to think clearly** | **0** | **1** | **2** | **3** | **4** |
| **4. I have been clumsy and uncoordinated** | **0** | **1** | **2** | **3** | **4** |
| **5. I have been forgetful** | **0** | **1** | **2** | **3** | **4** |
| **6. I have had to pace myself in my physical activities** | **0** | **1** | **2** | **3** | **4** |
| **7. I have been less motivated to do anything that requires physical effort** | **0** | **1** | **2** | **3** | **4** |
| **8. I have been less motivated to participate in social activities** | **0** | **1** | **2** | **3** | **4** |
| **9. I have been limited in my ability to do things away from home** | **0** | **1** | **2** | **3** | **4** |

Because of my fatigue during the past 4 weeks....

| | Never | Rarely | Sometimes | Often | Almost always |
|---|---|---|---|---|---|
| 10. I have had trouble maintaining physical effort for long periods | 0 | 1 | 2 | 3 | 4 |
| 11. I have had difficulty making decisions | 0 | 1 | 2 | 3 | 4 |
| 12. I have been less motivated to do anything that requires thinking | 0 | 1 | 2 | 3 | 4 |
| 13. My muscles have felt weak | 0 | 1 | 2 | 3 | 4 |
| 14. I have been physically uncomfortable | 0 | 1 | 2 | 3 | 4 |
| 15. I have had trouble finishing tasks that require thinking | 0 | 1 | 2 | 3 | 4 |
| 16. I have had difficulty organizing my thoughts when doing things at home or at work | 0 | 1 | 2 | 3 | 4 |
| 17. I have been less able to complete tasks that require physical effort | 0 | 1 | 2 | 3 | 4 |
| 18. My thinking has been slowed down | 0 | 1 | 2 | 3 | 4 |
| 19. I have had trouble concentrating | 0 | 1 | 2 | 3 | 4 |
| 20. I have limited my physical activities | 0 | 1 | 2 | 3 | 4 |

| | | | | | |
|---|---|---|---|---|---|
| **21. I have needed to rest more often or for longer periods ability to do things away from home** | **0** | **1** | **2** | **3** | **4** |

# Appendix 9

## First Draft of the MS WIS – 122 items

1. I push myself to keep working
2. I feel guilty about taking time off
3. I've got a terrible memory
4. I need to spread my work out
5. I worry about money
6. I don't have enough energy to do my job like I used to
7. As the day goes on my condition gets worse
8. My job is physically impossible
9. I need to take more rests during the day.
10. My speech problems affect my work
11. My speech is slurred sometimes
12. It takes me longer to get my words out
13. My condition affects my thinking
14. It slows me down in everything I do
15. I worry about the future
16. I have thought about changing my job
17. I feel stigmatised at work
18. People treat me differently
19. My condition is worse when I get stressed at work
20. I've thought about reducing my hours
21. I'm finding my job is about all I can manage
22. I can't talk to my work colleagues about my condition
23. I flag very quickly
24. I am very worried about my ability to keep working
25. Its affected my job prospects
26. Its affected me money wise
27. My condition is holding me back
28. Its frustrating because it is affecting my earnings
29. I'd like another job but I'm restricted to what I can do
30. I feel drained by the end of the day
31. I am finding things more and more difficult at work
32. I have trouble standing upright sometimes
33. There are some things I can't do any longer
34. Some days I find it hard to cope
35. I find my work exhausting
36. Its difficult to keep going some days
37. I really have to concentrate on what I am doing

38. I fall over easily
39. It affects my eyesight
40. I don't have the stamina to work like I used to
41. I have to pace myself
42. I feel guilty about getting others to help me
43. It takes me longer to do some things at work
44. When I get in from work I'm absolutely shattered
45. I find it difficult to control my bladder
46. My self confidence has gone
47. Its affecting my walking
48. It restricts where I can go
49. I have to plan my day more
50. I don't like to ask for help
51. I'm concerned about my memory
52. It is affecting the feeling in my hands
53. Steps and stairs are a struggle for me
54. Its changed the way I do some things at work
55. I'm less steady when I'm tired
56. I have trouble concentrating
57. My balance is poor
58. My hands are clumsy now
59. My vision is affected
60. Standing is very painful
61. I am constantly aware of my limitations
62. I fall asleep at work
63. My colleagues are not supportive
64. My employers are not supportive
65. Tiredness affects my concentration
66. Financially, I can't afford to stop working
67. I am constantly thinking about my condition
68. My colleagues don't understand
69. I don't like it when people treat me differently
70. Its painful walking
71. Getting to the toilet can be difficult at work
72. As the day goes on I get more tired
73. I have used my holiday so that I don't have to go sick
74. My hands don't seem to work properly
75. I make more mistakes when I am tired
76. I bump into things really easily
77. My words don't come out as I want them to
78. I can't think as quickly as I used to

79. I am tired all the time
80. Getting around at work is hard for me
81. I'm in pain most of the time
82. I can't stand for long periods of time
83. My condition restricts what I can do
84. I worry about people finding out at work
85. I get short tempered more easily
86. I struggle on rather than ask for help
87. I get mentally tired
88. Sometimes in the afternoon I can get really, really tired
89. I need more flexibility at work
90. My work suffers when I am tired
91. I feel I need to prove myself more at work
92. My employer has stopped me doing certain things
93. Its reduced my effectiveness to do my job

94. Its getting more and more difficult to walk
95. I get double vision
96. I push myself to go to work because I don't want to give in to my condition
97. It gets me down
98. I'm struggling at work
99. If I don't reduce my hours I may have to give up work
100. It's always there in the back of your mind
101. I have to be careful not to overdo it at work
102. I'm less confident now
103. I have difficulty remembering details
104. The building I work in makes things harder
105. I'm always dropping things
106. Its difficult reaching up
107. I can't remember important numbers like phone numbers
108. I'm more forgetful now
109. I don't like relying on other people
110. I can't do my job as well as I would like to
111. I get irritated more easily
112. Its hard when your condition is so unpredictable
113. When I can't do as much if affects my colleagues
114. Sometimes I can't face being at work all day
115. My condition is worse if I get too hot at work
116. I have to rely on other people for some parts of my job
117. My writing is affected

118. I am more tired than I used to be
119. I have to make notes now and write things down
120. I am not as physically strong as I used to be
121. I can't do things that I used to be able to do
122. I don't feel physically well enough to do my job

# Appendix 10

## Second Draft of the MS WIS – 120 items

1. I push myself to keep working
2. I feel guilty about taking time off
3. I've got a terrible memory
4. I worry about money
5. I don't have enough energy to do my job like I used to
6. As the day goes on my condition gets worse
7. My job is physically impossible
8. I need to take more rests during the day.
9. My speech problems affect my work
10. My speech is slurred sometimes
11. It takes me longer to get my words out
12. My condition affects my thinking
13. It slows me down in everything I do
14. I worry about the future
15. I have thought about changing my job
16. I feel stigmatised at work
17. People treat me differently
18. My condition is worse when I get stressed at work
19. I've thought about reducing my hours
20. I'm finding my job is about all I can manage
21. I can't talk to my work colleagues about my condition
22. I flag very quickly
23. I am very worried about my ability to keep working
24. Its affected my job prospects
25. Its affected me money wise
26. My condition is holding me back
27. Its frustrating because it is affecting my earnings
28. I'd like another job but I'm restricted to what I can do
29. I feel drained by the end of the day
30. I am finding things more and more difficult at work
31. I have trouble standing upright sometimes
32. There are some things I can't do any longer
33. Some days I find it hard to cope
34. I find my work exhausting
35. Its difficult to keep going some days
36. I really have to concentrate on what I am doing
37. I fall over easily

38. It affects my eyesight
39. I don't have the stamina to work like I used to
40. I have to pace myself
41. I feel guilty about getting others to help me
42. It takes me longer to do some things at work
43. When I get in from work I'm absolutely shattered
44. I find it difficult to control my bladder
45. My self confidence has gone
46. Its affecting my walking
47. It restricts where I can go
48. I have to plan my day more
49. I don't like to ask for help
50. I'm concerned about my memory
51. It is affecting the feeling in my hands
52. Steps and stairs are a struggle for me
53. Its changed the way I do some things at work
54. I'm less steady when I'm tired
55. I have trouble concentrating
56. My balance is poor
57. My hands are clumsy now
58. My vision is affected
59. Standing is very painful
60. I am constantly aware of my limitations
61. I fall asleep at work
62. My colleagues are not supportive
63. My employers are not supportive
64. Tiredness affects my concentration
65. Financially, I can't afford to stop working
66. I am constantly thinking about my condition
67. My colleagues don't understand
68. I don't like it when people treat me differently
69. Its painful walking
70. Getting to the toilet can be difficult at work
71. As the day goes on I get more tired
72. I have used my holiday so that I don't have to go sick
73. My hands don't seem to work properly
74. I make more mistakes when I am tired
75. I bump into things really easily
76. My words don't come out as I want them to
77. I can't think as quickly as I used to
78. I am tired all the time

79. Getting around at work is hard for me
80. I'm in pain most of the time
81. I can't stand for long periods of time
82. My condition restricts what I can do
83. I worry about people finding out at work
84. I get short tempered more easily
85. I struggle on rather than ask for help
86. I get mentally tired
87. Sometimes in the afternoon I can get really, really tired
88. I need more flexibility at work
89. My work suffers when I am tired
90. I feel I need to prove myself more at work
91. My employer has stopped me doing certain things
92. Its getting more and more difficult to walk
93. I get double vision
94. I push myself to go to work because I don't want to give in to my condition
95.It gets me down
96. I'm struggling at work
97. If I don't reduce my hours I may have to give up work
98. It's always there in the back of your mind
99. I have to be careful not to overdo it at work
100.I'm less confident now
101.I have difficulty remembering details
102.The building I work in makes things harder
103.I'm always dropping things
104.Its difficult reaching up
105.I can't remember important numbers like phone numbers
106.I'm more forgetful now
107.I don't like relying on other people
108.I can't do my job as well as I would like to
109.I get irritated more easily
110.Its hard when your condition is so unpredictable
111.When I can't do as much if affects my colleagues
112.Sometimes I can't face being at work all day
113.My condition is worse if I get too hot at work
114.I have to rely on other people for some parts of my job
115.My writing is affected
116.I am more tired than I used to be
117.I have to make notes now and write things down
118.I am not as physically strong as I used to be

119.I can't do things that I used to be able to do
120.I don’t feel physically well enough to do my job

# Appendix 11

## The 21 –Item Multiple Sclerosis Work Instability Scale (MS - WIS)

Please read each statement thinking about your Multiple Sclerosis.
Please choose the response that applies to you *at the moment:*

| | TRUE | NOT TRUE |
|---|---|---|
| 1. I push myself to keep working | ☐ | ☐ |
| 2. I don't have enough energy to do my job like I used to | ☐ | ☐ |
| 3. As the day goes on my condition gets worse | ☐ | ☐ |
| 4. My job is physically impossible | ☐ | ☐ |
| 5. People treat me differently | ☐ | ☐ |
| 6. There are some things I can't do any longer at work | ☐ | ☐ |
| 7. I have to pace myself | ☐ | ☐ |
| 8. I feel guilty about getting others to help me | ☐ | ☐ |
| 9. It takes me longer to do some things at work | ☐ | ☐ |
| 10. I don't like to ask for help | ☐ | ☐ |
| 11. It is affecting the feeling in my hands | ☐ | ☐ |
| 12. My hands are clumsy now | ☐ | ☐ |
| 13. My employers are not supportive | ☐ | ☐ |
| 14. Its painful walking | ☐ | ☐ |
| 15. My hands don't seem to work properly | ☐ | ☐ |
| 16. Sometimes in the afternoon I can get really, really tired | ☐ | ☐ |

17. I push myself to go to work because I don't want to give in to my condition ☐ ☐

18. If I don't reduce my hours I may have to give up work ☐ ☐

19. I have to be careful not to overdo it at work ☐ ☐

20. I have to rely on other people for some parts of my job ☐ ☐

21. I am more tired than I used to be ☐ ☐

Please check you have ticked a box for every statement on this page

# Appendix 12

## Working yet worried: Development & evaluation of a model of Occupational Rehabilitation for people with Multiple Sclerosis

**Abstract:**

Work contributes to adult identity, confers financial benefit and status and can improve quality of life. At diagnosis most people with Multiple Sclerosis are in full time education or employment, but within ten years most have left work. The reasons for this are multiple but include limited recognition of work difficulties by health care professionals and services which are perceived as inappropriate or difficult to access. There has been limited research into this area. We need to know what people with MS feel would be most helpful, how that service should be promoted and delivered and whether such services are effective and cost efficient. This preliminary study aims to address these issues using a combination of qualitative and quantitative approaches. The main outcome from the study will be a service model with supporting materials that maybe used in a variety of settings. In addition the study will identify the most appropriate outcome measures for evaluating the effectiveness of a vocational rehabilitation service and a costing system, thus facilitating cost benefit analysis as well as providing data for the design of future studies.

## Aim of the study

The aim of this study was to define the intervention in practice in terms of content and cost, and to identify which outcomes captured the impact of the intervention. We hypothesized that the intervention would not impact on measures of physical disability, but would influence measures of well being and measures of work ability.

## Methodology

### Participants

Participants were recruited from the National Hospital for Neurology and Neurosurgery (NHNN) University College London Hospital NHS Foundation Trust, London, UK. This hospital delivers a comprehensive MS service covering all stages of the disease from diagnosis to the management of severe disability. Inclusion criteria were as follows: a definite diagnosis of MS; in employment or education within the past 6 months; clinically stable; and functional spoken English. Ethical approval for the study was obtained from the joint research ethics committee of the NHNN and the Institute of Neurology.

Twenty-three people (16 female, mean age 40 years, range 24-63 years) were recruited to the study. Fifteen participants had relapsing remitting MS, two secondary progressive MS and six primary progressive MS; the mean duration of MS was 6.3 years. Fifteen of the patients were university educated.

## Intervention

Each participant was comprehensively assessed by a senior neurological occupational therapist (OT) experienced in vocational rehabilitation. The initial assessment was office based and involved obtaining a description from the patient of their education and training, the tasks they performed at work, and the physical and social environment within which they worked. In addition during this assessment the therapist worked with the individual to identify their strengths and weaknesses, define problems and solutions by identifying the various components of the tasks, the various factors that contributed to any difficulties and the individual's work-related goals.

Potential approaches to problems included firstly improving performance e.g., through fatigue management or physiotherapy, secondly compensating for impaired performance e.g., removing environmental barriers, improving the ergonomics of work stations, and thirdly modifying performance e.g. by working with employer to reduce the demands of the task e.g., by structuring the day by setting aside specific times for different elements of the work. Follow-up sessions included the provision of advice about and modeling disclosure, information about the disability discrimination act, work planning including work place accommodations and information about Access to Work.

After each session an action plan was developed for both OT and client. Each participant had access to as many treatment sessions as were necessary to complete the plan; where appropriate referrals were made to other rehabilitation professionals including physiotherapists and neuro-psychologists.

### Cost data

For each participant both contact and non-contact time was recorded describing both the task performed and the time taken to undertake the task. In addition the time spent by other health care professionals was also recorded. The cost of the intervention was calculated as the product of total time spent and mean hourly rates defined in The Units Costs of Health and Social Care Report. 95% confidence intervals were calculated using a bootstrap method (10,000 resamples, with replacement). Measures of work performance, health status and wellbeing were recorded both at recruitment and completion of the intervention.

## Outcome measures

Work status was measured using: (1) The MS impact on work questionnaire which identifies barriers to work caused by symptoms and by the environment (2) The MS work assessment scale (3) The work limitations questionnaire, these three scales have been shown be reliable and valid but no studies of responsiveness have been performed, (4) The MS work instability scale (Mc Fadden et al, Personal Communication). Work instability is the mismatch between an individual's functional abilities and the demands of his or her job. This scale is under development, and preliminary work suggests it is valid and reliable. All patients were asked to complete a seven point transition question after completion of the study asking how much change there had been in their ability to cope with the demands of the work place with potential answers including from much worse, somewhat worse, slightly worse, no change, slightly better, somewhat better, much better.

Health status was measured using the self report Barthel Index (BI), the Multiple Sclerosis Impact Scale (MSIS -29) and the Multiple Sclerosis Walking scale (MSWS-12). Well-being was measured using the Medical Outcomes Survey Short Form 36 (SF-36) and the General Health Questionnaire (GHQ). These five scales are well-established with good validity and reliability. In addition the Occupational well-being scale, a scale under development, designed to measure well-being in the work-place was also recorded [Doble – personal communication].