

**THE MOTHERHOOD CHOICE:
DEVELOPMENT AND EVALUATION OF A
DECISION AID FOR WOMEN
WITH MULTIPLE SCLEROSIS**

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Presentation of Thesis

This thesis is presented as a series of **published/submitted** manuscripts. The first chapter contains a review of the literature, introducing the research rationale, aims and hypotheses. Chapters two, three, four and five contain the following manuscripts. The candidate is the principle author of each of these papers.

- Chapter Two: Sponiar, M.C., Sharpe, L., Butow, P., Fulcher, G. (2007)
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- Chapter Five: The Motherhood Decision Aid: Mechanisms of Treatment.
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The final chapter provides a summary and conclusions of the research findings, limitations and suggestions for the direction of future research.

The study presented in this thesis represents research undertaken by the candidate in conjunction with other researchers in the Clinical Psychology Unit and the Medical Psychology Research Unit at the University of Sydney, and the Multiple Sclerosis

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The candidate was involved in all aspects of the study, and was responsible for coordinating the study under the supervision of Associate Professor Louise Sharpe, and associate supervision of Professor Phyllis Butow and Dr Gary Fulcher.

The contributions of the candidate include:

Study design: The candidate was responsible for identifying the topic, recruiting the study sample beginning with the large-scale mail-out to 1410 female members of the MS Society in New South Wales and Victoria, Australia. The candidate designed the initial preliminary decision aid and ran the focus groups in the qualitative study. The candidate selected the questionnaire measures and compiled the demographic questions and the knowledge questionnaire.

Data collection, entry and analyses: The candidate was responsible for all aspects of data collection, entry and analyses. The candidate formulated the hypotheses. All data presented in this thesis was analysed by the candidate under the guidance of Associate Professor Louise Sharpe.

Manuscripts: The candidate was the principle author and wrote all first drafts of the manuscripts presented in this thesis, responsible for the conceptualisation and the interpretation of the data, under the supervision of Associate Professor Louise Sharpe and Professor Phyllis Butow.

Abstract

Multiple sclerosis (MS) is the most common neurological disease affecting young adults. MS affects approximately 1 in 1000 people and, like other autoimmune diseases, women are more likely to be affected than men. The illness typically onsets between the ages of 20 and 40, and hence usually affects women of child-bearing age. The course of the MS is often unclear for years after diagnosis and since most women are diagnosed in their child-bearing years, they often have to make reproductive choices before their prognosis is clear and while the future remains uncertain.

For women with MS, starting a family is an individual choice that needs to balance the importance of motherhood for the woman and her partner against the risks that she will be unable to care for the infant or child as a result of increasing disability. In other areas of medicine where finely balanced decisions are required, there has been a recent proliferation of decision aids that aim to inform people of the benefits and risks of opposing courses of action. In addition, decision aids help patients to weigh their values against the risks and benefits to make an informed decision. Despite the existence of over 200 decision aids to help patients consider decisions related to their medical conditions, not one exists that deals with the decision of whether or not to have a family for women with a chronic disability, such as MS.

This thesis developed and evaluated a decision aid for women with MS to help them decide whether to start, forego or enlarge their families. The study utilised the criteria set out for the development of decision aids, according to the Cochrane Systematic Review of Patient Decision Aids (O'Connor et al., 2003). The first aim was to determine the proportion of women who are undecided about the motherhood choice

and for whom a decision aid may be relevant. Results found that the motherhood choice was relevant to 46% of the women who responded to an initial mail-out.

The second study aimed to establish women's current concerns and thoughts regarding pregnancy and motherhood, and their response to the pilot decision aid. Twenty women participated in qualitative interviews and results supported previous findings that the mother's health concerns, coping with parenting and societal attitudes are significant concerns when considering this decision. This study further identified concerns from different groups that had a direct impact on the decision to have children, including the experience of parenting, the child's well-being and the timing and pressure of the decision.

The main study was a randomised controlled trial of the decision aid aiming to determine whether the decision aid facilitated decision-making in women with MS. The study confirmed that the decision aid presented a balanced view to women, increased knowledge, reduced decisional conflict, increased decisional self-efficacy and certainty of the decision, and was free from adverse effects on psychopathology.

The final component of the study was a 12 month follow-up which aimed to explore the long-term effectiveness of the decision aid and what aspects were valued by the women who received it. It was found that over time, women in the intervention group did maintain their certainty, but women in the control group also became more certain of their choice. At follow-up, the difference in certainty was no longer significant between the two groups. However, women did report that the intervention was useful in (a) providing access to information previously unavailable or difficult to obtain, (b)

facilitating communication between women, their partners and health care professionals, (c) aiding them in considering and utilising their networks of support, and (d) preparing them for potential difficulties.

In summary, this thesis developed and evaluated a decision aid for women with MS who are considering motherhood. The results showed that many women were undecided and, in the absence of good information on the topic, many women had concerns about pregnancy and parenthood. The decision aid was shown to be effective across a range of measures and free from adverse psychological effects. Hence, this evidence-based resource can now be recommended for those women with MS who are currently contemplating motherhood.

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CHAPTER 1: INTRODUCTION

1.1 Introduction and Overview

Multiple Sclerosis (MS) is a chronic neurological disorder of the central nervous system affecting approximately 1 in 1000 people. It is more common among women and its onset is typically in the childbearing years of the 20s and 30s (Birk & Kalb, 1992, Cook, Troiano, Bansil & Dowling, 1994). Despite advances in the medical treatment of MS, it remains a serious illness that is associated with progressive disability for most individuals with MS. Due to the progressive and unpredictable nature of MS, coupled with the time of onset being one associated with family planning, women with MS are often faced with decisional conflict regarding the motherhood decision (Smeltzer, 2002). The already physically and emotionally demanding time of pregnancy and childbearing may be exacerbated by a potentially disabling illness. However, it has been shown that generally the course of MS remains unaffected by pregnancy in the long term and there are no higher rates of birth defects or miscarriage (Watkiss & Ward, 2002), although the first 3 to 6 months postpartum are often a time associated with increased relapse rates (Cook, Troiano, Bansil, & Dowling, 1994) as well as being a physically and emotionally stressful period. Therefore, the presence of MS means there are additional factors to consider in the decision-making process. Only when women are aware and informed of the various issues involved can an informed decision be made.

Motherhood is a valued social role for the majority of women. The motherhood decision can be defined as the choice to forego, start or enlarge a family (Smeltzer, 2002). For

women with MS, there are a number of issues that need to be considered as part of the motherhood decision. Four specific factors about which women need to be informed to make informed choices have been identified (McNary, 1999), as follows: (a) the neuro-physical features of MS; (b) the psychological consequences of MS; (c) the culture of each individual woman and her family; and (d) the historical context in which women with disabilities contemplate motherhood. These themes have been found to influence women in their reproductive choices and to determine the ease with which women and their partners are able to make this decision.

Although in recent years, there has been an increase in the amount of research on the physical consequences of pregnancy and childbirth on the health of women with MS and their infants, there is a paucity of research on how women reach their decision. What literature is available suggests that women with MS more often choose voluntary termination of pregnancy (Mueller, 2002) and are over-represented amongst childless women (Damek & Shuster, 1997). Research also suggests that women with MS continue to experience negative reactions towards their decision to have a child from society and health care professionals and have difficulty accessing good information to allow informed decision-making (Grue & Laerum, 2002; Thomas, 1997).

This thesis aims to bridge the gaps in the literature by (a) examining the number of women experiencing uncertainty regarding the motherhood choice; (b) understanding the reasons why different women choose to have families or to forego them, and (c)

developing and evaluating a decision aid to provide women with MS with help in making informed choices regarding motherhood.

1.2 Physical Characteristics of Multiple Sclerosis

Symptoms of MS are caused by demyelination in the central nervous system (CNS) and include muscle weakness, lack of coordination and balance, numbness and tingling, visual disturbance, tremors, spasticity and dysfunction of voiding and bladder. These symptoms frequently lead to decreases in quality of life, disturbances in psychological functioning and changes in social role functioning (Hart, Fonareva, Merluzzi & Mohr, 2005). Therefore, MS is associated with functional limitations in self-care and independent living. Because MS is characterised by progressive disability which is unpredictable, it is difficult to envisage the course and prognosis of the illness medically and physically for each individual. As a result it is difficult to feel in control of daily living and planning for the future, subsequently compromising the family planning process as it is difficult to consider the possibility of significant disability or ongoing fatigue problems in the years to come (Birk & Kalb, 1992).

There are a number of courses of MS that have been identified and the course varies considerably, with some patients experiencing a progressive course from the outset, and others following a more benign course. At present, there are five identified courses of MS:

1.2(a) Relapse-Relmitting MS

This is the most common course of MS and it is characterised by periods of relapse from which the person partially or completely recovers, and illness which may be inactive for long periods of time. Approximately 80% of people diagnosed with MS begin their course with this pattern of symptoms. Of those initially diagnosed with relapse-remitting MS, for 50% the disease course changes to secondary-progressive MS within ten years and for 90% of patients within 25 years (Ferrero, Pretta, & Ragni, 2004).

1.2(b) Secondary-Progressive MS

As described above, the majority of people with relapse-remitting MS will eventually develop secondary-progressive MS. People with secondary-progressive MS commonly experience an initial period of the relapse-remitting course, followed by steady progression of the disease with occasional relapses (Lechtenberg, 1995). Patients may experience less recovery from attacks, a decline in functioning both during and between attacks and/or fewer attacks with progressive disability.

1.2(c) Progressive-Relapsing MS

In contrast to those with relapse-remitting MS, those with this course of MS typically experience continuing disease progression between periods of clear acute relapse with or without full recovery. This is the least common course of MS with only 5% of people fitting this pattern of the illness at diagnosis.

1.2(d) Primary-Progressive MS

Primary-Progressive MS also has a slow onset, but a continual progression of disability either without remissions, or with minor improvements that are only temporary. Acute attacks are not generally experienced, rather occasional plateaus in disease activity. Ten percent of people with MS begin with this course, however diagnosis is usually made after a period of time where progressive disability has been ongoing without remission.

1.2(e) Benign MS

A frequent definition used to define benign MS is where there is “minimal or no disability, equivalent to a score on the Expanded Disability Status Scale (EDSS) equal to or less than 3.0 (fully ambulatory), at least 10 years after disease onset” (Glad, Nyland & Myhr, 2006, p. 55). Prevalence studies have varied in their results, with the frequency of benign MS ranging from between 10 and 40%, depending on the follow-up time. After 10 years, 40% of people with MS have a benign course, however this drops to 20% after 20 years, suggesting that even benign MS may be a transient phase (Glad, Nyland, & Myhr, 2006; Hawkins & McDonnell, 1999; Weinshenker, Bass, & Rice, 1989).

Half of those diagnosed with MS will need a walking stick for mobility within 15 years of diagnosis (Weinshenker, Hader, Carriere, Baskerville, & Ebers, 1989). However, the course of the MS is often unclear for years after diagnosis. For example, it can be ten

years after diagnosis before a patient can be said with certainty to have a benign course of the illness.

Disease progression is typically assessed radiographically via Magnetic Resonance Imaging (MRI) highlighting any new lesions, examining changes neurologically and electrophysiologically and by assessment of physical and cognitive functioning. It is difficult to determine prognosis on an individual basis, but the National MS Society Information Sourcebook (2005) states that those who do better tend to have fewer initial attacks in the years after diagnosis, longer periods between attacks and complete recovery from attacks. Furthermore, attacks of the sensory type, for example being marked by symptoms such as numbness or tingling, are associated with better long-term prognosis. On the other hand, early tremor, being unco-ordinated and having trouble walking are signs that the disease may be more progressive in nature. Progression is also marked by more frequent attacks without complete recovery and a higher number of lesions detected on MRI.

1.3 Neuropsychological Characteristics of Multiple Sclerosis

In addition to the physical complications of MS, the disease is often associated with neuropsychological deficits. Studies investigating the specific neuropsychological profiles among the subtypes of MS have found some significant differences between the various courses of the illness. One study investigating the differences in cognitive impairment of relapsing remitting, secondary and primary progressive MS (Huijbregts, Kalkers, de Sonneville et al., 2004) found that all MS groups had detectable cognitive

deficits, but different cognitive profiles. The most severe deficits existed on all tasks in those with secondary progressive MS, and significantly more deficient in tasks requiring higher-order working memory, except where speed of information processing played an important role. Primary progressive patients were the next most impaired, also performing poorer than controls on all tasks, and than those with relapse remitting MS who only differed from controls on three out of five tasks. People with relapse remitting MS were impaired on higher-order working memory tasks and showed relatively poor verbal fluency (Huijbregts, Kalkers, Sonnevile et al., 2004).

In a sample of 108 patients with relapse-remitting MS, 71 patients with secondary-progressive MS, 55 patients with primary-progressive MS and 67 healthy controls who underwent neuropsychological assessment, they found that deficits were most severe in secondary-progressive MS patients, followed by primary-progressive MS and then relapse-remitting MS patients. They also found evidence that there is a heterogeneity in the picture of cognitive impairment amongst the different courses of MS. Relapse-remitting MS patients were found to perform significantly better than primary-progressive MS and secondary-progressive MS patients on tests requiring specific working memory operation which also required central processing speed. It is thought that the more widespread white matter disease evident in the progressive types of MS results in deficient processing speed. On tasks requiring working memory ability with less emphasis on processing speed, primary-progressive MS patients performed better than those with secondary-progressive MS and relapse-remitting MS, explained by the

spatial working memory deficits in secondary-progressive MS and relapse-remitting MS (Gaudino, Chiaravalloti, DeLuca, & Diamond, 2001).

One of the most common complaints among patients with MS is a decline in memory, however some aspects of memory are affected more than others. Patients' ability to register and store information has been reported to be intact in MS patients that are not experiencing an exacerbation or chronic progressive deterioration, while their ability to recall information is significantly reduced (Beatty & Scott, 1993). This significantly impaired recall contrasts with normal performance in recognition, suggesting that the deficiency lies in the retrieval of information they have learned, rather than the initial acquisition of information. This hypothesis is supported by a study investigating the acquisition and storage deficits in MS, which reported that patients require more trials to learn verbal information, although once acquired, they are able to recall and recognize the information to the same extent as healthy controls (De Luca, Gaudino, Diamond, Christodoulou & Engel, 1998). The same study found a slightly different pattern with visual information whereby MS patients also required more trials to learn visual information yet also performed significantly worse than controls at both recall and recognition, thus suggesting deficiencies in both acquisition and storage of visual information.

Information processing speed is another domain which has been found to be impaired among patients with MS. It is an impairment that manifests globally within neuropsychological functioning as opposed to being associated with a few tasks or specific conditions, and has been likened to the general cognitive slowing that

accompanies the ageing process. Moreover, information processing speed has been found to influence other cognitive domains such as abstract reasoning and working memory (Kail, 1998). Specifically, MS patients have been noted to have a slower response speed and tend to process visual information at a reduced rate when compared to healthy controls particularly in more complex task conditions (De Sonneville et al., 2002).

Studies measuring attentional ability amongst people with MS have also found deficits in various attentional domains. Significantly reduced performance has been found on measures of focused, divided and sustained attention, with the performance of patients with primary-progressive MS being significantly lower than those with secondary-progressive MS (Bobholz & Rao, 2003). This manifests in MS patients being more susceptible to distraction and irrelevant targets and exhibiting a slowed response speed on tasks of attentional flexibility (De Sonneville et al., 2002).

MS is associated with axonal loss in addition to the inflammation and demyelination of the CNS. This demyelination and axonal degeneration causes interruption between neural connections (Piras, et al., 2003). While some research suggests that cognitive deficits become worse as a function of disease duration (Schiffer & Caine, 1991), axonal loss can occur early on in the disease and can therefore explain the instance of cognitive impairment during these initial stages. It has been demonstrated that despite few physical symptoms, decline in cognitive functioning can occur early in the course of the illness with impaired performance on tests of executive functioning and working memory (Barak, Lavie, & Achiron, 2002).

Fatigue has been acknowledged as a detrimental factor in neuropsychological test performance and is also one of the common disabling complaints among patients with MS. Studies on the effects of fatigue on cognitive functioning have produced mixed results. Early studies reported that fatigue has a negative impact on cognitive functioning (Lezak, 1995). However more recent studies differing in experimental design reported that fatigued patients with MS have not been found to have impaired performance in comparison to healthy controls (Johnson, Lange, De Luca, Korn, & Natelson, 1997). Another study concluded that there were no differences in test performance when patients were tested during periods of high versus low fatigue (Parmenter, Denney, & Lynch, 2003), however it has also been purported that MS-related fatigue may have a systematic effect on performance, manifesting itself in later tasks (De Sonneville et al., 2002). Subjective feelings of fatigue have been found to be a poor indicator of actual cognitive ability, with performance either remaining stable from baseline to retest or improving, thus illustrating a normal practice effect (Beatty et al., 2003). This study also reported that MS patients show greater increase in subjective fatigue over the course of a normal day, yet do not exhibit greater cognitive fatigue. However it should also be noted that a third of the patients in this study were taking anti-fatigue medications, which can alleviate fatigue or improve mood so it is difficult to ascertain how this influenced their performance (Beatty et al., 2003).

It has also been suggested that prominent symptoms of depression, for example feelings of inferiority and failure, contribute to deficits in information processing speed and

executive function linked to working memory, and that negative automatic thoughts reduce the individual's attention and working memory ability (Feinstein, 1999). Although there is some disagreement in the literature regarding the exact nature and cause of cognitive impairment in MS, studies consistently show that 40-60% of people with MS experience clinically significant cognitive impairments (Huijbregts, Kalkers, De Sonneville et al., 2004). These are more pronounced amongst those with primary-progressive MS and tend to become more severe over time. High levels of cognitive impairment in patients with MS is likely to result in an inability to maintain employment, fewer social support networks and a greater need for support with activities of daily living (Mohr & Cox, 2001) and thus the presence of cognitive impairment in MS adds to the hardship experienced by these patients.

1.4 Psychological Consequences of Multiple Sclerosis

Not only is MS associated with physical and neuropsychological consequences, but MS has also been associated with a number of symptoms of psychological distress, disturbances in personality and increased prevalence of psychiatric disorders (Feinstein, 1999). The unpredictability of MS is one of the most psychologically challenging aspects of MS and patients with the illness commonly experience both anxiety and depression. Given the fact that each patient's outcome remains largely unclear with the threat of increased disability, it seems understandable that people with MS experience depression. Indeed depression is the most common psychiatric illness amongst people with MS (Gorman & Finkel, 2005).

Interestingly however, the lesions found in patients with MS occur in a number of brain regions associated with depression (Gorman & Finkel, 2005). As a result, there is a controversy in the literature concerning whether these disorders are by-products of the illness, reactions to medications used to manage the physical symptoms, or psychological reactions to the experience of the disease (Minden, 2000). It is possible that MS plaques interrupt the neural pathways involved in the regulation of mood. Disruption in white matter affecting axonal-neuronal conduction may be linked to a presentation resembling vascular depression (Gorman & Finkel, 2005). In a study assessing 95 patients with MS, 18 met the criteria for major depression and it was found, when comparing the regional brain volumes and lesion loads of the depressed and non-depressed, that major depression was correlated with right frontal lesion load and right temporal brain volume and that severity of symptoms correlated with total temporal volume and right hemisphere volume (Zorzon et al, 2001). Of course, this does not exclude the possibility that these people with MS had more cognitive impairment and that cognitive impairment is the cause of the depression. In a more recent study using MRI to compare 21 depressed MS patients with 19 non-depressed MS patients, it was found that there were more hyperintense lesions in the left inferior medial frontal regions and greater atrophy of left anterior temporal regions in the depressed MS patients and that these two regions accounted for 42% of the variance in depression (Feinstein, Roy, & Lobaugh, 2004)

On the other hand it is clear that psychosocial factors also contribute to depression. Patients often report that their depression is due to the unpredictability of the illness (Beatty, 1993). Specifically, that patients' loss of function is unpredictable and often

implacable. Patients with MS who are depressed, typically present as angry, irritable, worried and with a sense of hopelessness about the future (Beatty, 1993). In fact, the lifetime prevalence of major depressive disorder following a diagnosis of MS is approximately 50% and the lifetime rate of suicidal intent in patients with MS is considerably higher than that for the general population (Feinstein, 2002). Of concern is that despite the common occurrence of suicidal ideation amongst patients with MS, depression often goes undetected and untreated (Feinstein, 1999).

Research has suggested that psychosocial factors play a significant role in whether a person will develop depression, with coping style and social support receiving the most attention (Siegert & Abernethy, 2005). Studies on coping with MS have suggested that optimism as well as problem-focused and cognitive coping strategies predict better adjustment to the illness (Aikens, Fischer, Namey & Rudick, 1997; de Ridder, Schreurs & Bensing, 2000; Pakenham, 1999). These may include positive reframing, finding ways of compensating for the disability or maintaining health and goal setting. On the other hand, emotion focused coping or avoidance and escape have been associated with higher symptom levels (Siegert & Abernethy, 2005).

What has also been found to influence adjustment to MS is the perception the patient has of the illness, or their illness representation. Illness representations influence patients' affective psychological adjustment as well as fatigue levels and it has been suggested that addressing aspects of illness beliefs may strengthen patients' ability to adjust to an MS diagnosis (Jopson & Moss-Morris, 2003). Further, patients' individual perceptions of

their illness and its variability, as well as how they make sense of the illness's influence on day-to-day tasks have been related to depression and adjustment (Weinman, 1990). These findings indicate that psychological factors play at least some role in depression in MS and physical explanations alone are unlikely to be sufficient to explain depression in MS patients.

In a study exploring the prevalence of psychiatric disorders and measures of psychopathology in relapse-remitting MS outpatients, 50 outpatients with relapse-remitting MS and 50 healthy controls matched for sex, age and educational level, were assessed. MS patients reported a higher prevalence of psychiatric disorders as well as higher rates of non-mood psychiatric disorders. In a study of 50 MS patients and 50 healthy controls, it was found that the risk factors for depression amongst people with MS were female gender and severity of disability and there were higher rates of depressive psychopathology in the MS patients even when they were in the remitting phase of their illness (Galeazzi et al., 2005). A correlation was also confirmed between disability status and self-perceived depressive symptoms and it has been suggested that this supports the view that depression is a psychological reaction to the neurological illness (Zorzon et al., 2001). This finding supports a previous study which found that the greater the disability, the greater the feelings of uselessness, worthlessness and burden, which is a common subjective experience of patients with MS (Mohr et al., 1999).

Depression is highly correlated with anxiety so it has been suggested that there is likely to be a high prevalence of anxiety disorders amongst people with MS (Siegert &

Abernethy, 2005). There is evidence in the literature to support this notion, however the number of studies utilising meticulous methodology is limited in comparison to the studies on the prevalence of depression. In one study aiming to determine the prevalence of anxiety and depression in 88 patients with MS, it was found that 34% met the criteria for probable caseness for anxiety on the Hospital Anxiety and Depression Scale (HADS) (Smith & Young, 2000). However it has been suggested that as the sample were patients attending an MS clinic it is possible that they would be experiencing higher rates of anxiety than would be found in a community based sample of people with MS (Siebert & Abernethy, 2005). Further, whether these people with MS had an anxiety disorder is unclear.

Other emotional disturbances occasionally found amongst people with MS include euphoria, pathological laughing and weeping as well as mania and there has been debate over whether these symptoms are a result of brain damage directly or a psychological reaction to diagnosis, highlighting the need to distinguish the impact of depression from that of the neurological disease itself (Beatty & Scott, 1993). A study aiming to establish the prevalence of pathological laughing and crying in MS and to define the associated neurological, emotional and cognitive correlates screened a sample of 152 patients with MS for pathological laughing and crying (Feinstein, Feinstein, Gray & O'Connor 1997). Pathological laughing and crying was defined by sudden, involuntary displays of laughing or crying or both, without associated subjective feelings of depression or euphoria. It was found that the prevalence of pathological laughing and crying in MS was 10% and was not associated with exacerbations of the illness but was associated with

more advanced disease. Patients did not differ from controls in levels of depression or anxiety but had a significantly lower IQ.

It has also been found that a diagnosis of MS can trigger symptoms of posttraumatic stress disorder as a consequence of sudden concerns about the progression of the disease and the future of the individual (Chalfant, Bryant & Fulcher, 2004). Recent studies support a relationship between stress and disease exacerbation, showing that stress increases the likelihood that an individual will experience an exacerbation of their symptoms (Ackerman et al., 2002; Sibley, 1997). Sibley (1997) found that stress in relation to work and marriage was more likely to be followed by a relapse as opposed to negative life events. Ultimately, it should be acknowledged that the experience of an exacerbation in MS symptoms provides an additional source of stress for the patient.

1.5 Quality of Life and Health-Related Quality of Life

It is not surprising given that patients have to cope with the physical, neuropsychological and psychological consequences of MS, that MS in its entirety affects quality of life. A few studies to this affect then relate to role functioning and the potential for loss of important roles which increase the risk of poor quality of life. Quality of life, a term relating to an individual's perception of their general well-being and role fulfillment in both physical and psychosocial areas of their lives, has been reported to be negatively influenced by the presence of MS (Forbes, While, Mathes & Griffiths, 2006). Further, quality of life has been found to be significantly poorer in people diagnosed with MS

compared to those diagnosed with other chronic illnesses, such as rheumatoid arthritis, diabetes and epilepsy (Hermann et al., 1996).

An MS diagnosis represents a significant adjustment in patients' lives. When a member of a family becomes chronically ill, previously formed ideas about the role of that family member, what their future will look like and how they will be able to participate within the family are questioned. The psychological consequences (outlined above) that result from these considerations, coupled with the uncertainty of the illness and its associated physical degeneration, then impacts on their quality of life (Chalfant et al., 2004). In a study assessing physical and psychological health, social relationships as well as home and community environments, it was found that a lower quality of life is reported amongst people with MS than the general population (McCabe & McKern, 2005).

Health-related quality of life is more focused on the effects of disease and treatment on physical and psychological functioning as well as overall outlook on life. Assessment of health-related quality of life usually includes assessing patients' mobility, fatigue, pain, cognitive function, general health and the impact of these symptoms on their functioning (Hart, Fonareva, Merluzzi & Mohr, 2005). Problems associated with MS, such as fatigue, pain, employment problems, depression and relationship problems were found to have a negative effect on health-related quality of life in a study aiming to explore the interrelationship between a number of common MS problems and health-related quality of life independent of the effect of physical disease impact (Forbes et al., 2006). Further,

health-related quality of life was most compromised in patients experiencing multiple problems.

1.6 Benefit-Finding

There is evidence that people with MS tend to search for meaning and that benefit-finding helps strengthen role function and is therefore associated with better quality of life. Finding some positive meaning in illness and other adverse life events has been found to enhance coping ability and lessen the risk of depression (Katz, Flasher, Cacciapaglia & Nelson, 2005). This process of finding benefit from adversity has been termed benefit-finding, whereby existing beliefs are redefined as a form of cognitive adaptation to lessen the impact of the situation. Using these new found meanings as coping mechanisms is known as benefit-reminding (Van Der Wende, 2000). These terms were originally developed to apply to response to trauma and have recently been broadened to include chronic illness. Benefit-finding has been found to be a psychosocial effect of MS as patients tend to experience a deepening of relationships, enhanced appreciation of life and an increase in spiritual interests (Mohr et al., 1999). Patients with MS have described finding meaning through a deepening of faith and relationships with family (Forbes et al., 2006).

Finding meaning in the face of threatening events is suggested to be protective of mental health and may also be protective of physical health (Taylor, Kemeny, Reed, Bower & Grunewald, 2000). Similarly, benefit finding in the face of illness has been linked to both

psychological and physical health, and it appears as a central concept in cognitive models of adaptation to threatening circumstances (Katz et al., 2001; Sharpe & Curran, 2006).

1.7 Impact of MS on life planning and child-bearing decisions

A diagnosis of MS undermines much of the planning that would normally be occurring in this stage of a woman's life (Confavreux, et al, 1998). Since most women are diagnosed between 20 and 40, in their child-bearing years, women often have to make reproductive choices before their prognosis is clear and while the future remains uncertain. Further, since 50% of patients are likely to have serious mobility problems within ten years of diagnosis, waiting for clarity in prognosis may mean that increased disability makes parenthood more difficult than it might previously have been. This necessitates most women with MS making a decision about motherhood during a period of considerable uncertainty.

In addition to the uncertainty, the fact that MS onset is in early adulthood, that is after maturation, results in additional challenges for the individual as their already developed sense of identity may be questioned or doubted (Cole & Cole, 1993). All of these issues mean that individuals diagnosed with MS face many psychological barriers as they are faced with potential changes in self-image as well as role changes as a spouse and sexual partner, parent, and income provider. This can have effects on patients' quality of life.

Intuitively, it would seem that health-related quality of life indicators, such as diagnosis, severity of illness and disability, would impact views on motherhood. For example,

women may fear that they will not be able to adequately care for a child during times of a disease exacerbation. Further, if people with MS perceive themselves as having poorer quality of life, many women may be making decisions about motherhood based on a negative view of what their future will look like.

It has however, been reported that the experience of parenthood can lead to an improvement in overall quality of life for infertile women (Abbey, Andrews & Halman, 1994). Relatively little is known about the impacts of voluntary childlessness on the quality of life of women with MS. However the instance of childlessness remains higher amongst women with MS than the general population (Damek & Shuster, 1997). It is possible that voluntary childlessness may be one predictor of lower quality of life. That is, women with MS may be deciding to remain childless due to perceived risks or negative societal attitudes. If this is the case, then it is possible that women are negatively impacted by their choices. Providing this group of women with information as well as enhancing their ability to think through their underlying value system may aid them in making an informed decision and hence improving their quality of life. The information and support provided by health professionals is critical. It has been stated that the relationship between the patient and health care professional is a critical factor that can influence attitudes towards motherhood, decisions about contraception, and whether to keep or terminate an unplanned pregnancy (Becker & Krumm, 2006).

In summary, a diagnosis of MS marks a significant event in any person's life with marked changes in physical ability. Considerable deficiencies in cognitive function can

also occur and patients report a lower quality of life as a consequence of these changes. Many patients also suffer from depression and other psychological disorders are commonly experienced. Since MS occurs more frequently amongst women, during the child-bearing years of their 20s and 30s, intuitively, this poses challenges for women with MS with regard to motherhood decision-making.

1.8 Parental Disability

While there is a relatively small body of literature on MS and pregnancy decision-making, there is a larger literature on the effect of general disability and motherhood. Past research into the reproductive experiences of disabled women has revealed that ‘risk’ is a common theme that emerges when making the decision to become a parent (Thomas, 1997). Generally, common concerns refer to the health of the baby as well as the mother’s own health, the fear of passing on the illness or any possible risk due to drug treatments. What may result are feelings of guilt and anxiety should the child inherit their illness and the interpretation of this as irresponsible and unjust.

It has also been found that disabled women tend to perceive others as judging them to be inadequate mothers (Thomas, 1997). As a consequence, many women feel the need to demonstrate their competency to the outside world, further adding to feelings of insecurity as well as emotional and physical distress. This tends to result from the generalist view of disabled individuals as receivers of care as opposed to being capable of caring for another (Grue & Laerum, 2002). However, parenting impact is not purely influenced by the presence of a chronic illness or disability. Other variables include

components of family status, such as single parents and social support, the child status itself, which includes variables of age, gender and temperament, and finally, family process aspects, including parenting style and communication (Kelley, Sikka & Venkatesan, 1997).

While the physical symptoms of MS may affect mothers' agility and limit the extent of physical interaction between a mother and her child, MS has been found not to have a significant negative impact on the interaction between mothers and daughters (Crist, 1993). This highlights the probability that being reared by a parent with a chronic illness, with its associated physical disabilities, will not have a harmful effect on the child.

Furthermore, a substantial body of literature defending the rights of disabled women to bear children emphasises love and instilling a strong sense of self-worth in one's child as the basis for being a good mother (Cole & Cole, 1993; Killoran, 1993; Kocher, 1993) and argues that the disabled person has grown accustomed to changing the way they carry out their daily activities and parenting is no different. Contrary to what evidence has shown, women with MS have on occasion been discouraged by health professionals from having families and from nursing their babies for fear of further deterioration of their condition after childbirth (Poser & Poser, 1983).

1.9 Multiple Sclerosis and Pregnancy

Historically, in the absence of scientific evidence, women with MS were discouraged from becoming pregnant. Prior to 1950, the general consensus was that pregnancy accelerated the course of MS and when pregnancy did occur, a termination was often

recommended by medical professionals (Cook et al, 1994). The issue of whether pregnancy affects the course of MS has been the subject of much research (Cook et al, 1994; Damek & Shuster, 1997; Hutchinson, 1999; Lorenzi & Ford, 2002; Poser & Poser, 1983; Watkiss & Ward, 2002) and there is now consensus in the literature, based on recent prospective studies, that pregnancy in women with MS neither exacerbates their illness nor is a risk to the child (Confravreux, Hutchinson, Hours, Cortinavis-Tourniaire, & Moreau, 1998; Confavreux & Vukusic, 2002; Ferrero et al., 2004; Lorenzi & Ford, 2002; Meuller, 2002; Watkiss & Ward, 2002). The reality is that due to the unpredictability of the illness, in the long term the condition may get worse whether or not pregnancy occurs.

During pregnancy, the risk of relapse is reduced, but following pregnancy, women are more at risk of relapse in the three to six months following their child's birth (Confavreux & Vukusic, 2002; Cook et al., 1994). This is problematic since the first six months post-natally are stressful for any new mother. The increased risk of a relapse further increases stress for the mother and her partner. After this time the relapse rate tends to return back to the pre-pregnancy baseline level (Ferrero et al., 2004). MS symptoms that commonly worsen during pregnancy, despite the reported reduction in exacerbations, include fatigue, bladder and bowel dysfunction, spasticity and difficulty with mobility (Giesser, 2002) and this may make pregnancy an additional burden for patient who are affected.

Studies have not supported a likely prognosis of long-term disability in MS as a result of pregnancy. No significant association has been found between level of disability and

number or timing of pregnancies, MS onset and deteriorating MS condition (Weinshenker, Hader et al., 1989) and women who become pregnant following an MS diagnosis seem to have a reduced risk of developing the chronic progressive course of the illness from a previous relapse-remitting course (Runmarker & Anderson, 1995). What must be noted, however, is the possibility that women with the severe chronic progressive form of MS may avoid pregnancy, or may be unable to become pregnant, so this could produce biased results.

A clearer idea of the severity of an individual's MS and the ensuing disability becomes more apparent approximately five years after an MS diagnosis and as a consequence women are often advised to delay pregnancy for five years in order to gain a better idea of the likely degree of disability support necessary for parenting (Smeltzer & Kelley, 1997). It is probable however that this request may add further stress to the decision-making process whereby a possibility of developing the severe chronic, progressive form of the illness may mean avoiding pregnancy altogether.

Studies on the effects of maternal MS on infant health and development report that there are no adverse consequences for the baby. In one study 37 women with MS, 15 pregnant and 22 childless women, were followed up for 3 years (Worthington, Jones, Crawford & Forti, 1994). Those in the pregnant group experienced a higher frequency and severity of relapses in the first 6 months post partum but were below the rates expected 6-24 months post partum. Of note is that babies born were in the normal range with regard to weight and head circumference (Worthington et al., 1994).

In a large study investigating the effect of maternal MS on delivery and birth outcome in vaginal births, 851 509 intended vaginal births registered in the Medical Birth Registry of Norway from 1988 until 2002 were studied (Dahl, Myhr, Daltveit, & Gilhus, 2006). Of these, 449 births had noted a diagnosis of MS in the mother leaving 851 060 births as the control group. Fifty five MS births and 45 935 control births were excluded because of planned Caesarean section. Results showed no difference between the groups for complications leading to emergency Caesarean section. However trends which approached significance were evident. Women with MS tended to more often have induction of labour, a slower progression to second stage of labour and increased use of forceps all approached significance, with these features more apparent in the MS group. In comparison to control babies, babies of women with MS had a lower mean birth weight and length. However, the frequency of serious complications, such as stillbirths, the perinatal mortality rate and the rate of birth defects were not significantly different between the groups (Dahl et al., 2006). MS-related disability and fatigue may offer an explanation for the higher rate of planned Caesarean sections as well as the higher rate of forceps use. The slower progression to second stage of labour may also be explained by MS-related symptoms, such as neuromuscular perineal weakness, spasticity, fatigue and exhaustion. However, importantly, despite being smaller, the babies of women with MS were essentially healthy.

Medications for MS are effective in reducing the length of relapses and improving the number of symptoms that people experience (Lechtenberg, 1995). However, a number of

the medications used to treat MS are not advised during pregnancy. Many have been shown to be harmful to the developing baby, although in other cases the effects are simply unknown. Given that the various medications often used in MS can affect the unborn child, one of the issues that is important to consider is the risk of going off medication while trying to conceive as well as during pregnancy.

Patients with MS are prescribed a number of disease-modifying therapies depending on their MS subtype. Immunotherapy drugs such as Avonex, Betaferon, Copaxone and Rebif are beneficial for people with relapse-remitting MS and work to reduce disability, the frequency of relapses and disease activity (Lechtenberg, 1995). However, it is not known from the available studies what the effects of these drugs are on the unborn baby.

Miscarriages while on treatment with interferons have been reported, however there are also cases of healthy babies born to women who continued with their interferons during pregnancy (Gilmore, Pennell & Stern, 1998). It is also not yet known if the drugs are present in breast milk. Because of the uncertainty it is normally advised to stop taking Avonex, Betaferon and Copaxone one month before trying to conceive, during pregnancy and while breastfeeding. It is advised to stop taking Rebif three months before trying to conceive, during pregnancy and breastfeeding.

Methotrexate is one type of drug that is known not to be safe during pregnancy (Weinreb, 1994). Studies have shown that methotrexate can cause cranio-facial and limb defects in babies and it can also cause central nervous system abnormalities and leads to an increase in the likelihood of miscarriage (Ferrero, Pretta, & Ragni, 2004). Methotrexate should be

avoided during lactation and should not be taken while a mother is breastfeeding as it has been associated with several potential problems, including immune suppression, neutropenia, adverse effects on growth, and carcinogenesis (Ferrero et al., 2004).

Steroids and corticosteroids are used to ease inflammation at the affected site, and control the severity of relapse. Intravenous steroids are administered only with extreme exacerbations and they should be avoided during the first trimester of pregnancy because it is thought that the drug can affect the unborn baby (Ferrero et al., 2004). Steroids increase risks of congenital abnormalities, and can slow growth while the baby is in the uterus. They can be present in breast milk and should be avoided while breastfeeding. However, dexamethasone is not associated with congenital defects and it is sometimes used to stimulate foetal lung maturation in patients with premature labour (Ferrero, Pretta, & Ragni, 2004). Dexamethasone crosses the placenta, though methylprednisolone is metabolised before crossing the placenta so is preferred during pregnancy. Methylprednisolone also seems to have little effect on the developing foetus (Ferrero et al., 2004).

Mitoxantrone, an anticancer drug, is known as Novantrone and is more often used for those people with secondary-progressive and progress-relapsing MS. It is an immunosuppressant and studies in rats report low foetal birth weight, retarded development of the kidney and an increased incidence of premature delivery (Ferrero et al., 2004). This drug is known to be present in breast milk and should not be taken while

pregnant or breast-feeding. Moreover, the effects of this drug on future fertility remain unclear.

1.10 The Motherhood Choice

Despite evidence showing that pregnancy in MS is neither dangerous to the mother, nor a risk for the baby, women continue to view having children in the face of MS as a risky decision (Smeltzer, 1994). Additionally, the motherhood choice is complicated further by pressure from medical professionals to delay pregnancy for five years. Delaying pregnancy for five years not only means the possibility of developing a more chronic type of MS, but may introduce problems of conception that are associated with older mothers, thus making the motherhood choice even more complicated.

Despite the fact that research now clearly shows that pregnancy is not medically contraindicated for women with MS, it has been found that many women with MS have avoided pregnancy and parenthood because of perceptions of doubt and disapproval from those around them (Wates, 1997). In a population cohort study, pregnancy course, birth outcomes, and need for rehospitalization within two years after delivery were compared for 198 women with multiple sclerosis and a comparison group of 1584 women without MS. Interestingly, demographic results highlighted that a greater proportion of women with MS (30%) had a previously induced termination of pregnancy than women in the control group (20%) (Mueller, 2002). Moreover, despite the changing advice that is recommended for women with MS concerning pregnancy, research shows that there continues to be higher rates of termination of pregnancy for women with MS who

become pregnant than for the general population (Mueller, 2002). Further, the frequency of childlessness in women with MS is greater than in the general population (Damek & Shuster, 1997).

1.11 Involuntary Childlessness

While women with MS are not infertile, there remains a higher number of terminations amongst women with MS, as well as a higher frequency of childlessness (Damek & Shuster, 1997; Mueller, 2002; Wates, 1997). To date, there is no research on the psychological impact of voluntary childlessness amongst women with MS or women with other disabilities. However, the effect of childlessness on women's psychological function has been extensively studied amongst women unable to conceive. While speculative, it seems intuitively likely that this group of women, who decide not to have children due to their illness, may be experiencing similar psychological effects, such as grief, anxiety, depression and a threatened sense of self.

The lifetime prevalence of infertility during childbearing years has been reported to be between 17-26% (Schmidt, 2006). Infertility has been identified as one of the most highly distressing experiences and fundamentally different from any other stressor (Peterson, Newton, Rosen & Schulman, 2006). In fact, 50% of women unable to have children report infertility as the most distressing experience of their lives (Freeman, Boxer, Rickels, Tureck & Mastroianni, 1985). The experience of infertility brings about a number of stressors such as problems with sexual functioning, relationship pressure and changes in social and family networks (Newton, Sherrard & Glavac, 1999). Further to

these, is the emotional impact of the perceived loss of having children and of being able to relate to other people with children. Infertility has also been found to pose a unique threat to women's sexual identity and sense of self (Andrews, Abbey & Halman, 1992) and anxiety and depression are reported commonly amongst those who experience infertility (Jordan & Ferguson, 2006).

Social identity theory suggests that people form their identity via a process of comparison to others (Tajfel, 1978). Further, theories have put forward the idea that for women, motherhood is the stage of reaching "full womanhood" (Morrell, 1994). It has been argued that involuntarily childless women would compare themselves to other women and feel a loss of identity as a result of being unable to fill the motherhood role. Infertility has also been associated with feelings of failure, anger, shame, confusion and helplessness (Letherby, 2002).

Research has most commonly focused on the impact of infertility on the individual, but more recently there has been a body of literature exploring how each partner experiences the event and how they cope together as a unit (Anderson, Sharpe & Irvine, 2003; Peteron, Newton & Rosen, 2003; Peterson et al., 2006; Schmidt, 2006). One study assessed couples who were referred for infertility treatment for infertility-related stress, adjustment within their relationship and depression. Couple congruence, or a feeling of agreement within a couple about how they define stress and their appraisal of the severity of a stressful event (McCubbin, Thompson, Thompson & McCubbin, 1993), was found to be a significant predictor of depression in infertile women but not men. That is, where

there was incongruence in the marital relationship over the need for parenthood, a higher number of women (21%) scored in the depressed range in comparison to men (9%) thus highlighting that while infertility is a stressful experience for a couple, women perceive it as more stressful and are more affected psychologically than are men. Of course, it is difficult to extrapolate these findings to women with MS who make a choice not to have children. However, this likely depends on whether a woman feels that they are making a choice or decision. Intuitively, if women make a considered and informed decision, it is less likely that they will regret the decision and experience negative psychological impacts.

In recent years, the emphasis in health care has shifted from a focus solely on medical indicators to an emphasis on quality of life indicators. It is well documented, across different illnesses, that quality of life is only moderately associated with disability and other illness characteristics (Sharpe, Sensky & Allard, 2001; Wang, Mayo & Fortin, 2001). The same is true for the association between disease variables and psychopathology. The World Health Organisation (WHO, 1980) has attempted to describe the relationship between impairment, disability and patient-perceived handicap. According to this system of classification, impairment is defined as the actual damage due to an illness. The impairment in MS refers to the demyelination of the central nervous system. In contrast, disability refers to the tasks that the person is no longer able to perform, such as the inability to walk without a stick. Importantly, patient-perceived handicap is determined by the degree to which the person perceives that their disability impedes them in achieving their major life goals or continue in their valued social roles.

That is, patient-perceived handicap would depend on whether or not the inability to walk unaided affected major life roles. Clearly, women's choices about motherhood would affect the degree of patient-perceived handicap.

1.12 Making Decisions About Motherhood

Even those women who become pregnant express concerns about the effect of MS on their pregnancy and vice versa. Indeed, pregnant women with MS continue to view their decision to conceive as risky (Smeltzer, 2002). In a study aiming to understand how women with MS make decisions about pregnancy and childbearing, 15 pregnant women with MS were interviewed about their decisions to become pregnant and asked about the various factors that influenced their decision (Smeltzer, 2002). Most women described the decision to have a baby as a difficult one, complicated further by a lack of a reliable and definitive source of information, uncertainty about the future of their MS, and the effect of pregnancy on their MS. Several factors were identified as considerations in this decision-making process: a) available information about pregnancy in MS, b) uncertainty about the risks of pregnancy in MS, c) availability of support from others in their decision making and the prospect of support if needed in the future, d) any potential psychological benefits of having a baby, e) the previous course of MS, and f) the paucity of alternatives to pregnancy for women with a chronic illness who desired to be a parent. Hence, despite evidence to suggest that pregnancy in women with MS is safe, women with MS continue to conceive at lower rates than healthy women. Further, those who choose to have a family, continue to view their decision as a risk despite good scientific knowledge to the contrary.

A small number of qualitative studies have been carried out in order to determine the unique themes that arise specifically for women with MS when considering becoming a parent (McNary, 1999; Smeltzer, 1994, 2002). Using a descriptive case-study design to identify themes arising in the motherhood decision, a sample of four women with MS who were pregnant participated in a semi-structured interview covering eight broad topic areas: a) childhood aspirations, b) childhood aspiration to motherhood, c) messages about motherhood received from family of origin, d) post-diagnosis changes in childhood aspirations, e) post-diagnosis changes in childhood aspiration to motherhood, f) post-diagnosis changes in career aspirations, g) important considerations entering motherhood decision, and h) salience of MS among considerations entering motherhood decision (McNary, 1999). The 12 most salient themes that arose for all of these women with MS included: a) motherhood culture, b) value of motherhood, c) motherhood self-concept, d) superwoman, e) career, f) independence, g) MS as a family disease, h) MS as a physical concern, i) self-focused anxiety, j) loss, k) social support systems, and l) partner concerns (McNary, 1999).

1.12(a) Motherhood culture, value of motherhood, motherhood self-concept

This theme referred to the motherhood culture in the women's family of origin and the influence that it had on their motherhood self-concept and the value placed on motherhood. Women felt that being a mother was special and a blessing and women agreed that being a mother is an assumed part of life. Women spoke of their mothers staying home to care for them when they were young and how

they were in control like, “a finely choreographed ballet” (McNary, 1999, p. 97) and then spoke of their own fears of being unable to provide a child with this same level of care. These fears centred on deteriorating strength and energy levels as well as high levels of stress.

1.12(b) Superwoman, career, independence

This theme was reflected in statements where women described themselves as perfectionists and the belief that MS would not beat them. Typically, women likened their independence to stubbornness and this reflected many being adamant that they would be able to manage work and parenting and their illness on a full time basis.

1.12(c) Social support systems, partner concerns

Many women spoke about the importance of having a good support network around them who would be able to help if they were to experience an exacerbation. Some recommended that this is a vital aspect to be sorted out during the process of decision-making. Women also spoke about their experience of having different ideas about parenting to their husbands and the effect that this had on their ability to receive adequate support from their partners.

1.12(d) MS as a family disease, MS as a physical concern, self-focused anxiety, loss

These themes specific to MS were reflected in all four participants. There were concerns about the impact any visible symptoms would have on their children's relationships with peers at school as well as financial costs for families should women become too ill to work or require expensive medical treatment. Women also expressed concerns about the possibility that they may deteriorate physically and be unable to perform the tasks required to fulfil the role of a mother. These tasks included anxiety about an inability to keep children safe as a result of slowed reaction time. The theme of loss was relevant in the sense that women felt that because of MS they would be unable to fulfil ambitions they had always held.

Other common fears and concerns that women with MS have during the process of decision making have been the unpredictability of MS and the effect that pregnancy may have on the course of their MS (Smeltzer, 2002). Five categories of concerns have been identified as important issues in the decision making process: a) concerns about the pregnancy itself, including the decision to become pregnant, b) the effect of MS on future child-bearing plans, c) labor and delivery concerns, d) concerns and issues related to breastfeeding, and e) concerns about the baby's well-being and child care (Smeltzer, 2002). With respect to deciding whether to become pregnant, the contributing factors identified as making the decision more difficult included a lack of available and reliable information and at times, conflicting advice, uncertainty about the future course of their MS and the possible effect that pregnancy may have on that course. What was also

identified in this study were the salient factors influencing their final decision, including information regarding pregnancy and MS, uncertainty of the risks of pregnancy in MS, availability of social support both in terms of the decision making itself, as well as future help prospects, potential psychological benefits of having a baby, the individual's previous MS activity and the lack of alternatives to pregnancy available for women suffering from chronic illness or disability. All the women interviewed in this study went through with their pregnancies and utilised a method of risk-benefit analysis to come to their decision, which concerned weighing up the potential risks and benefits involved in becoming pregnant.

Despite the reported concerns about pregnancy and the post-natal period, what appears to be the greater issue for women with MS is not the pregnancy, but the childrearing. It is difficult to predict the likelihood of being physically able to care for a child some years ahead and the possibility of childrearing as being impractical in the face of chronic illness can be a distressing thought for many women. Fatigue may be particularly worse in the postpartum months when the mother may be withstanding the nutritional drain of breastfeeding and experiencing an irregular schedule, coupled with increased relapse rates (Lechtenberg, 1995).

Women with MS considering pregnancy and childrearing would be faced with an exaggerated experience of the already stressful concerns associated with bringing a child into the world. These concerns include role changes associated with parenthood, the wellbeing of the new baby and their confidence in being a good parent (Chalmers, 1982).

Additionally, women with MS have to consider a number of issues that are not relevant to healthy women. For example, women must consider possible effects of MS on pregnancy and the effect of pregnancy and the post-natal period on MS. Many of the medications used to treat MS are contra-indicated during pregnancy and women must consider the effect on MS of ceasing their medication, sometimes before conception. Practical difficulties also present and women worry about whether they will need a caesarean section, be able to have epidural anaesthesia during delivery or whether they can breastfeed their baby (Ferrero et al., 2004). MS can affect a woman's ability to care for an infant and perform parenting tasks in the short or long term. Finally, women worry about the risk of their child inheriting MS (Ferrero et al., 2004; Smeltzer, 1994).

Among mothers with MS it has been reported that lack of emotional, instrumental and informational support have been significant issues (Harrison & Stuifbergen, 2002). Information pertaining to mothers being able to care for themselves as well as their infant in the presence of a disability has not been easily obtained, possibly due to the history of those with severe disabilities being discouraged from becoming pregnant. Nonetheless the motherhood decision is a significant decision for any woman, whether she has MS, any other medical condition or no illness at all and identifying the main concerns for this group of women has important implications for health professionals in their treatment of women going through this decision-making process. It is important to understand how people share decision-making with their healthcare professionals. The following section explores medical decision making and interventions to assist the process as it relates to the motherhood decision for women with MS.

1.13 Shared Decision-Making

Clearly, motherhood is a decision that requires careful thought in the best of circumstances, but the decision is complicated for women with MS who need in addition to consider medical risks and physical consequences. Therefore, such decisions need to be considered by women and partners in conjunction with physicians who can advise on issues or prognosis for the individual woman. The literature highlights four existing models of doctor-patient decision-making (Emanuel & Emanuel, 1992). These are the Paternalistic, Interpretive Decision-Making, Shared Decision-Making and Informed Decision-Making models and it has been suggested that there is much overlap in their interpretation (Charles, Gafni & Whelan, 1997, 1999). These models differ in the level of participation of the patient in choosing and shaping their 'treatment'. In the paternalistic model, the doctor, as the person with most knowledge of the subject matter, makes all the decisions about the patient's treatment after evaluating the disease, treatment options and likelihood of different outcomes. It is assumed that due to their knowledge, the doctor will make a decision in the patient's best interests. The paternalistic model predominated until the last few decades, which has seen a shift towards more patient involvement. In the interpretative model, the responsibility of choice still lies with the doctor, however, the doctor also considers the values and preferences of the patient. The shared decision-making model emphasises the role of both parties, that is the doctor and the patient. Within this model, the two parties reach a decision together after the patient receives all necessary information regarding risks and benefits and alternative options available and communicates their values and preferences to the doctor. Shared decision making has also been associated with terms such as informed decision making, concordance,

evidence-based patient choice, enhanced autonomy, and mutual participation (Braddock, Edwards, Hasenberg, Laidley & Levinson, 1999; Ford, Schofield & Hope, 2003; Ruland & Bakken, 2002). Finally, informed decision-making is described as a model whereby the authority lies with the patient in making the final decision after the doctor presents them with all available treatment options (Wirtz, Cribb & Barber, 2006).

Of these models, shared decision is now seen by most commentators as the gold standard (Butow & Tattersall, 2005). The shared decision-making model evolved from patient-centred medicine which emphasises that doctors should practice holistically, appreciating the patient's subjective experiences (Weston & Brown, 1995). It is seen as the option which respects patient autonomy and contribution, while avoiding over-burdening the patient, who often lacks expertise and may be emotionally vulnerable.

Shared decision making occurs via a process of comprehensive communication between the doctor and patient and it has a number of key characteristics. Shared decision-making must involve both the doctor and the patient, otherwise the process is not shared.

Additionally, both parties must play an active role in the decision-making process and this may require fulfilling a number of roles. Roles might include gathering information, recording and interpreting it, advising the patient of treatment options and relaying the risks and benefits associated with each option as well as negotiating issues such as timing or place of treatment (Charles et al, 1997). Further, the process of sharing information is necessary for shared decision-making to occur. Typically, the health professional explains all treatment options available, thereby beginning a process of evaluating each

option in collaboration with the patient. During the process of shared decision-making, patients are informed of the nature of the illness and its prognosis, as well as being given a number of options for treatment. Each option is reviewed in collaboration with the patient and the pros and cons of each are considered. Pros and cons may include weighing up things like risks and benefits, cost and convenience. This provides an opportunity for the patient to discuss at length their different perspectives and consider their own personal values, concerns, preferences, expectations and ideas. In doing this with their doctor, they are provided with an opportunity to hear their physician's recommendations in the context of a particular individual (Charles et al., 1997). A final characteristic of shared decision-making is that both parties agree to the final decision when it is made.

1.14 Personal Characteristics Influencing Patients' Ability to Share Decisions

Shared decision-making, while commonly seen as desirable, is not necessarily an easy process for either doctor or patient (Butow & Tattersall, 2005). The likelihood that an individual patient will engage in shared decision making depends in part on their degree of self-efficacy, or their perception of their ability to make the specified decision (Bandura, 1986). The term self-efficacy refers to an individual's own perceived ability to perform a specified behavior or set of behaviors (Makoul & Roloff, 1998). Self-efficacy is a construct that comes from Social Cognitive Theory, that brings together cognitions, behaviour and the environment via reciprocal interaction (Makoul & Clayman, 2006). This theory highlights that behaviours are largely determined by the outcome and efficacy expectations that patients associate with them. For example, an outcome expectation may be that eating less salt will reduce one's blood pressure, while an

efficacy expectation refers to that person's belief about whether they will be able to eat less salt.

Individuals with more confidence, or who perceive themselves to have greater confidence with regard to a specific course of action, are more likely to engage in that behaviour.

Further to this, these people are also more likely to initiate communication

(Bodenheimer, Lorig, Holman & Grumbach, 2002; Makoul & Roloff, 1998), adjust better to illness and treatment (Airlie, Baker, Smith & Young, 2001) comply with treatment plans and adhere to health behaviours recommended to them by their health professionals (Marcus, Selby, Niaura & Rossi, 1992). Thus people who feel capable of making a decision are more likely to engage in shared decision-making.

Self-efficacy is often dependent on the comprehensiveness of an individual's understanding of all the facets that make up a particular decision (Makoul & Roloff, 1998). These facets may include having a thorough understanding of the facts surrounding the decision, including medical facts about the illness and any associated side effects, understanding the doctor's perspective, and the ability to gain further clarification if needed. Self-efficacy also includes the patient's confidence in their ability to get the facts about the risks and consequences associated with each of the options available, ask questions without feeling incompetent, to ask outside parties for advice and also to handle unwanted pressure from others. Research on self-efficacy has highlighted the importance of the health professional in checking and clarifying the understanding of the patient to help them to be able to make an informed choice (Gazmararian, Williams,

Peel & Baker, 2003). With regard to MS and the motherhood decision, the literature as outlined previously has shown that the medical outcomes are finely balanced, suggesting that discussion around the importance of personal values for each individual is essential. Thus the degree to which shared decision-making is preferred is based on the degree to which values are primary and medical outcomes are finely balanced (O'Connor, Legare & Stacey, 2003).

Of further importance within the shared decision making model, is the issue of follow-up. Medical decisions are not always made immediately, but in many cases are decided upon gradually over a period of discussion. Shared decision making provides an avenue whereby there may be an initial presentation of options on behalf of the health professional, after which the patient may take this information home to discuss with their significant others or with their healthcare team (Charles, et al., 1997, 1999). Thus, an essential component in patient care is to follow up with the patient for further discussion whereby any new issues may be raised or questions from the family involved may be answered in order to provide them with a clearer picture. This is particularly the case with reference to MS and motherhood, where decision-making is ongoing and involves multiple people, such as partners and health professionals.

It is important to recognize that, while conceptually, shared decision-making assumes an equality or partnership, the decision-making is unlikely to be shared equally. Its emphasis lies in the belief that the perspectives of patients as well as health professionals are of equal importance, and that they both have a role in decision-making.

1.15 Decision-Making about Motherhood in MS

The motherhood decision is not a once off decision in its entirety, but rather a process of decision-making that is often reconsidered many times. Once the initial decision to have a child has been made, other family planning issues arise such as how many children to have, what size gap to leave between siblings and whether to terminate or continue should an unplanned pregnancy occur. When planning a family, there exist many social, economic and emotional decisions that must be faced and having to factor in a chronic, unpredictable illness exacerbates the complexity of an already complex decision.

Smeltzer (2002) conducted a qualitative study of pregnant women with MS. In describing the decision to become pregnant, women likened this to a “risk-benefit analysis” (Smeltzer, 2002, p.147). The participants described a lack of available information about pregnancy in MS which led them to be uncertain about the risks to them or their child. They balanced these fears against the perceived psychological benefits of having a baby and the lack of alternatives (e.g. adoption) for women with a progressive neurological condition. Further, women specifically referred to lack of available support from others in their decision-making. Surprisingly, there is no research on shared decision making in this context. Historically, doctors simply advised women against having children, and according to women’s accounts, more recently doctors tend to regard the decision as a woman’s decision and abstain from advising them.

Decision-making may be defined as a process of making choices between different courses of action or inaction. However there are often situations whereby the possible

outcomes of different alternatives may not be all positive or even known, and can often result in combinations of both negative and positive outcomes. Therefore, the right choice for an individual is not clear and they find themselves in a dilemma of uncertainty. This situation in which there is uncertainty and conflict over choices where possible results may include risk, loss or regret, is known as decisional conflict (O'Connor, Jacobsen & Stacey, 2002). As it has been ascertained that an MS diagnosis complicates family planning decisions, it would be expected that women and their partners would experience much decisional conflict when faced with decisions regarding their family's future.

1.16 Interventions to Support Decision Making About Motherhood in MS

In order to provide support for health care decision-making, a number of strategies have been identified as helpful. These include providing information about options and outcomes to increase knowledge, realigning individual expectations of outcomes to ensure they are realistic, clarifying values to decipher what is most important for each individual, and increasing individual's skills in decision making using guidance and coaching techniques (O'Connor et al., 2002). Preparing the client and clinician for the decision making process and, where appropriate, scheduling follow-up counselling may also improve the quality of the choice. Preparation is necessary, especially for those who may be experiencing difficulty engaging in decision-making, or who may be vulnerable to excessive stress and anxiety as a result of considering and re-considering options. It is also useful to prepare clients for communication with the clinician and include follow-up counselling for a period following the initial decision. This is an important stage where factors such as inadequate knowledge, unrealistic expectations, unclear values, unwanted

pressure and inadequate support may be further addressed. Additionally, self help skills in handling pressure, seeking social support and managing consequences may be learned.

Providing a structure such as this ensures an individually tailored and supportive decision making process which provides appropriate information to ease the stress associated with decisional conflict as much as possible. Patients can learn about any likely benefits or risks and weigh them with their own personal values. One approach, which tries to utilise this structure to facilitate shared decision-making between health professionals and patients, is the decision aid.

1.17 Decision Aids

Decision aids are being increasingly utilised in the medical profession to assist patients in making informed decisions regarding choices of treatment, therapy and various types of medical tests (Feldman-Stewart et al, 2007). They differ from usual patient information handouts and education materials by providing descriptions of various risks and benefits involved as well as discussing the likelihood of each outcome occurring. Handouts and brochures generally quote statistics that are not easily understood and do not easily allow individual patients to judge their own personal likelihood of an outcome occurring (O'Connor et al., 1998). Decision aids may be tailored according to a patient's clinical risk profile, while also assessing and utilising their personal values in order to come to a conclusion, which is aligned with their own value system. The rationale for decision aids is to provide the patient with knowledge that ensures patients are well informed about the medical risks involved in their choice, and to lead them to weigh the medical

considerations with their own attitudes and values (Deyo, 2001). As a result, the high levels of stress and anxiety associated with decisional conflict are reduced so patients can play a more active role in decision-making. This process aims to reduce decisional conflict and minimise regret over the decision, once made.

Decision aids are of particular benefit for health decisions that are marked by a new situation or diagnosis, have uncertain risks and benefits and require more effort in making a decision rather than implementing that choice (O'Connor, Rosten & Fiset, 1999).

Common components of decision aids include information presented in structured format to increase patients' knowledge about the relevant medical issues. Information is tailored to consider the risks and benefits in a balanced manner to ensure realistic expectations of outcomes. Various issues that are important in considering the decision are discussed and supplemented with exercises to help patients clarify their own values relevant to the decision they are making. In addition, decision aids often include the experiences of other people who have been in a similar situation and describe their retrospective accounts of making their decisions (Feldman-Stewart & Brundage, 2004).

Decision aids facilitate the clients' ability to make possible outcomes tangible and functional. They aim to enable clients to better judge the likelihood of different outcomes for them personally, thus making each benefit and risk for them somewhat clearer. After completing a decision aid, the client has in their possession a visual summary of choices, outcomes and probabilities based on accurate, comprehensible and balanced information (Lawrence et al., 2000).

Most decision aids are related to decisions to have or forego a particular intervention (e.g. screening in prostate cancer) or to decide between two treatments (e.g. mastectomy versus lumpectomy in early breast cancer). One example is a decision aid for women considering hormone therapy after menopause (O'Connor et al., 1998). This decision aid uses a self-administered, self-paced booklet and audiotape and is designed to be used by women at home in preparation for a follow-up visit to their doctor. The illustrated booklet includes general information, worksheets enabling the woman to weigh benefits and risks, clarify her values, identify current health practices and preference for her level of participation in decision-making and list any questions to ask at the visit. The booklet also suggests that the women review the worksheet with their doctors. Ninety-four women participated in the trial to validate the decision-aid and they completed questionnaires assessing their knowledge, their current predisposition towards taking hormone replacement therapy, decisional conflict, personal expectations and importance ratings of benefits and risks associated with hormone replacement therapy. Results showed that after using the decision-aid, participants' level of general knowledge about hormone replacement therapy had increased, and their personal expectations of the risks and benefits were more realistic. Further, women were reported to feel more certain of their choice, informed, clear about their values as well as more supported in decision-making (O'Connor et al., 1998).

Another example is a cross-cultural consumer-based decision aid for screening mammography (Lawrence et al., 2000). In this study, an evidence-based decision aid was developed for European American and Mexican American women between the ages of

50 and 70, who are at average risk of breast cancer, of varying educational levels and who speak either English or Spanish. In its validation, the decision aid was developed and evaluated by a multidisciplinary team including oncologists, biostatisticians, social scientists, graphics and educational specialists and lay people. Forty-nine European Americans and 54 Mexican Americans constituted the sample to test its reliability and validity. Results showed that women could comprehend the information, integrate it with their own personal value system and they were also able to communicate their preferences for screening mammography.

A systematic review of randomised trials of patients' decision aids in improving decision making and outcomes has been reported recently (O'Connor, Stacey, Entwistle, Llewellyn-Thomas et al, 2003). Compared with controls, patients receiving decision aids had a higher knowledge of options and outcomes, more realistic expectations, less difficulty in reaching a decision, more active participation in decision making, and no differences in anxiety levels, or satisfaction with decisions or the decision making process.

Two types of decisions have been identified (O'Connor, Legare & Stacey, 2003): (a) those decisions where there is good medical evidence that one treatment has more benefits than harm for a patient; and (b) where there is not strong evidence to favour one choice over another and the preference for treatment is related to a person's values. Decision aids are particularly useful to the latter type of decision where the options are finely balanced and it is the patient's values which are important in determining the

preferred course of action (O'Connor et al., 2003). This is particularly relevant for the motherhood decision in MS. Starting a family is an individual choice that needs to balance the importance of motherhood for the woman and her partner against the risks that she will be unable to care for the infant or child as a result of increasing disability. Surprisingly, despite the fact that motherhood decisions are clearly the type of decision ideally suited to the development of a decision aid, no aids exist of this nature for any illness group. To date, there is no information available to women with MS who are currently considering the motherhood decision. This is despite the fact that MS is an illness that predominantly affects women of childbearing age.

1.18 Developing a Decision Aid

The C.R.E.D.I.B.L.E. criteria were developed by the Cochrane Systematic Review of Patient Decision Aids (O'Connor et al., 2003) to guide the development of decision aids.

These criteria are as follows:

(C) Competently developed: The decision aid must include the components that promote quality decision making and be developed by researchers with appropriate qualifications. Further, in the development process it is important to undertake a needs assessment and a review of the draft materials both by experts and consumers.

(R) Recently Up-dated: The data included in the decision-aid should be based on evidence available within the past two years.

(E) Evidence-Based: Statements describing the benefits and harms that are included in the decision aid should be based on scientific studies or systematic reviews.

(DI) Disclosure of conflicts of Interest: Sponsorship or conflicts of interest are disclosed.

(BL) Balanced presentation of options, benefits and harms: The decision aid should present balanced information and be viewed as balanced by most users.

(E) Efficacious at improving decision-making: Evaluations should include a randomised controlled trial and should demonstrate that the decision aid is effective in improving knowledge, is acceptable to users, is free from adverse effects, and shows other benefits, such as reduced decisional conflict or increased decisional self-efficacy.

1.19 Aims and Hypotheses

The aim of this research project is to determine the need for a decision-aid for women with MS in New South Wales and Victoria. The second aim is to explore the relevant themes associated with motherhood decision-making amongst women at various stages of this decision-making process. The primary aim of the project is to develop a decision-making aid with the function of assisting women with MS in making their decision of whether to become a parent. The tool will be evidence-based, and will be designed for use in clinical practice to facilitate decision-making amongst women with MS who are considering the motherhood decision. Finally, the decision aid will be evaluated as to whether it provides information that increases knowledge about pregnancy in MS, facilitates decision-making in women with MS who are in the process of deciding whether or not to have a child and influences the decision. Finally, the research will attempt to determine the long-term impact of the decision-aid on motherhood decision-making at 12-month follow-up. All ethics documentation for this research project is included as Appendix A.

This research project is broken up into four phases:

Phase 1 of the study is a needs assessment, which aims to determine the proportion of women for whom the motherhood decision is relevant and to identify participants for phases 2 and 3. It is anticipated that the motherhood decision will have relevance to a large percentage of women with MS between the ages of 20 –40.

Phase 2 of the research is a qualitative study that aims to explore the themes associated with motherhood that are relevant for a heterogeneous sample of women with differing views regarding the motherhood choice. Previously no research has focused on the issues relevant to those women who choose to forego motherhood, or those who are yet to make a decision. This study will attempt to fill this gap in the literature. This phase will also serve as a pilot study to gain feedback from both consumers and physicians on the decision aid that has been developed by the authors. Given that phase 2 is qualitative, speculative hypotheses will not be made. However, we do expect that by representing the views of women who have yet to decide or have decided against having children, new themes will emerge that have not previously been documented in the literature.

Phase 3 of this project is a randomised controlled trial of the decision aid. The aims of this phase are to determine whether the decision aid (a) increases knowledge; (b) reduces decisional conflict; (c) affects a woman's decision regarding motherhood, (d) increases decisional self-efficacy for those who make a decision, and (e) is free from adverse effects on psychopathology. For the randomised controlled trial, it is anticipated that the

decision aid will increase knowledge about pregnancy and MS, reduce decisional conflict, increase decisional self-efficacy and that women will be more certain about their choice. It is not expected that the decision aid will push women in one direction or another. In terms of the psychological difficulties commonly experienced among people with MS, increases in anxiety or depression are not expected in women with MS as a result of the decision aid.

The final phase of this study is a follow-up to be conducted at 12 months following participants' initial involvement in the research. The aim of the follow-up study is to assess the status of womens' motherhood decision since post-intervention, using a quasi-qualitative design, in order to determine the long-term impact of the decision aid and the mechanisms through which it operated. It is expected that some women will have become pregnant and those women who have become pregnant will be asked about their experiences of being pregnant and what impacted on their decision-making process, regardless of whether they were in the treatment or control group. Those allocated to the intervention group will also be asked about their use of the decision-aid. This aims to determine whether the decision aid impacted on the way in which women approached their decision and their subsequent experience of pregnancy and/or early motherhood. It is anticipated that participating in the study as part of the intervention group will have had an impact on the motherhood choice for women with MS and that these women will be more certain of their choice as compared to before they began their participation in the study.

1.20 Significance to MS

Many women with MS are faced with a decision whether or not to start a family at a time where the full impact of the disease on their life is unknown. To date, there is no informational resource available to women with MS who are currently considering the motherhood decision. This is despite the fact that MS is an illness that predominantly affects women of child-bearing age. Further, those who choose to pursue a family have numerous concerns about their health and the health of their child. Research suggests that, despite strong scientific evidence confirming the safety of pregnancy in MS, doctors remain reticent to offer advice on motherhood to women with MS (Smeltzer, 2002). Research also shows that the number of women with MS who choose not to have children is higher than in the general population or other illness populations (Mueller, 2002). These data suggest that women may be making uninformed decisions about their families.

While there has been an increase in research into the physical outcomes of pregnancy in MS with large scale methodologically rigorous studies (Confavreux et al., 1998; Cook et al., 1994; Dalmas, Texier, Ducloy-Bouthors & Krivosic-Horber, 2003; Damek & Shuster, 1997; Deatrick, Brennan & Cameron, 1998; Ferrero et al., 2004; Giesser, 2001, 2003; Hutchinson, 1999; Lorenzi & Ford, 2002; Mueller, 2002; Poser & Poser, 1983; Runmarker & Anderson, 1995; Verdru, Theys, D'Hooghe & Carton, 1994; Vukusic et al., 2004; Weinshenker, Hader et al., 1989), the psychological impacts surrounding motherhood in MS are largely neglected aside from a few qualitative studies limited to pregnant women or those who choose to have children (McNary, 1999; Smeltzer, 1994,

2002). We currently have very little information about how many women are undecided, why women choose not to have children and whether women are making informed decisions.

If the decision aid is demonstrated to be effective in promoting women's decision making, it could be widely applied in routine clinical practice with women with MS. The present design will determine whether the decision aid is effective on a range of relevant variables. The design will ensure that the decision aid is successful in increasing knowledge, reducing decisional conflict and increasing decisional self-efficacy. Importantly, the inclusion of measures of psychopathology will ensure that the intervention does not have unintended negative consequences for women with MS.

CHAPTER 2: REPRODUCTIVE CHOICES OF WOMEN WITH MULTIPLE SCLEROSIS

2.1 Introduction

Multiple sclerosis (MS) is the most common neurological disease affecting young adults. MS affects ~1 in 1000 people, and, as with other autoimmune diseases, women are more likely to be affected than men. Women with MS are typically diagnosed during the childbearing years (age 20–40 years) (Smeltzer, 2002). The course of MS varies considerably, with some patients experiencing a progressive course from the outset and others following a more benign course. However, the disease course is often unclear for years after diagnosis. For example, 10 years may pass after diagnosis before a patient is certain to have a benign course of the illness (Jopson & Moss-Morris, 2003). Hence, a diagnosis of MS is given at a time when women would ordinarily be making decisions about motherhood.

Despite the evidence that pregnancy does not accelerate the course of MS, women with MS continue to be overrepresented among those who remain childless and who present for termination of pregnancy (Wates, 1997). Although qualitative research suggests that most women with MS have a strong desire to become mothers, many of them have avoided pregnancy and parenthood because of perceptions of doubt and disapproval from those around them or their own fears of being unable to cope with the demands of pregnancy or parenting (Wates, 1997).

The motherhood choice can be defined as the choice to forego, start, or enlarge a family, including facets such as how many children to have and how much time to leave between

children. The literature suggests that the decision to become a parent has been complicated in recent years because of the career opportunities available to women (Meyers, 2001; Sevon, 2005) and increased availability of contraception and abortion (Tardy, 2000). Nonetheless, most healthy women report intending to have children. In a cohort of the Australian Longitudinal Study on Women's Health, 91% of young women wanted children (Lee et al., 2005).

The decision becomes complicated when health is compromised, as with human immunodeficiency virus/autoimmune deficiency syndrome (HIV/AIDS) or sickle-cell anemia. For example, a study exploring reproductive decision making in women who were healthy carriers of sickle-cell anemia found that the women had difficulty discussing their carrier status with their partners, risking rejection and the loss of opportunity to have children due to the risk of giving birth to an affected child (Asgharian, Anie, & Berger, 2003). Little is known about whether these concerns change the reproductive decisions of the women. In contrast, the HIV/AIDS Surveillance Report from the US Centers for Disease Control and Prevention (2001) stated that 84% of women with HIV and 79% of women with AIDS were diagnosed during their reproductive years, yet only 32% of women of reproductive age wanted to have children (Heard, Sitta, & Lert, 2007). However, both sickle-cell anemia and HIV/AIDS carry potential risks to the unborn child, which is not the case for MS. Hence, we cannot generalize from these findings as to whether the diagnosis creates ambiguity regarding reproductive choices for women with MS.

Because of the progressive and unpredictable nature of MS, coupled with the usual age at onset during childbearing years, women with MS are often faced with decisional conflict

regarding the motherhood decision. The presence of a chronic illness would exacerbate the already physically and emotionally demanding time of pregnancy and the postnatal period. For most women, the presence of MS may necessitate that they consider additional factors in the decision-making process. Surprisingly, little research has focused on this issue. Studies to date relating to pregnancy and MS have only used small samples ($n < 15$) of women who are either pregnant or already have children, which may not be representative of all women with MS. Therefore, whether women are, in fact, undecided about motherhood is unknown (McNary, 1999; Smeltzer, 1994, 2002).

To our knowledge, this study is the first quantitative research to determine what proportion of women with MS are undecided about motherhood. The results offer important information about the needs of women with MS.

2.2 Method

The MS Society of New South Wales (NSW) and Victoria, Australia, maintain a database of members who have agreed to be contacted about research that is endorsed by the society. The MS Society sent a mailing to all female members between the ages of 20 and 40 years, constituting a potential sample of 1410 members.

Members were sent a 1-page questionnaire asking them to complete basic demographic information (eg, age, marital status, MS type) and to indicate where they were in the decision-making process about motherhood. They were asked to return the questionnaire to the researchers in a postage-paid envelope. A copy of the questionnaire is included as Appendix B.

2.3 Results

The women in the sample had a mean age of 33 years (SD 4.30 years) and had been diagnosed with MS for an average of 6.67 years (SD 4.58 years). Most of the respondents had relapsing-remitting MS (80%), 13% were unsure of their type of MS, and smaller proportions had primary progressive (2%) or secondary progressive (4%) disease. These figures are typical of MS samples with the age and sex inclusion criteria of our sample.

All statistical output is included as Appendix C.

Of the 461 women who responded to the initial mailing, 212 indicated that they were undecided about the motherhood choice (Table 2.1). One hundred and fifty-eight women already had children and did not want to have more. Only 52 women indicated that they did not want to have children, and 39 indicated that regardless of MS, they want to have children.

Table 2.1

Status of motherhood decision making for women with MS during childbearing years

Decision status	N	Percentage
Unsure/considering it		24.7
Have children/considering more	212	21.3
Have children/not considering more	158	34.3
Do not want children	52	11.3
Want children	39	8.5
Total		100.0

As shown in Table 2.2, 80% of respondents had the relapsing-remitting form of MS. Of those, 50% were undecided about motherhood. Thirteen percent of women were unsure of their MS type. Of these, 44% were undecided. In contrast, 70% of those with primary

progressive and 81% with secondary progressive MS had decided to either have no children or no more children.

Table 2.2

Reproductive decision status of women with MS during childbearing years, by MS type

Decision status	MS type					Total
	RRMS	SPMS	PPMS	Don't know	Benign	
Unsure/considering it	91	2	0	12	0	105
Have children/considering more	69	1	3	11	0	84
Have children/not considering more	101	8	2	19	0	130
Do not want children	31	5	5	4	1	46
Want children	30	0	0	6	0	36
Total	322	16	10	52	1	401

MS, multiple sclerosis; RRMS, relapsing remitting; SP, secondary progressive; PP, primary progressive

Table 2.3

Results of women who have children vs. women who do not

	N	%
No children		
Want	39	19.9
Unsure	105	53.6
Do not want	52	26.5
Total	196	100
Children already		
Unsure	98	38.3
Do not want	158	61.7
Total	256	100
Total undecided	212	46

2.4 Discussion

We hypothesized that many women with MS would be undecided about their reproductive plans. The results show that almost half of the women who responded had not yet made their decision. Another third of women reported that they already had children and did not want more. Amongst women who did not have children, 26.5% had chosen voluntary childlessness (11.3% of the total sample). In the general population, the instance of voluntary childlessness amongst healthy women is 9% (Lee & Gramotnev, 2006).” In the two groups of women who had made their decision to either begin or forego starting a family, the motherhood decision would likely have been a necessary process, in the past, to reach this conclusion. Unfortunately, the study did not look into how many of these women received their diagnosis before starting their families or for how many the motherhood choice was affected by MS. However, if the decision was relevant only to those currently undecided or yet to decide, half of the women surveyed would be represented. Clearly, then, these results support the conclusion that the motherhood decision is relevant to women with MS.

The breakdown of women with each type of MS into the various statuses of decision making was interesting. Most studies of MS and motherhood have been on women with relapsing-remitting disease, because it is the most common type. The situation is the same in the current study, with 80% of respondents ($n = 322$) having relapsing-remitting MS, of which half ($n = 160$) were undecided in their motherhood choice.

Among those with secondary progressive MS ($n = 16$), most (81%) either decided against having children or already had children and did not want to expand their families.

Secondary progressive MS begins its course as relapsing-remitting MS; therefore, this group was older by an average of 3 years and had already made motherhood decisions. Nonetheless, a relatively high proportion (31%) of women in this group had decided to remain childless.

Similarly, although only 10 women in the sample had primary progressive MS, 5 of them had decided against having children, and only 3 had children. Although a small sample, these data suggest that more extreme progressive disability has a greater influence on future life choices, such as family planning, than less disabling forms of disease.

A smaller proportion ($n = 52$) of women did not know the course of their MS. We assume that their prognosis was unclear to them. A similar proportion of these women (44%) to those with relapsing-remitting disease (50%) were considering motherhood. However, they were doing so without clear information about their prognosis, which would have important implications for informed decision making. For example, women who are not informed about their type of MS and its associated prognosis may decide to forego

motherhood because they have assumed that their illness will progress quickly to severe disability.

2.5 Limitations

The study relies on self-report, which may mean that the MS types reported were not entirely accurate, particularly in that many women were unsure of the diagnosis.

Nonetheless, the study shows that women were attempting to make their reproductive choices despite lacking this information.

The low response rate of the mailing (33%) may indicate a response bias. Women may have failed to return questionnaires for various reasons. However, the possibility that those who responded are those for whom this issue was most relevant is likely. Even if this is the case, the motherhood decision was so common among those surveyed that it was relevant (at the time) to 15% of the possible sample (ie, 212/1410).

How relevant MS was in the reproductive decision-making process of women who already had children remains unknown. A high proportion of women reported that they already had children and did not want anymore. Knowing whether they were diagnosed before having children and, if so, whether MS complicated their decision making would have been useful. However, national trends show that women are delaying motherhood, this decision will become increasingly influenced by MS for women following diagnosis.

Finally, the study did not have a comparison group, which would help determine whether rates of indecision among women with MS are more prevalent than those for women

without MS. However, recent data suggest that most healthy women, when asked, reported that they want to have children (Lee, et al, 2005).

2.6 Conclusion

To our knowledge, this is the first study to determine in a large sample of women with MS in the childbearing years, what proportion remains undecided about motherhood. The study indicated the relevance of this issue for many women with MS. Prior research suggested that women face considerable conflict about the motherhood decision (Smeltzer, 2002). Further research is necessary to explore the needs of this group.

A follow-up of the women who were undecided, to explore the reasons for their indecision and highlight the factors associated with MS that might be increasing their uncertainty, would be useful for addressing these issues in clinical practice. The development of a decision-making aid may help women access appropriate information and consider how their values should be weighed against other relevant information, thus allowing for less conflict regarding the decision.

CHAPTER 3: THE MOTHERHOOD CHOICE: THEMES ARISING IN THE DECISION-MAKING PROCESS FOR WOMEN WITH MULTIPLE SCLEROSIS

3.1 Introduction

MS is the most common chronic neurological disorder among young adults. It most often strikes women and its onset is typically in the childbearing years (20s-30s) (Confavreux, Hutchinson, Hours, Cortinavis-Tourniaire, & Moreau, 1998). Due to the progressive and unpredictable nature of MS, coupled with the time of onset being one associated with family planning, women with MS are often faced with decisional conflict regarding the motherhood decision as the already physically and emotionally demanding time of pregnancy and childbearing may be exacerbated by a potentially disabling illness. Therefore, the presence of MS necessitates additional factors to consider in the reproductive decision-making process.

Women with MS considering pregnancy and childrearing can be faced with an exaggerated experience of the already stressful concerns associated with having a child. These may include role changes associated with parenthood, the wellbeing of the baby and their confidence in being a good parent (Chalmers, 1982). Additionally, women with MS have to consider issues that are not relevant to healthy women. Common concerns amongst these women include any possible effects of MS on pregnancy and the effect of pregnancy and the post-natal period on MS. Many medications used to treat MS are contra-indicated during pregnancy and women must consider the impact of ceasing their medication on their MS. Practical difficulties also

present and women worry about whether they will need a caesarean section, be able to have epidural anaesthesia during delivery or whether they can breastfeed (Ferrero, Pretta, & Ragni, 2004). Symptoms of MS in the post-natal period, during which women are at higher risk of relapse, can affect a woman's ability to care for an infant and perform parenting tasks in the short or long term. Finally, women worry about the risk of their child inheriting MS (Ferrero, Pretta, & Ragni, 2004; Smeltzer, 1994).

Only three qualitative studies have been carried out to understand the concerns of women with MS considering motherhood (McNary, 1999; Smeltzer, 1994, 2002). The most salient themes that were identified included 'independence', 'MS as a physical concern', 'MS as a family disease', and the importance of the 'motherhood self-concept' (McNary, 1999). These themes were added to the usual concerns associated with motherhood, such as 'career', 'financial resources' and 'partner concerns'. In a study of pregnant women, women identified a lack of available and reliable information, conflicting advice, uncertainty about the course of their MS and the possible effect that pregnancy may have on the disease as their major concerns (Smeltzer, 2002).

Although these prior studies give insights into the experiences of women with MS choosing motherhood, this remains an under-researched area. All three available studies have relied on interviews with pregnant women (McNary, 1999; Smeltzer, 1994, 2002). Hence the views of women choosing *not* to have children have not been represented. Similarly, there is no information on how women with MS fare early in the post-natal period. Importantly since those studies, there is now good evidence to suggest that MS does not adversely affect pregnancy or the infant, nor does pregnancy

adversely impact the long-term course of MS (Confravreux, Hutchinson, Hours, Cortinovis-Tourniaire, & Moreau, 1998; Watkiss & Ward, 2002).

The aim of this study is to document the main themes and concerns for women with MS considering motherhood. We included the views of women who had decided against having a family, as well as those with children and those who were undecided. As a result, this qualitative study will offer be the first to document the experiences of women with MS who are considering motherhood.

3.2 Method

As presented in chapter 2, we conducted a large mail-out in January 2005 to 1410 women, aged between 20 and 40, from the NSW and Victorian MS Societies asking them to indicate what stage they were at in the motherhood decision. Women were also asked whether they would be interested in taking part in a larger study on this topic.

Four-hundred and sixty-one women responded and were divided into four groups: (1) those who decided against having children (n=52), (2) those who had children and did not want anymore (n=158), (3) those who decided to have children (n=39), and (4) those who were undecided about their motherhood choice (n=212). Women from the above groups who lived in Sydney and responded within four weeks of the mail-out were eligible to participate. From these women, consecutive women were chosen to represent the four groups of participants described above. Recruitment continued until theoretical saturation (no new themes emerging in two consecutive groups) was reached. This occurred after four focus groups and one individual interview.

Women were contacted and invited to attend a focus-group or individual interview in February or March 2005. Women unable to attend a focus group were offered a telephone interview. All women contacted agreed to participate. Semi-structured questions were used to guide discussion covering womens' experiences of making this decision, including the factors they were weighing up, their experience of accessing relevant information from organisations and health professionals and the attitudes of those around them with regard to their decision. For women who had made a decision, how they reached their decision was discussed, as well as their experience of having a child, any challenges of parenting and how they coped in the post-natal period. See table 3.1.

Table 3.1

Interview questions for focus groups

How has your MS diagnosis affected your decision to have children?

What sorts of factors were you weighing up in making your decision?

What was your experience of accessing information about this topic?

What has been your experience of having a child?

Have there been any challenges for you in parenting? If so, what were they?

What was the post-natal period like for you?

Did you experience different attitudes from those around you towards having a child?

The focus-groups were facilitated by the candidate. Each focus-group or interview lasted for approximately 60 minutes, was audio-taped and transcribed (range: 20 mins – 76 mins).

This study had ethical approval from the University of Sydney Ethics Committee and Multiple Sclerosis Australia.

Audio-taped data was transcribed and analysed qualitatively using the Framework method of analysis developed by the National Centre for Social Research, a method in which themes are developed both from the research questions and from the accounts of research participants, to identify the themes associated with the motherhood decision (Towle, Godolphin, Grams & La Marre, 2006). After transcription, the transcripts were independently read by the candidate and the Primary Supervisor (LS) to identify the major themes and develop a coding framework. The coding framework identified semantic content relevant to the subject matter that could be organised into meaningful groups (Braun & Clarke, 2006). The different codes were then sorted into potential themes and all coded data was collated within the identified themes. The researchers then met with the Associate Supervisor (PB) to confirm the themes and coding framework. Any disagreements concerning themes were discussed until consensus was reached.

3.3 Results

Twenty women with MS participated, with a mean age of 32 (range 20-40) years and diagnosed with MS on average 6.5 years ago (see table 3.2). Sixty percent of the women had relapse-remitting MS, 10% had secondary-progressive MS and 10% had primary-progressive MS. Twenty percent did not indicate their MS type. Five participants were unsure of whether to start, forego or enlarge their family, five definitely wanted children, six had decided not to have children and four already had children and did not want more.

Table 3.2

Demographic information of the participants

<i>Demographic Information</i>	<i>Frequency</i>
Age (at time of focus group)	
20-25	3
26-30	5
31-35	6
36-40	5
Not reported	1
Time since diagnosis	
1-5 years	9
6-10 years	6
11-15 years	4
16-20 years	1
Type of MS	
Relapse-Remitting	12
Secondary Progressive	2
Primary Progressive	2
Don't Know	4
Number of children	
Pregnant	2
None	12
One	4
Two	1
More than two	1
Motherhood decision status	
Unsure	5
Definitely want children	5
Do not want children	6
Already have children, don't want more	4

The following are the main themes that emerged from the qualitative analysis:

*3.3(a) Health and well-being**My health during the post-natal period*

For women who were undecided, a major concern was their future well-being. They described fears of the post-natal period, which is typically characterised by increased relapses and how this would impact on their ability to care for a new infant.

“It's the 3-6 months after having the baby you are tired and having to get used to breastfeeding and getting up every 4 hours, it's going to be difficult and you're more likely to relapse”.

Coming off medication

The uncertainty around how their health would be affected if they discontinued medication for conception and pregnancy (and breastfeeding) was also a common concern for these women.

“Being able to stay on medication was really important. Being able to manage my relapses was like really, really important”.

“I'm really afraid of going off my medication and having a bad exacerbation”

Current health reflective of future health

Additionally, the current state of women's MS influenced their self-efficacy in decision-making because they viewed their current state as a reflection of how their future health would be.

“I've only been diagnosed for a year and I've had 4 relapses in a year so I haven't been going great...it may continue to go this way...I think I've got to be a bit better about how I'm going with MS, and my husband, before I know which way to go”.

“There's a part of me that would love to have children but there's also a really big part of me that questions what effect MS is going to play in their life...it's made a big impact in my life so what's it going to do with a child”

3.3(b) Child's well-being

Passing on MS

Despite the very small objective risk, a large proportion of women worried that they would pass MS on to their child. Some spoke of the guilt that they would experience if their child inherited MS and the devastation at watching their child suffer. Some were unaware how small the chance of passing MS on was (3-5% if one parent has the illness) indicating a lack of available information. In contrast, some who knew the facts and had children, expressed anger at the insensitivity of people questioning their decision to start a family in case they passed on their illness.

“I thought about having kids and it just upset me because I didn't want to pass MS on to them”.

“The small percentage of being able to pass it on that would be particularly devastating that my child developed it”.

“I would have loved to have had children...but I wouldn't take the risk of them getting MS.”

“A lot of people who don't know anything about MS have asked me about my child inheriting the illness...it really annoys me”.

“Would it be so bad? Is your life so terrible that you'd rather not have it? Because mine isn't. Even when I've been really, really, really sick, I'd still choose to be here. I just don't understand where people are coming from when they ask that...”.

Child being a carer for their parent

Many women who were undecided were concerned that their child may end up caring for them as their disease progresses. Numerous women stated that having to care for a parent is not a childhood, and that childhood is a precious time which should not be taken away from any child. These women reported that they would feel guilty and feel like a 'bad mother' if this occurred.

"I don't want to bring up a child who is going to have their childhood robbed from them by being a carer for their parent".

"I don't want the child to end up looking after me...for me that's just not a childhood".

Harming the child

A prominent fear amongst women, particularly those who had decided not to have children and those with more progressive illness, was whether they could keep their child safe, especially in the early years of the child's life. Many questioned whether they would be sufficiently agile to protect their child from harm or to keep up with a child. Due to the uncertainty of their prognosis, women were fearful that future progression might affect their ability to care for their child. Women worried about dropping their baby or falling over and hurting their child.

"The child running away...putting their finger in a power point...not having the reaction time that's going to be fast enough to stop the child from being harmed".

3.3(c) Coping with parenting

Fatigue

All of the women currently deciding whether to begin or enlarge their families expressed concern regarding their ability to parent a child in the face of a progressive and unpredictable illness. These participants reported that worsening fatigue was a source of anxiety as it would make parenting almost impossible. They all explained that taking care of themselves was difficult enough and doubted their ability to cope with the increased fatigue.

“I don't think we want children but it's just a real sort of factor, I need to be alright to be able to look after kids. I'd like to be active with my kids, I want to...do everything that everyone wants to.”

“There's no point getting to the point when you're just so tired you can't even cook or anything...that's not going to help your baby at all”.

Financial concerns

Related was the concern that should parenting become compromised, financial resources would become strained if partners had to cut back on work to help with parenting. This fear was especially difficult to plan for because the uncertain nature of MS prognosis for many of these women.

“I worry about financial figures as well. If I'm actually too tired to look after a child then my husband will have to stop work and that concerns me, especially it's so expensive in Sydney so that worries me.”

“Definitely financial is one. I know that we really want to be financially stable before we sort of move in to having kids, just in case...all the what-ifs I get sick”.

Support

The importance of a good support base revealed itself as a major concern for women in all groups. Women who had not yet decided whether they would start a family were uncertain about asking for help believing that they should be able to cope as well as any able-bodied woman. Some were even unwilling to have children if they were not certain that they would be able to cope most of the time. Women who had decided not to have children often did not have support nearby and this was noted as an important factor in their decision to forego a family. Women who had already had children spoke, with the benefit of hindsight, about the importance in asking for help. However, many noted that when they first had their babies they were reluctant to do so.

“I’ve got a very supportive family and my partner is very supportive so I know I won’t be stressing about those things”.

“You’ve got to start to realise what you can and can’t do and it’s a hard thing to accept.”

“I’d always ring mum and ask her to come and stay with me I need a bit of help and she’d be there for a week and I’d be fine after that.”

“I know we’ve got a great support network...but I personally feel if I can’t do it 9 times out of 10 I don’t want to rely on someone else to do it”.

“I just don't think it's fair to rely on someone else to look after it for me. I know in times of emergency, yes I could call on someone but I don't want to rely on them all the time”.

3.3(d) Societal attitudes and women's reactions

Reactions from family, friends and health professionals

Most women who had previously been through this decision-making process reported experiencing negative attitudes from both health professionals and family members regarding their future family planning. All women reported that at least some family members and friends assumed that these women would not have a family. Even those who did not assume that the woman would remain childless, assumed that there would be difficulties with pregnancy or labour, such as the need for a Caesarian or the inability to breastfeed.

“I remember my neurologist saying, ‘you'll probably never have children’. I'm very much like, ‘don't tell me what to do’ because I like to make my own decisions”.

“I went to see the GP...to check results of whether I was pregnant...she said, ‘Oh yes you're pregnant’ she looked in the file - I have MS. She said oh you shouldn't be having children!”

“The whole way through my pregnancy I'd hear, ‘so I guess you'll be having a Caesarian?’ I'm like, ‘no way’.”

Not letting MS beat you

Despite this, there was a strong union amongst all the women with children that, despite their illness, they valued their lives and would still choose to be alive even if they were forewarned about developing MS. In addition to this, the women who already had children all reported that they went through a stage of defiance where they would not let anyone try and influence their decision. They said they would do the opposite when it was recommended that they slow down and ignored any offers of advice, whether helpful or unhelpful.

“I didn't go around asking people because I didn't really care what they said.”

“I can do whatever I want and I'll do it. And whatever you say is not going to impact.”

Experience of parenting

Women who had children described the challenges they had faced with their pregnancy or parenting. None reported regret over their decision to start a family and all spoke of the joy of motherhood and the importance it held for them in their lives. Interestingly, every woman in the sample who had children described a period of post-natal depression, which they attributed to their reluctance to readily ask for help from others and the pressure of coping with their new infant as well as their MS.

“You've got to realise what you can and can't do...it's a hard thing to accept...when every other parent in the neighbourhood pops their kid in the pram and goes down to feed the ducks”.

“I lost my mother 10 years ago from cancer so I've always wanted to have kids and be a mother. I wanted to have that bond I never had when I was an adolescent, so it's important at one point but it's just making a careful decision now.”

“I would just bounce back from whatever was thrown in my way...I had postnatal depression but refused to let anyone know. I put on a false bravado in front of everybody...and then I crumble because no one offers me help.”

“I was superhuman. I could do anything and everything and no one could stop me. If they dared to get in the way I would try even harder and then I would crumble when no one else could see me.”

Timing and pressure of the decision

In contrast to previous research where women reported that they were often advised to wait 5 years to decide, the women in this study reported common advice from health professionals was that to start a family happen before the age of 30 and before their MS progressed. Hearing this made many women, who were in the process of making this decision, anxious as to the timing of their decision. Women, particularly those in their 20s, commonly reported that they felt it was necessary to rush their decision. This put pressure on their relationships, as they were often not at a stage where they might otherwise have been ready to have children. Women spoke about how unhelpful, unnecessary and even untrue they found some of the advice they had received.

“I don't want to be older and like...secondary progressive...I wouldn't want to be at that stage and for it to be too late for me to start thinking more about having kids if I hadn't had kids.”

“Part of my brain is geared around where I do have MS and in the last 11 years and especially the last 5 it has impacted me...I'm in a wheelchair...I don't have the energy to get around...if I'm considering having a child now or in another 10 years, can I still do it at the age of 40”.

“My neurologist saying you've got to do it before you're 30, that nearly broke my husband and I up”.

“I've always wanted to have kids...you might say I've made a decision, younger rather than wait older before things get worse...cause I probably would have waited 5 years or more”.

“I took my partner to my neurologist early in our relationship to discuss this issue. We, as a couple, were not ready at the time to have children and now I feel I am too old. Information on this earlier would have helped make the decision and opened up discussion.”

3.4 Discussion

The aim of this study was to provide a qualitative investigation of the views of women with MS about the issues relevant to their motherhood decision. Prior research has only encapsulated the views of women who have decided to have children. Our results supported those findings (Smeltzer, 1994) identifying similar themes such as mother's health concerns, coping with parenting and societal attitudes. This study further identified concerns from different groups that had a direct impact on the decision to have children amongst those who had already decided, including the experience of parenting, the child's well-being and timing and pressure of the decision.

Those women who had not yet decided to have children as well as those who had decided not to have children reported different concerns from those with children already. Those who did not have any children held fears about the possibility of their children inheriting their MS. Of concern is that this fear was, for a number of women, disproportionate to the level of risk. For example, the risk of inheriting MS when one parent has MS is 3-5%, a similar risk to juvenile diabetes (2.9%) (Sandler, 1990) and yet a few women reported deciding against a family largely because of the fear that this unlikely outcome would occur.

Women who were unsure of their decision had more concerns around their ability to cope on a number of levels. They questioned their own stamina during labour and were concerned they would be unable to be as functional during the post-natal period when relapses tend to increase (Cook, Troiano, Bansil, & Dowling, 1994). They worried about having to come off medication in order to conceive safely and were fearful of the permanency of any exacerbations occurring while off medication. The ability to parent with the level of fatigue that can occur amongst those with MS was questioned by these women. They also felt that having children would make them financially unstable, particularly during times of relapse when role changes would need to occur in the household and income could not be guaranteed. Women who had decided not to have children held the same concerns though were surer that they would *not* be able to cope with these possibilities.

Those women who had decided to go ahead and have families reported that, in retrospect, they shared the same concerns before making their final decision. Of interest was that, in reaching their decisions, they had developed a defiant attitude.

They tended to do the opposite of what people would recommend with regard to their general health and ignored signs from their bodies that they needed to rest. Indeed, this may account for the very high rates of reported post-natal depression amongst those women in this group. While it is not clear that they would have met formal criteria for post-natal depression, the fact that these women identified their defiant attitude as a source of stress is important because this may be an unintended consequence of uninformed and unhelpful attitudes from society and health professionals. Over time this group had come to learn to realise when they need to slow down and had become more comfortable with asking for help from their support base. Despite their acknowledged difficult experiences during the post-natal period, this group of women nonetheless found meaning in their role of a mother and cherished the bond they have formed with their children.

Those whose illnesses had exacerbated recently, and who did not yet have children, were also worried about their current state of health and being uncertain of the rate of progression of their MS. Being unwell as a new parent was a major concern because of the impact that this would have on the rest of the household as well as other family members. Most women felt that asking for help and for their child to grow up having had to care for their mother at various times rendered them a “bad mother” and were unwilling to become parents while this possibility for their future remained. They had adamant views on what childhood should and should not entail and were of the opinion that having to care for a sick parent is an experience children should not have to endure.

There are several limitations to the study that must be addressed. Firstly, the women recruited for this study were all members of the MS Society and were willing to attend an allocated focus group or interview session. It is possible that those who are members of the MS Society are more likely to seek out and read information regarding their own health and therefore be more interested and aware of what their fears and concerns are. Further to this, it is likely that these women were more able to discuss their concerns than non-responders. Secondly, women were contacted as responses were returned and hence, there may be a bias in the early responders relative to late responders. Additionally, demographic information, such as marital status, level of education and occupation was not collected so it is unknown whether these factors have influenced or biased the sample in any way. Finally, given the qualitative design and the small sample size, these data should be considered hypothesis generating.

These limitations notwithstanding, this is the first study to investigate the concerns of women with MS who are at all the different stages of the motherhood choice. The results supported previous research suggesting that womens' concerns are currently not being addressed adequately. Women themselves reported misunderstandings about information, such as the rate of heritability. Further, they also discussed unhelpful attitudes of well-meaning health care providers which in some instances had unintended negative consequences.

These results indicate that there is a need for access to more information about the effect of MS on pregnancy and child-rearing and the effect of pregnancy on MS and its progression. It seems that such information would be valuable to women in the

midst of this decision-making process. Moreover, it seems that educating health care professionals and the wider community in these issues may also have positive consequences for women with MS who do chose to pursue motherhood.

CHAPTER 4: THE MOTHERHOOD CHOICE: A DECISION AID FOR WOMEN WITH MULTIPLE SCLEROSIS

4.1 Introduction

The motherhood decision is defined as the choice to forego, start or enlarge a family. For women with chronic illnesses or disabilities, this is not an easy decision (Asgharian, Anie, & Berger, 2003; Grue & Laerum, 2002). The issue of reproductive choices amongst women with disabilities is an important area that has received a lot of attention (Grue & Laerum, 2002; Heard, Sitta, & Lert, 2007; Killoran, 1993). Despite the principles of normalization having been adopted in Westernized societies for more than two decades, women with disabilities continue to remain over-represented amongst those who remain childless (Killoran, 1993). Such women often view motherhood as ‘risky’ (Thomas, 1997). Common concerns relate to the ongoing health of both baby and mother, fear of the baby inheriting the illness and risks associated with drugs taken during and after pregnancy to manage the illness (Birk & Kalb, 1992). Operating within a medical discourse which emphasizes minimization of risk can intensify these concerns. Women with chronic illnesses or disability and their partners are sometimes made to feel that having children is irresponsible (Grue & Laerum, 2002; Killoran, 1993). Hence, this is an area in which health care professionals are inadvertently communicating in a way that women experience as unhelpful.

MS is a progressive, unpredictable neurological disorder of the central nervous system and its onset is more common amongst women of child-bearing age (Birk & Kalb, 1992; Cook,

Troiano, Bansil, & Dowling, 1994). The likely activity of the disease for any individual is often unclear for at least five years after diagnosis so women often have to make reproductive choices while their future remains uncertain. Since 50% of patients are likely to have mobility problems within ten years of diagnosis, waiting for clarity in prognosis may mean that increased disability makes parenthood more difficult than it might previously have been (Ferrero, Pretta, & Ragni, 2004). Because the onset of MS occurs in early adulthood, it may challenge a woman's self image, including their present and future images of themselves as spouses, sexual partners, parents and income providers.

Prior to 1950, the general consensus was that pregnancy accelerated the course of MS and women were discouraged from becoming pregnant (Cook, Troiano, Bansil, & Dowling, 1994). When pregnancy did occur, a termination was often recommended. Women with MS were on occasion discouraged by health professionals from nursing their babies for fear of further deterioration of their condition (Poser & Poser, 1983). While most women possess a strong desire to become a mother, many women with MS have avoided pregnancy and parenthood because of perceptions of doubt and disapproval from those around them (Wates, 1997).

The issue of whether pregnancy affects the course of MS has been the subject of much research (Cook, Troiano, Bansil, & Dowling, 1994; Damek & Shuster, 1997; Hutchinson, 1999; Lorenzi & Ford, 2002; Poser & Poser, 1983; Watkiss & Ward, 2002) and there is now consensus in the literature, based on large prospective studies, that pregnancy in women with MS neither exacerbates their illness nor is a risk to the child (Confavreux, Hutchinson, Hours, Cortinavis-Tourniaire, & Moreau, 1998; Confavreux & Vukusic, 2002; Ferrero,

Pretta, & Ragni, 2004; Lorenzi & Ford, 2002; Mueller, 2002; Watkiss & Ward, 2002). In the long term the condition may get worse whether or not pregnancy occurs. Despite this changing evidence concerning pregnancy and MS, higher rates of pregnancy termination (Mueller, 2002) and childlessness (Damek & Shuster, 1997) persist in women with MS than for the general population, and pregnant women with MS continue to view their decision to conceive as risky (Smeltzer, 2002).

The presence of MS complicates the motherhood decision, and there are a number of issues which women need to understand before an informed decision can be made (Mueller, 2002). Four specific factors which women need to consider have been identified (McNary, 1999), as follows: (a) the neuro-physical features of MS; (b) the psychological consequences of MS; (c) the culture of each individual woman and her family; and (d) the historical context in which women with disabilities contemplate motherhood. These themes have been found to influence women in their reproductive choices and to determine the ease with which women and their partners are able to make this decision. However, to date, there are no available patient resources to assist women who are contemplating a decision or the health professionals who are counseling them in their decision.

Decision-making may be defined as a process of making choices between different courses of action or inaction. Many decisions involve weighing up uncertain positive and negative outcomes, leading to uncertainty and vacillation (decisional conflict) (O'Connor, Jacobsen, & Stacey, 2002). The motherhood decision is not a once off decision, but rather a process of decision-making over time. Once the initial decision to have a child has been made, other family planning issues arise such as how many children to have, what size gap to leave

between siblings and whether to terminate or continue should an unplanned pregnancy occur. When planning a family, there exist many social, economical and emotional factors that must be considered (Feldman-Stewart et al., 2007), and having to factor in a chronic, unpredictable illness exacerbates the complexity of an already stressful situation (Smeltzer, 2002).

A number of strategies have been identified as helpful to facilitate decision-making about health care. These include providing information about options and outcomes to increase knowledge, realigning individual expectations of outcomes to ensure they are realistic, clarifying values to determine what is most important for each individual, and increasing individuals' skills in decision making using guidance and coaching techniques (O'Connor, Jacobsen, & Stacey, 2002). Such strategies are commonly employed in decision aids.

Decision aids have been defined as interventions designed to help people make specific and deliberate choices among options, including the status quo, by providing at the minimum information on the options and outcomes relevant to a patient's health (O'Connor, Roston, & Fiset, 1999). They aim "to facilitate patient involvement in decisions about their health care, with the goal that each person's decision be informed and consistent with his/her values" p.47 (Feldman-Stewart et al., 2007). They typically contain relevant evidence-based information presented in a simple, clear, graphical form, and lead patients through a process of clarifying their values and weighing the pros and cons of the options prior to decision-making. A recent systematic review showed that, compared with controls, patients receiving decision aids had a higher knowledge of options and outcomes, more realistic expectations, less difficulty in reaching a decision, more active participation in decision

making, and no differences in anxiety levels, or satisfaction with decisions or the decision making process (Elwyn et al., 2006).

Decision aids are being increasingly utilised in medicine to assist patients in making informed decisions regarding choices of treatment, therapy and medical tests (Elwyn et al., 2006; Volk & Spann, 2000). They are of particular benefit for health decisions made in the context of a new situation or diagnosis, in which there are certain risks and benefits (O'Connor et al., 1998), such as the motherhood decision in MS. To date, there is no information available to women with MS who are currently considering motherhood. Hence, there are few educational resources that health professionals can recommend or use to initiate discussion with women with MS faced by this decision. If the decision aid is demonstrated to be effective in promoting women's decision making, it could be applied by a range of health care professionals in routine practice with women with MS.

This study aims to evaluate a decision aid with the function of assisting women with MS to decide whether or not to start or enlarge their families. Specifically, we hypothesize that the decision aid will increase knowledge about pregnancy in MS; reduce decisional conflict; increase decisional self-efficacy and make women more certain of their decision. We did not expect that the decision aid would affect anxiety, depression or the direction of their decision.

4.2 Method

Eligible women were women diagnosed with MS, aged between 20 and 40 at the time of recruitment, were currently unsure about the decision to start or enlarge their families and

who could read and write English sufficiently to complete questionnaires. The MS Societies of New South Wales and Victoria have databases of members who agree to participate in research endorsed by the Society. The MS Societies sent mail-outs to all women between 20 and 40 years (n=1410) between March and November 2005 inviting them to participate in the study by returning a one-page questionnaire about their motherhood decision-making, the results of which were reported in chapter 2. Women were also asked to indicate if they would agree to be involved in a randomized controlled trial of a decision aid.

Women who consented were then sent baseline questionnaires with a reply-paid envelope. A list of random numbers corresponding to intervention vs control were generated using the Excel Bernoulli function and linked to participant identification numbers. The random sequence was concealed electronically until the point of allocation. Women were randomly allocated after being sent their baseline questionnaires. Women in the intervention group were sent the decision aid which they were asked to read within two weeks. They were telephoned two weeks later to ensure they had read and understood the decision aid, to answer any questions and to screen for distress. Telephone conversations did not exceed 20 minutes and confirmed that all women had read the decision aid. After the telephone conversation or at the corresponding time for control women, the post-intervention questionnaires were sent to participants to be returned by mail.

This study had ethics approval from the University of Sydney Ethics Committee and Multiple Sclerosis Australia.

A draft of the decision aid was developed by the authors following a literature search of the databases PSYCHINFO, MEDLINE and SCIENCEDIRECT, using the search terms PREGNANCY, MOTHERHOOD, PARENTING both individually and combined with MULTIPLE SCLEROSIS from June until November, 2004. Publication dates of literature retrieval ranged from 1961 to 2004. The format for the decision aid was based on the Ottawa Decision Support Framework and followed the C.R.E.D.I.B.L.E. criteria, developed by the Cochrane Systematic Review of Patient Decision Aids (O'Connor et al., 2003). The criteria stipulates that decision aids must be Competently developed, incorporating components promoting quality decision-making. The researchers developing the decision aid should possess appropriate qualifications. The decision aid should be Recently updated, and include information based on evidence available within the last two years. It must also be Evidence-based, such that the information presented is based on scientific studies following a thorough literature search. There should be no Conflicts of Interest, and the decision aid should be Balanced. Finally, the decision aid must be Efficacious at improving decision-making and be free from adverse effects.

The decision aid provided general background information about MS, different types of MS and the likely prognosis associated with each type. Following was a section on the psychosocial impacts of MS on lifestyle factors relevant to decisions about parenting, including financial burden, changes in relationships, psychological disturbances and social role changes within the family.

Data about the effect of MS on child-rearing was described. Research confirming that pregnancy does not alter the course of MS was presented and that MS does not interfere

with fertility or miscarriage rates. The documented effects of MS on pregnancy, labour and delivery, were described, including the fact that 70% of women experience a remission during pregnancy. There was a section that described the best evidence about the safety of different medications during conception and pregnancy. The decision aid also discussed the effect of MS during the post-natal period in terms of increased relapse rates and fatigue. There was a section that addressed breastfeeding issues, such as needing to decide whether to breastfeed if medications were counter-indicated during lactation. There was also a section on commonly asked questions by women who are making this decision, such as fertility, the contraceptive pill and the risk of the child inheriting MS.

There was a section on the issues that may arise in parenting with MS. This section described the concerns that mothers with MS have reported. This included worries of their ability to provide physical affection to their child during relapse. The need for good support post-natally and the meaning associated with motherhood. To present a balanced view, the decision aid included patient stories, of one woman who chose to have and one who chose to forego children. The stories included the factors that influenced their decisions. These stories were developed on the basis of the qualitative interviews reported in chapter 3. The decision aid then included a number of options that women may consider, such as having a smaller family, having no children, leaving a larger gap between siblings etc. The choices were followed by exercises to help women consider their personal values and those of their partners. Women were encouraged to list important factors for and against each option and rate the importance of each factor. Women were also encouraged to consider what support they would have if they had children and how support people could help them.

The decision aid was piloted with 20 women with MS (those who took part in the qualitative interviews described in chapter 3), who commented on its readability, balance and usefulness and gave feedback on the content. The feedback was generally positive, however, some exercises were simplified on the basis of their comments. Fifteen neurologists, who specialise in MS, were also sent the decision aid by mail and given an opportunity to comment on the accuracy of the information. Eight neurologists wrote back with positive feedback and a further seven phone calls were made to the remaining neurologists. There were no factual errors identified. Feedback on the decision from women is included as Appendix D. The decision aid is included as Appendix E.

4.2.1 Measures

Primary outcomes for this analysis were decisional conflict, decisional self-efficacy, knowledge and certainty of decision-making. Secondary outcomes were depression and anxiety. This analysis was conducted on an intention to treat basis.

The following questionnaires were administered and can be found in Appendix F:

4.2.1(a) A *demographic questionnaire* elicited age, duration and type of MS and time since last exacerbation. Participants indicated whether they wanted to have children before receiving their MS diagnosis, whether they currently have children and whether their diagnosis complicated their decision to have children by ticking yes or no. Women were asked to rate their certainty about having children on a continuum where –5 meant they would definitely not have children now or in the future, 0 meant they were unsure, and +5 meant they definitely would have children at some stage. In addition to the score from -5 to +5 which indicated the direction of

a person's choice, we calculated a score from 0-5, to indicate certainty of choice regardless of direction. For example, both -5 and +5 were scored as 5 to indicate women were certain of their decision.

4.2.1(b) As there is no questionnaire which specifically addresses the issue of pregnancy in MS, a *knowledge questionnaire* was developed based on the information provided in the decision aid. Participants were asked to answer 10 questions relating to pregnancy and child-rearing in people with MS. Scores were derived by adding the number of correct answers given.

4.2.1(c) *The Decision Conflict Scale 4th Edition* (A. O'Connor, 1999) : The Decisional Conflict Scale is a 16-item Likert scale that has been widely used to evaluate patients' decisions regarding a range of healthcare decisions. The scale consists of five subscales: uncertainty; feeling uninformed; feeling unclear about one's values; feeling unsupported in decision-making; and the perception of the effectiveness of the decision-making once it has been made. Items in each subscale are scored on a 5-point scale from 1 (strongly agree) to 5 (strongly disagree), with reverse scoring for 5 negative statements. Item scores are summed and divided by the total number of items to yield the average item score, with total scores ranging from 1 (low decisional conflict) to 5 (high decisional conflict). A 2-week test-retest reliability coefficient of 0.80 has been reported, and the internal consistency for the total scale has ranged from 0.78 to 0.92 when the scale was applied to a variety of groups, including patients with cardiac or respiratory disorders who made decisions

about influenza immunization and women aged 50–69 years who made decisions about being screened for breast cancer (O'Connor, 1995).

4.2.1(d) *Decision Self-Efficacy Scale* (A. O'Connor, 1995): The Decision Self-Efficacy Scale has been used with patients confronting a range of decisions. This measure is an 11-item Likert scale, which was administered to determine the level of self-confidence and belief that participants had in their own ability to make this decision. Respondents obtain a score between 0 (extremely low self-efficacy) to 100 (extremely high self-efficacy). It has an internal consistency coefficient of 0.92.

4.2.1(e) *Center for Epidemiologic Studies Depression Scale (CESD)* (Radloff, 1977): The CESD is a short 20-item, self-report scale intended to measure depressive symptoms in the general population. Scores are obtained by the sum of the 20 item weights. It has an internal consistency coefficient of 0.89. A score of 16 or greater is considered depressed. Research has confirmed the reliability and validity of this measure within the MS population (Verdier-Taillefer, Gourlet, Fuhrer, & Aléprouvitch, 2001). This measure was preferred for this population over the Beck Depression Inventory (BDI) (Beck, Ward, Mendelson, Mock, & Erbaugh, 1961) due to the overlap between the somatic items in the BDI and medical symptoms in MS patients (Mohr et al., 1997).

4.2.1(f) *State-Trait Anxiety Inventory (STAI)* (Spielberger, 1983): The STAI is an extensively used self-administered inventory comprising self-report scales for measuring state and trait anxiety. The state anxiety form was used in this study to

compare any changes in anxiety over time. The scale consists of 20 items and respondents rate how well statements reflect how they feel on a 4-point scale. Reliability coefficients exceed .90, the inventory has good construct validity and it is used with MS populations (Tsivgoulis et al., 2007).

4.2.2 Analyses

Baseline differences between the intervention and control group were assessed using parametric (for continuous variables) and non-parametric (for dichotomous variables) t-tests to identify covariates. A series of (group: intervention; no intervention) x 2 (time: pre; post) mixed model ANCOVAs were performed, using SPSS 15 for Windows, to determine the effectiveness of the intervention.

According to the Cochrane systematic review, decision aids, on average, increased knowledge by 19 pts, improved realistic perceptions by 40%, reduced decisional conflict by 9 pts, reduced passive decision-making by 30% and reduced the proportion of undecided people after counselling by 57% (O'Connor, Legare, & Stacey, 2003). A sample of 150 is sufficient to detect each of these differences with at least 80% power at a 0.05 level of significance.

4.3 Results

4.3.1 Demographic variables

As reported in chapter 2, 461 women responded to the MS Society invitation to participate, of whom 212 were eligible. Of these, five participated in the qualitative study reported in chapter 3 and pilot phase to review the decision aid. The eligible participants were

randomized to receive the decision aid or no intervention. One-hundred and fifty-two women (78%) completed pre-treatment questionnaires. Forty-two women did not complete measures because they did not respond ($n=19$), became pregnant ($n=13$) or withdrew ($n=10$). At post-treatment, 13 participants failed to return questionnaires due to withdrawal ($n=3$) or non-response ($n=10$).

The mean age for the intervention group was 31.95 ($SD=3.79$) and 31.10 ($SD=5.64$) in the control group (see Table 4.1). There were no significant differences between the intervention and control groups for age ($t(1,149)=-1.093, p = 0.276$) or any of the demographic or outcome measures (see Table 4.2), with the exception of whether the diagnosis of MS had complicated their decision of whether to have children ($t(1,149)=1.998, p=0.046$). Women in the intervention group were more likely to feel that MS had complicated their decision. Therefore all ANCOVAs were conducted controlling for this variable.

Table 4.1

Demographic Variables

Measures	Decision-Aid Group (n=78)		Control Group (n=61)	
		Mean (SD)		Mean (SD)
Age		31.95 (3.79)		31.10 (5.64)
Time since Dx		5.15 (3.45)		6.22 (4.19)
		N		N
Have children	Yes	45 (54%)	Yes	29 (43%)
	No	39 (46%)	No	39 (57%)
MS type	Relapse Remitting	75 (89%)	Relapse Remitting	63 (92%)
	Secondary Progressive	2 (2%)	Secondary Progressive	0
	Primary Progressive	1 (1%)	Primary Progressive	0
	Don't Know	6 (7%)	Don't Know	5 (8%)
Wanted kids	Yes	69 (82%)	Yes	52 (76%)
before Dx?	No	6 (7%)	No	1 (1%)
	Unsure	9 (11%)	Unsure	15 (22%)
Dx complicate	Yes	64 (77%)	Yes	42 (62%)
decision	No	19 (23%)	No	26 (38%)

Table 4.2

Outcome Measures

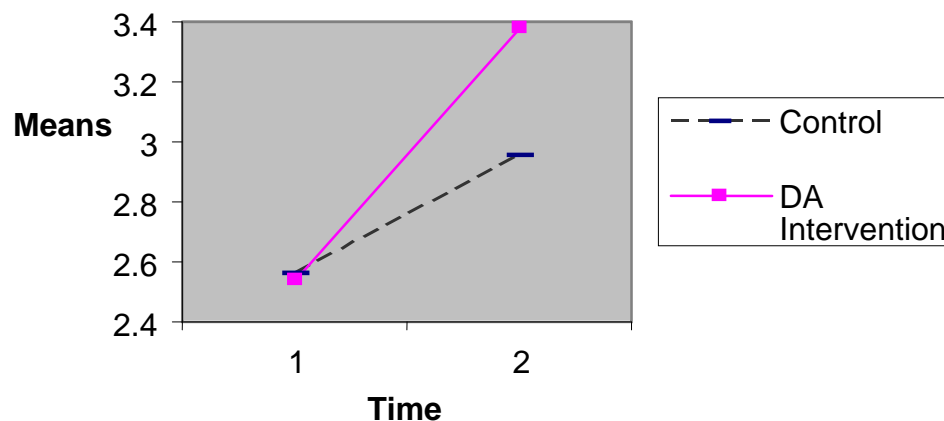
	Decision-Aid Group		Control Group		Effect Size
	PRE	POST	PRE	POST	
	mean (SD)	mean (SD)	mean (SD)	mean (SD)	
Knowledge	4.12 (1.81)	6.30 (2.18)	4.19 (1.93)	4.63 (1.86)	0.89
Decisional Conflict	2.48 (0.69)	2.07 (0.55)	2.34 (0.65)	2.29 (0.64)	0.72
Decision Self- Efficacy	77.69 (17.56)	86.09 (12.51)	82.23 (16.21)	82.56 (18.36)	0.51
Certainty	2.47 (2.04)	3.34 (1.85)	2.66 (1.84)	3.00 (1.81)	0.28
Balance	1.19 (2.98)	1.08 (3.68)	2.13 (2.44)	1.79 (3.03)	
Depression	14.74 (11.21)	13.09 (9.25)	13.95 (11.61)	12.48 (10.17)	
Anxiety	35.82 (11.98)	36.17 (12.03)	34.69 (11.59)	35.34 (11.35)	

4.3.2 Intervention Effects

The statistical analyses for this chapter are included as Appendix G. For certainty of the decision, there was a main effect for time at post-treatment, indicating that the passage of time significantly improved the certainty of all subjects, regardless of which group they were in ($F_{(1,135)}=8.739; p=0.004; \eta^2=0.061$). There was no main effect for group ($F_{(1,135)}=0.493; p=0.484; \eta^2=0.004$). However, there was a significant interaction effect, such that those in the intervention group shifted towards a more certain choice with regard to either choosing to have or not to have children, while the control group tended to remain relatively less sure ($F_{(1,135)}=2.854; p=0.047; \eta^2=0.021$) (See Figure 4.1). Although women became more certain of their decision, the decision aid did not systematically change women towards wanting or not wanting a child. That is, for the raw score on this continuum, there were no main or interaction effects ($F_{(1,135)}=0.079; p=0.779; \eta^2=0.001$).

Figure 4.1

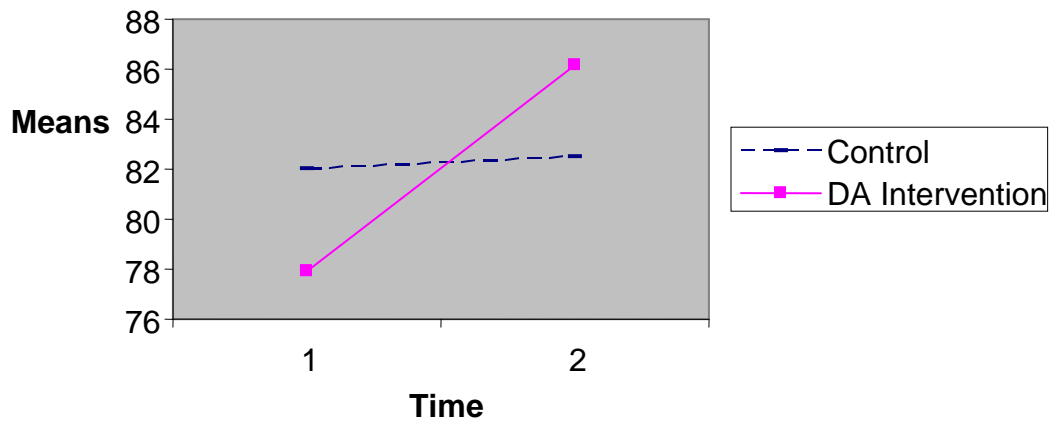
Continuum (absolute value)



For decision self-efficacy, the main effects for time ($F_{(1,135)}=2.486;p=0.117;\eta^2=0.018$) and group ($F_{(1,135)}=0.006;p=0.936;\eta^2=0.001$) were not significant. However there was a significant interaction effect ($F_{(1,135)}=8.375;p=0.002;\eta^2=0.058$) suggesting that women in the intervention group became more confident in their decision over time (See Figure 4.2).

Figure 4.2

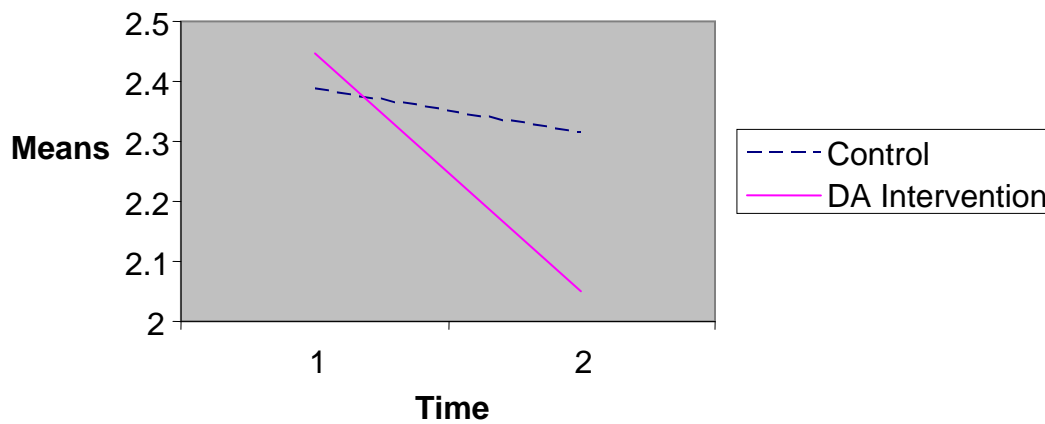
Decision self-efficacy



Between the pre and post assessments, decisional conflict reduced significantly in both groups (main effect for time: $F_{(1,133)}=11.997; p=0.001; \eta^2=0.083$). There was also an interaction between group and time indicating that decisional conflict was improved more over time in the decision aid group ($F_{(1,133)}=10.820; p=0.001; \eta^2=0.075$) (see figure 4.3). There was no main effect for group ($F_{(1,135)}=1.211; p=0.273; \eta^2=0.009$).

Figure 4.3

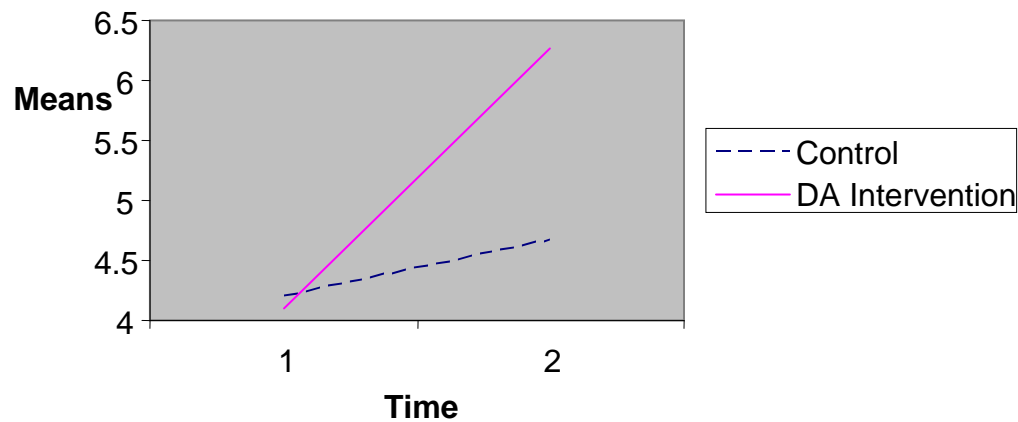
Decisional Conflict



At post-treatment, both groups showed a significant improvement in their knowledge regarding pregnancy in MS ($F_{(1,133)}=8.215;p=0.005;\eta^2=0.058$). There was also a main effect for group ($F_{(1,133)}=6.774;p=0.010;\eta^2=0.048$). Importantly the group x time interaction was also significant ($F_{(1,133)}=19.883;p<0.001;\eta^2=0.130$). That is, prior to treatment there was a small non-significant difference between the groups. Over time, both groups improved in knowledge, but this change was significantly greater in the group who received the decision aid (See Figure 4.4).

Figure 4.4

Knowledge



Levels of depression were not effected by the passage of time ($F_{(1,133)}=1.744;p=0.189;\eta^2=0.013$) nor were they different between the groups ($F_{(1,133)}=0.003;p=0.954;\eta^2=0.001$) and there was no significant interaction effect ($F_{(1,133)}=0.001;p=0.973;\eta^2=0.001$). A similar pattern resulted in levels of anxiety, with anxiety remaining unaffected by time ($F_{(1,133)}=0.291;p=0.590;\eta^2=0.002$) or group

($F_{(1,133)}=0.019$; $p=0.890$; $\eta^2=0.001$). As hypothesised, the interaction between time and group was also not significant ($F_{(1,133)}=0.001$; $p=0.991$; $\eta^2=0.001$).

4.3.3 Clinical significance

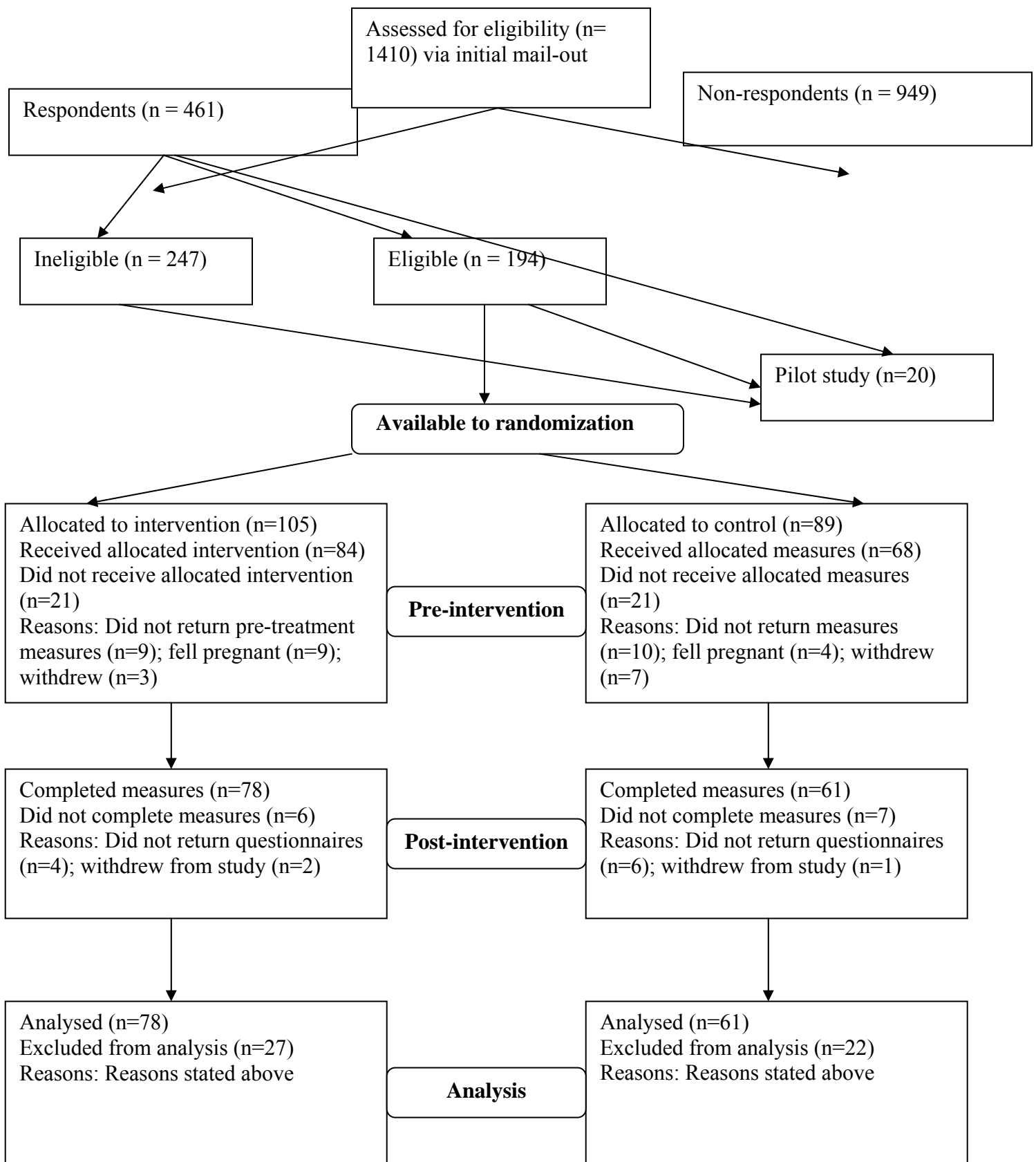
In addition to these analyses, it was also important to determine whether significant changes are of clinical significance. For this reason, scores on the Decisional Self-Efficacy Scale were reanalysed using a cut-off difference of 25% as indicative of a significant change in self-efficacy. For example, a participant who obtained a score of 50 at pre-intervention would have to score either below 25 or above 75 to indicate a clinically significant change in decisional self-efficacy. Scores on the Decisional Conflict Scale were similarly reanalyzed such that a change in score of 1, that is 20%, or more indicated a significant shift in decisional conflict. With regard to assessing the clinical significance of knowledge, conceptually it seemed that a score of 7 out of 10 reflected adequate knowledge about the subject matter to make a relatively informed choice. This cut-off score was decided a priori. With this in mind, both pre and post treatment knowledge scores were reanalysed to indicate whether subjects had adequate or inadequate levels of knowledge regarding the subject matter.

For self-efficacy, analyses indicated that there was no clinically significant change between the decision-aid intervention group and the control group ($X^2=4.475$; $df=2$; $p=0.107$). For decisional conflict, analyses revealed a clinically significant difference between the groups ($X^2=6.467$; $df=2$; $p<0.05$). Examination of the resulting proportions in each category indicates that whereas 7% of the control group became more conflicted over time, this was not true for anyone in the intervention group. In both groups, 87% remained the same and

only 7% in the control group became less conflicted, whereas 13% of the intervention group became less conflicted with regard to this decision. Therefore there are differences between the groups although clinical differences occurred in only a minority of women, but small, statistically significant changes were evident. With regard to subjects' levels of knowledge about MS and pregnancy, results revealed that at pre-intervention there was no significant difference between the groups, with 91% of the control group and 90% of the intervention group scoring less than 7 out of 10 on the knowledge measure ($X^2=0.123$; $df=2$; $p=0.940$). However, at post-intervention a clinically significant difference was revealed between the two groups with 47% of the intervention group having adequate knowledge about MS and pregnancy, as compared to only 20% in the control group ($X^2=11.147$; $df=1$; $p<0.001$).

Figure 4.5

Flow diagram



4.4 Discussion

The aim of this study was to evaluate a decision aid for women with MS in making the motherhood decision. We hypothesised that the decision aid would result in increased knowledge, self-efficacy and certainty and a reduction in decisional conflict. We also hypothesised that there would be no effect of the decision aid on anxiety or depression. These hypotheses were all supported. Similarly, there was no evidence of a difference in the direction of the decision, suggesting that the decision aid was balanced.

Despite careful attention to the study design and methodology there remain a number of limitations that need to be acknowledged in interpreting the results. Firstly, the study had substantial reliance on self-report. The demographic questionnaire, in particular, required women to report details such as the number of years since their diagnosis, their type of MS and whether they had wanted to have children before receiving their MS diagnosis. A number of women were not sure of their type of MS, which is concerning because it suggests that they are going through the process of making their motherhood decision with an absence of information about their individual illness and prognosis. The decision aid was also self-administered. The fact that the decision aid was effective with minimal face-to-face contact suggests that as an informational resource it is useful. It is more likely that in practice a tool such as this would be discussed face-to-face between health professional and patient. Its efficacy when administered in this manner has not been tested. However, research has consistently found that women with MS contemplating motherhood report no available information to help make an informed decision (McNary, 1999; Smeltzer, 1994, 2002). Hence, the provision of information in and of itself is likely to be important.

Although we had a high rate of recruitment and retention in this phase of the research, this was of those who initially responded to the mail-out, where a considerably lower response rate was achieved (33%). Therefore, it is possible that these women were more interested in this topic than other women with MS and may not be representative of this population. Nonetheless, presumably it is these women who would request supplementary information regarding MS and motherhood and therefore, those most likely to receive the intervention in clinical practice.

The final limitation is that while there were no differences between groups on any of the outcome variables or demographic variables, there was a difference between the groups in one question: whether MS had complicated their choice to have a family. In all other respects, randomization was effective and the groups were well matched. Further, in our analyses, we controlled for this variable. To ensure that these results were robust with and without this covariate, we analysed the data using ANOVA also and the pattern of results remained unchanged.

Women who had experienced a miscarriage during the treatment period were confused about where to place themselves with regard to the decision continuum at the post-intervention. While these women had made the decision to have children, they felt the likelihood of carrying a pregnancy to term was low. This highlights the issue that for some, the decision is taken out of their hands by physical realities.

These limitations notwithstanding, this study also has a number of strengths. Firstly, we developed a decision aid according to the C.R.E.D.I.B.L.E. criteria, developed by the

Cochrane Systematic Review of Patient Decision Aids (O'Connor et al., 2003).

Additionally, we consulted the Patient Decision Aid Checklist of the new International Patient Decision Aid Standards (Elwyn et al., 2006). The decision aid provides information about the positive and negative outcomes associated with each option to allow effective decision-making. It presents information in an unbiased, understandable way, using diagrams. The decision aid has methods for clarifying patient values and includes worksheets and questions that facilitate communication with health professionals and family members. The decision aid was systematically developed by researchers with appropriate credentials, and the area was thoroughly researched to determine needs (chapter 2). It was reviewed by potential users and experts and field tested with users (chapter 3). The decision aid was acceptable, balanced and understood by users. The decision aid was up-to-date with scientific evidence that was cited in a reference section and it used patient stories representing different choices. Finally the decision aid was shown to be effective in facilitating decision-making (chapter 4). These results are much more compelling indicating that nearly half the women versus less than a quarter are well informed after reading the pamphlet three weeks prior.

Importantly, women have consistently indicated that they have been unable to access an up-to-date informational resource about MS and pregnancy to facilitate appropriate decisions about their families (McNary, 1999; Smeltzer, 1994, 2002). This is problematic since women report that health professionals often have negative attitudes towards pregnancy and MS and hence there is considerable scope for the dissemination of misinformation. This decision aid is a first step towards the provision of an up-to-date resource that women can use with their partners and health professionals to facilitate decision-making.

Since this study, the MS Society of NSW has been using it as a resource for members of their society. The decision aid has been re-branded by the MS Society, advertised in their newsletter and made publicly available. Hence, there is a direct clinical benefit to patient care. As this is the first application of a decision aid to what is primarily a lifestyle choice, rather than a decision to have or forego a treatment or to determine between two treatment options, the question arises as to who would be an appropriate health care professional to administer the decision aid. Given that a number reported taking the decision aid to their general practitioners or neurologists, it would seem that they would be appropriate health care providers to introduce patients to this resource. However, MS Societies internationally employ a range of psychologists, social workers, nurses and other health professionals who provide counseling and support to women with MS and these practitioners may also find it helpful in initiating discussions about these issues.

Secondly, this decision aid could provide an important template for broadening the use of decision aids to other groups of women whose reproductive choices are affected by a medical illness. Such groups may include women with Systemic Lupus Erythematosus (SLE), infertility or who are HIV positive. Health professionals and researchers working with these women would need to adapt and evaluate the educational component in the pamphlet, however, many of the values exercises would remain similar. Indeed, health professionals working in these areas, where decision aids are yet to be available, could use these exercises to structure counseling sessions that focused on the motherhood choice.

Most decision aids are related to decisions to have or forego a particular intervention (e.g. screening in prostate cancer), or to decide between two treatments (e.g. mastectomy versus lumpectomy in early breast cancer) (Volk & Spann, 2000s). However, the motherhood choice, while being similar to the latter type of choice, relates more to a lifestyle choice that is complicated by a medical illness. While this study has developed an evidence-based decision-aid for women with MS that can be directly translated into practice, it also has the potential for wider applicability. It provides a direct model for other conditions where reproductive choices may be complicated by illness (eg SLE, infertility, HIV etc). Future research is needed to test the efficacy of decision-aids in other areas.

There is a need for future research to explore the relationship between stated preferences and actual behaviour to determine the effects of the decision aid in the longer term. A follow-up study would be useful to discover whether these women go on to have children or not, and experience less anxiety about risk during pregnancy and child-rearing.

In summary, we have developed a decision-aid to help women with MS consider issues relevant to their decision whether or not to have a family. The decision-aid was developed according to the gold standard CREDIBLE criteria and was acceptable to women in the study. The decision-aid was efficacious in improving women's certainty of their decision, their decisional self-efficacy and reducing their decisional conflict and there were no negative effects in terms of psychopathology. Hence, this decision aid can be readily used with women with MS contemplating motherhood.

CHAPTER 5: THE MOTHERHOOD DECISION AID: MECHANISMS OF TREATMENT

5.1 Introduction

Motherhood is arguably a decision that requires careful thought. Once the initial decision to have a child has been made, other family planning issues arise such as how many children to have, what size gap to leave between siblings and whether to terminate or continue should an unplanned pregnancy occur. When planning a family, there are many social, economic and emotional decisions that must be considered (Sevon, 2005) and having to factor in a chronic, unpredictable illness increases the complexity of the decision.

Multiple Sclerosis (MS) is the most common chronic neurological disorder among young adults. MS is largely progressive and unpredictable in nature, occurring more often in women in their reproductive years (Confavreux, Hutchinson, Hours, Cortinavis-Tourniaire, & Moreau, 1998). Women with MS are often faced with decisional conflict regarding the motherhood decision as the already physically and emotionally demanding time of pregnancy and childbearing may be exacerbated by a potentially disabling illness. For many women, the decision is not simple, and decisional conflict may be significant. Their choice must balance their family aspirations and career goals, and are further complicated by an illness characterised by uncertainty.

Women with MS considering pregnancy and childrearing need to consider not only the normal stressors associated with parenthood, the wellbeing of the baby and their confidence in being a good parent (Chalmers, 1982) but any possible effects of MS on pregnancy and the effect of pregnancy and the post-natal period on MS (Smeltzer, 1994). However, the majority of the research, to date, has focused on women who have already decided to have children. (e.g. (McNary, 1999; Smeltzer, 1994). Hence, the issues that are relevant to women who are currently considering this decision have not been adequately addressed.

In chapter 3, we interviewed 20 women with MS from various stages in the decision-making process. We confirmed the importance of previously identified themes. For example, some women had experienced negative attitudes from health-care providers or family and friends towards having children when they have a chronic, progressive illness. Women also voiced concerns about their own health and well-being as well as the well-being of their child. Another source of anxiety that affected the decision-making particularly of women who were undecided or who had decided not to have children was their ability to cope with the demands of parenting due to their MS. This related not only to whether to have children but also to when they should have children. That is, women reported the pressure to have children while they are less disabled although their longer term prognosis was unclear. This was because they feared that they would become too disabled to have a child later on. However, this pressure to bring forward child-bearing also had negative impacts on them, such as creating a stressful impact on their relationships as well as their own psychological well-being. Finally, women who had not had children were worried about the possibility of the child inheriting MS, despite the fact that this outcome was very

unlikely. However, for women who had already had children, these fears were outweighed by their experiences of being a mother, the meaning they have found in their role and the bond they have formed with their child. Finally and importantly, most of the women in the study agreed that currently their concerns were not being addressed adequately through the health care system.

The fact that women do not consider that they can easily get information about pregnancy and childrearing in MS is important because there are a lot of medical information that they need to facilitate informed decision-making. For example, many medications used to treat MS are contra-indicated during pregnancy and it is recommended that some should be stopped well in advance of conception. Hence, women must consider the impact of ceasing their medication on their MS. Practical difficulties also present and women worry about whether they will need a caesarean section, be able to have epidural anaesthesia during delivery or whether they can breastfeed (Ferrero, Pretta, & Ragni, 2004). Symptoms of MS in the post-natal period, during which women are at higher risk of relapse, can affect a woman's ability to care for an infant and perform parenting tasks in the short or long term. Finally, women worry about the risk of their child inheriting MS (Ferrero, Pretta, & Ragni, 2004).

Research suggests that, despite strong scientific evidence confirming the safety of pregnancy in MS, doctors remain reticent to offer advice on motherhood to women with MS (Smeltzer, 2002). Research also shows higher rates of termination of pregnancy amongst women with MS and that the number of women with MS who choose not to have children is higher than in the general population or other illness populations (Mueller, 2002). It has also been found that many women with MS have

avoided pregnancy and parenthood because of perceptions of doubt and disapproval from those around them (Wates, 1997). In chapter 2, the only large scale study assessing reproductive choices in women with MS to date, we found that 46% of a group of 461 women with MS aged between 20 and 40 were currently unsure about whether or not to have children. These results, taken together, confirm that the motherhood choice is not a straightforward decision for women with MS but that women do not feel that they are given sufficient access to accurate information to help them in their decision-making. In chapter 4, we conducted a randomised controlled trial of a decision aid, developed according to the CREDIBLE criteria (O'Connor et al., 2003) with 194 women with MS who were currently unsure whether or not they wanted children. They completed questionnaires measuring their decisional-conflict, self-efficacy, knowledge about MS and pregnancy, depression and anxiety as well as their motherhood choice at pre and post intervention. It was found that the decision-aid reduced women's decisional-conflict and increased their self-efficacy and knowledge of MS and pregnancy. The decision aid also resulted in a more certain choice that was not biased in either direction, and did not increase depression or anxiety.

As medicine has steered away from traditional paternalistic models towards models of shared decision-making, decision aids have been developed to support patients and their physicians in decision-making. They differ from usual patient information handouts and education materials by providing descriptions of various risks and benefits involved as well as discussing the likelihood of each outcome occurring (O'Connor et al., 1998). Decision aids have been shown to reduce uncertainty and decisional conflict in patients, increase their knowledge of the illness and the relevant

issues, provide more realistic expectations of outcomes and more consistency between patients' choices and values (Deyo, 2001). Shared decision-making assumes that the effectiveness of decision aids is because it allows patients to get relevant information and promotes communication between relevant parties (e.g. doctors and patients) regarding patient values and individual prognosis. However, while there is considerable evidence for the efficacy of decision aids (O'Connor, Roston, & Fiset, 1999), there is less information about the mechanisms through which patients find them helpful.

The aim of this study is to assess the long-term impact of a decision aid that was developed to help women with MS with their reproductive decision-making, using a quasi-qualitative framework. That is, we aim to determine whether the decision aid impacted on the way in which women approached their decision. It is expected that some women in both groups will have become pregnant and we aim to understand the way in which the decision aid may have changed their experience of pregnancy and/or early parenting. We predict that participating in the study as part of the intervention group will have had an impact on the motherhood choice for women with MS and that these women will be more certain of their choice as compared to the control group.

5.2 Method

5.2.1 Participants

The MS Societies of New South Wales and Victoria (Australia) maintain a data-base of members who have agreed to be contacted about research that is endorsed by the Society. The participants in this study were initially recruited (as described in chapter 2), via a large-scale mail-out to all women with MS aged 20 to 40 (n=1410), from this

database in January 2005. Four-hundred and sixty-one women responded (33% response rate), of whom 212 were currently undecided about whether or not to have a family. All women who were undecided (except 8 who took part in the qualitative phase reported in chapter 3) were invited to enter the RCT, 194 (recruitment rate = 92%) agreed (see chapter 4). These women were followed-up at 12 months for the current study. Unfortunately, only 106 women (55%) completed the follow-up (61 in the intervention group and 45 in the control group). Those remaining women who did not complete the study, failed to do so due to changed contact details or non-response after four failed attempts to make contact. Every woman with whom contact was made agreed to participate.

5.2.2 Procedure

We developed and evaluated in a randomised controlled trial, a decision aid with the aim of facilitating an informed choice that is in line with women's own personal values (O'Connor, 1995). In chapter 2, we completed a needs assessment which found that a decision aid would be useful for women with MS in making decisions about motherhood. In chapters 3 and 4, a preliminary version of the motherhood decision-aid was piloted amongst 20 women with MS from different stages of decision-making. These women attended focus groups after receiving a copy of the preliminary decision-aid and their feedback was sought. Women found the decision-aid to be comprehensive and reported that it covered most of the issues they would find relevant. Additional information women thought relevant was incorporated into the decision aid at this stage and it was then sent to 15 neurologists who commented on its accuracy. The details of the development of the decision aid are presented in

chapters 3 and 4. It was developed in accordance with the CREDIBLE criteria (O'Connor et al., 2003).

Women allocated to both the control and intervention groups in the randomised controlled trial, were telephoned 12 months after their initial participation in the study and completed a semi-structured interview over the phone which lasted approximately 5-20 minutes. Interviews began in July 2006 and finished in March 2007. All women were initially asked three questions: (1) whether their participation in the study had any effect on their decision; (2) if they had fallen pregnant since participating in the study; and (3) to indicate their current position on the motherhood choice continuum (outlined below). Interviews with women who had not fallen pregnant were terminated at this point, and those who had fallen pregnant (n=22) were asked further questions such as what factors influenced their decision, their use of the motherhood decision-aid in reaching a decision, whether they had any difficulties with their pregnancy and the post-natal period, their experiences as a result of having made a decision about motherhood, and the support they now require. These interviews lasted between 5 and 15 minutes. See Table 5.1 for the full list of questions asked during the interviews. The interviews were audio-taped and transcribed.

Table 5.1

Questions asked at follow-up

1.	You may remember when you last participated in this study, you were asked to place yourself on a continuum from –5 to 5 where –5 meant you definitely will not have children now or in the future and 5 meant you definitely will have children now or in the future and 0 meant you were unsure. Where do you see yourself in terms of this decision now?
2.	Did your participation in this study have any effect on your decision?
3.	Since you began participating in this study have you fallen pregnant?
4.	I wonder whether you could tell me a bit about the factors that influenced your decision, those that you weighed up in considering whether or not to have a family.
5.	[If in the decision aid group] Did you use the decision aid at all in reaching a decision? Do you think that the decision aid might have alerted you to some issues that you might not otherwise have considered?
6.	Did you have any difficulties with your pregnancy? Do you think that these were related to your MS?
7.	Have there been positive experiences that you have had as a result of deciding to start a family?
8.	What sort of support did you need when coming to your decision? Who did you talk with about the decision? Was anyone unsupportive of your choice?
9.	What kind of support do you require now as a new mother? Do you think that you would need this support regardless of whether you had MS? Is there any specific help that you need that you believe that you wouldn't need if you did not have MS?
10.	When you look back now at your decision to become pregnant, is there any issue that you would think about differently in making that decision now that you have experienced pregnancy and/or motherhood? Are you pleased with the decision that you made? Have you had any regrets?
11.	Some women find that the post-natal period is a difficult one for them? What was most difficult for you in managing the post-natal period? Do you think that it was made more difficult by the fact that you had MS? In what way? Many women find that their mood can be very low at times when trying to manage a newborn infant, did you experience any symptoms of post-natal depression? What were they?
12.	Have you and your partner thought about whether or not you might have more children?

5.2.3 Measures

As part of the follow-up questionnaire, women were asked to rate their certainty about having children on a continuum where –5 meant they would definitely not have children now or in the future, 0 meant they were unsure, and +5 meant they definitely would have children at some stage. Two scores were derived. Firstly, a score from -5 to +5 was given indicating the degree to which they definitely did want children. In addition, we calculated a score from 0-5, to indicate certainty of choice regardless of direction. For example, both -5/+5 was scored as 5 to indicate women were certain of their decision and 0 meant women were undecided.

5.2.4 Analysis

We compared completers and drop-outs using independent t-tests to determine whether there were any differences between the groups. Regarding the preliminary three questions administered to all participants, differences between the intervention and control group were assessed using parametric (for continuous variables) and non-parametric (for dichotomous variables) tests. Two (group: intervention; no intervention) x 3 (time: pre; post; follow-up) mixed model ANCOVAs were performed on the continuum measures, using SPSS 15 for Windows, to determine the effectiveness of the decision aid after 12 months.

Interviews with women in the intervention group were recorded and analysed qualitatively in order to identify the main issues. We analysed the data using Thematic analysis which is a method for analysing and reporting themes within data (Braun & Clarke, 2006). The process involved a number of phases. Firstly, the interpreters familiarized themselves with the depth and breadth of the content via a process of

rereading the data. Initial codes were then generated by identifying features of the data that could be assessed in a meaningful way. The data was then collated into a number of themes.

This study had ethics approval from the University of Sydney Ethics Committee and Multiple Sclerosis Australia.

5.3 Results

5.3.1 Demographics

One hundred and six women with MS, between the ages of 20 and 40 agreed to participate in the study. The mean age of women in the intervention group was 33.22 (SD=3.69) and 32.93 (SD=4.38) in the control group. The mean age of the women who completed the study (to 12-month follow-up) was 31.98 (SD=3.99). There was no significant difference in age between the women who completed the study and those who did not ($F=0.297$, $p=0.586$). Women in the intervention group had been diagnosed, on average, 5.55 years ago (SD=3.39) and 6.43 (SD=4.29) in the control group. Out of the women who completed the follow-up, the mean time since diagnosis was 5.93 years (SD=3.82) and there was no significant difference in time since diagnosis between the women who completed the study to follow-up and those who did not ($F=2.279$, $p=0.133$). Ninety-three percent of women in this study had relapse-remitting MS, 1% had secondary-progressive MS, another 1% had primary-progressive MS and 5% were unsure of their MS type.

5.3.2 Quantitative Results

All statistical analyses for this chapter are included as Appendix H. For women's certainty over their motherhood decision, there was a main effect for time at follow-up, indicating that the passage of time since post-treatment significantly improved the certainty of all subjects, regardless of which group they were in ($F_{(1,102)}=6.464; p=0.013; \eta^2=0.060$). There was no main effect for group ($F_{(1,102)}=1.041; p=0.310; \eta^2=0.010$) and no interaction effect ($F_{(1,102)}=0.851; p=0.358; \eta^2=0.008$). With regard to the raw score on the decisional continuum, there was a main effect for time since post-treatment, indicating that with time, both groups wanted children less ($F_{(1,102)}=4.101; p=0.045; \eta^2=0.039$). There were no group or interaction effects.

At follow-up, significantly more women in the decision aid intervention group indicated that their participation in the motherhood choice study had an effect on their decision ($\chi^2=14.662, df=1, p=0.000$).

Women were also asked whether they had fallen pregnant since participating in the study. Seven women in the control group had fallen pregnant and 15 in the intervention group. Results indicated that there was no significant difference between the groups in the number of women who fell pregnant during the study ($\chi^2=1.410, df=1, p=0.235$). Those women who had fallen pregnant since first participating in the study were asked whether they had experienced any difficulties with their pregnancy and if so, whether these were due to their MS. Only eight women had experienced difficulties out of 22, including four women who had miscarriages. Three (3/7; 43%) of these women who had miscarriages were in the control group, with one woman in

the intervention group (1/14; 7%) reporting her third miscarriage since participating in the study. The number of miscarriages occurring between the two groups ($\chi^2=3.86$, $df=1$, $p=0.055$) failed to reach significance, although, closely approached significance and was in the direction of fewer miscarriages in the intervention groups. Other difficulties experienced included extreme fatigue, morning sickness and swelling in the hands and feet. With regard to MS-specific difficulties, one woman reported two relapses and another took longer than usual to achieve full remission from an attack.

5.3.3 Qualitative Interview

The qualitative interview highlighted a number of mechanisms of change in decision-making, which the participants identified as being helpful. The following are the main themes that emerged as facilitating decision-making from the qualitative analysis:

5.3.3(a) Provision of Comprehensive Information

A common response amongst those who used the decision aid was the amount of information about pregnancy in MS that it provided, which these women had previously found difficult to access. Women reported that previously they had tried to seek information from a number of sources but were unaware of the sorts of information that they needed and hence were unable to request it. Thus the decision aid served as a resource where relevant information could be found in a single place.

“It did give me better information about having a baby in MS and I know MS won’t be effected in the long term and I will always have the decision aid to refer to if I decide to have another child”.

“My husband and I are very into researching MS anyway but it was good to clarify things. It gave us extra information we hadn’t been able to access anywhere. It clarified nothing is for certain, but it was reassuring”.

“The decision aid enhanced my knowledge and I got information that I hadn’t thought of. It pointed out medical facts that I’d have to think about or be prepared for”.

5.3.3(b) Communication

An additional effect of the decision aid that women highlighted was the facilitation of communication between these women and their partners and healthcare professionals. In fact, some women sought information and support from other sources where they could discuss options with other women who had been in the same situation.

“It helped me to talk to doctors and the case studies were helpful because I didn’t know anyone with MS who had kids. I have now spoken and met other women with MS and kids and I’ve gone to the MS Society and my doctor so it helped me seek out more information”.

“The study prompted me to speak to the MS Society who put me in touch with someone else going through the same thing”.

“It was also good for my husband to read and my family because they were all worried that I was going to relapse”.

5.3.3(c) Utilising support

Women who had become pregnant highlighted the importance of identifying sources of support. All women identified their husbands as their main support. Most women also named their close family members, such as their parents or siblings, and friends as important figures. Some women spoke of the disapproval they had received from some friends or family members.

“My husband had to be happy with the decision. I looked at who would be around to help. My parents-in-law are close by and so are friends”.

“My husband, how he will help me, and how me getting sick will impact the family”.

“I see a psychologist regularly. My partner and I did research together and now he understands and helps me more. My mum has MS also and was very unsupportive, so was my grandmother, so we don’t speak now”.

5.3.3(d) Planning for potential difficulties

The decision aid outlined potentially difficult times, such as the post-natal period, which women reported enabled them to prepare and plan for problems. Some women had experienced post-natal depression (PND) with previous pregnancies and were aware of the risk of this occurring again. They sought the help of their healthcare providers and support network in planning how to proceed if they developed a relapse of PND. Other sources of potential stress were being unable to breastfeed if going straight back on medication was necessary and working out a strategy for night time feeds if fatigue became a

problem for the mother. This awareness empowered women to feel prepared for potential difficulties increased their self-efficacy in being able to cope.

“Since my first pregnancy was more difficult, I had morning sickness 3 times a week for 20 weeks, but this time I have begun exercise with a physio to help prevent pelvic instability and short term osteoporosis.”

“We have a lot of friends and family on standby and my husband is taking off one day per week for the rest of the year.”

“I had PND with my second child and I'm worried it will happen again, it has been suggested I go straight onto antidepressants once the baby is born.”

“Every new mum needs support, I go to mother's group and it's not only for people with MS. I also had PND after my first two children so I'm aware of the counsellor and doctor.”

“I had PND with my first two kids, so I'll seek counselling and go to the GP at the hospital if necessary.”

5.3.4 Impact of reaching a decision

Most women spoke of the gladness and excitement they felt at making a decision to start a family. Other positive experiences also included feeling less stressed once the process of deciding was over, and more in control. Women also described how the process of deciding to start a family had brought them closer to partners and family.

“Having the baby has been huge. We’ve had lots of support and happiness from others around us”.

“No betaferons for a year! Being a ‘normal’ person, and live a normal life for a while”.

“Less stress once the process of deciding is over. It was the only thing talked about for three months”.

“I have a stronger bond with my partner, we’re expanding our family and starting a new phase of life”.

5.4 Discussion

The aim of this study was to determine the qualitative impact of the motherhood decision-aid twelve months later and to determine the likely mechanisms of change. We hypothesised that women in the intervention group would be more certain of their choice compared to the control group. As predicted, women in the intervention group reported that participating in the study impacted their decision-making process more than women in the control group. The short-term results presented in chapter 4 found that women in the intervention group did become more certain as compared to the control group at post-treatment, however, at 12 month follow-up we found that this difference was no longer significant. Rather, after 12 months, both groups had become more certain of their choice. Interestingly, however, 12 months later women in both groups were less likely to want children.

This study also gathered qualitative data from women, who have been participants in the motherhood study since January 2005, regarding their experience of going through the decision-making process and the impact this study has had on the choices they have made regarding motherhood. Women in the intervention group reported the benefits of using a motherhood decision aid. They identified that the decision aid gave them information they had difficulty accessing and also prompted them to seek out more information from their health-care providers. This is important, because, the information in the decision aid is targeted towards population level statistics and not the individual prognosis and characteristics of the woman. Clearly, if the aid is helpful in prompting women to (a) understand what information they need to know; and (b) to gain this information through medical resources, then they will be able to access individually tailored information. Women also reported that it improved their communication with their partners by allowing them to use the decision aid to clarify the values of their partner. Again, since child-rearing is a decision that involves both parents, the fact that the decision aid is reported to have prompted discussions between couples is very encouraging.

In addition to allowing women to gain more information from their immediate environment, a number of women sought out opportunities to meet others in similar situations in order to gain additional support. Women reported feeling less isolated as a result. They reported that it was helpful knowing that there were other women in the same position as them. Women who had become pregnant described their personal experiences of having decided to have children and how important this has been to them and their own value system. Additionally, they were able to express their

difficulties, either with parenting or the pregnancy, and to share their insight into how this impacted on them personally.

Despite careful attention to the study design and methodology there remain a number of limitations that need to be acknowledged in interpreting the results. Firstly, the women recruited for this study were all members of the MS Society. It is possible that these would be people who are more likely to seek out information regarding their health and therefore would be more interested in a decision aid. This may mean that this group of women value the decision aid more than other women.

A second limitation is the drop-out rate. At 12-month follow-up only 55% of the sample who participated in the randomised controlled trial could be contacted due to change of contact details or being unavailable for a phone interview. This may indicate a response bias. The women who were unavailable for phone interview may have been unavailable for a number of reasons. It is possible that those who dropped out of the study were those who felt the study was no longer relevant for them. However, there did not appear to be any systematic difference between those who completed and those who did not. Further, every single woman who could be contacted agreed to participate in the study, suggesting that the women remained motivated and interested in the study. Nonetheless, although our initial randomized controlled trial was well powered, due to drop-out, power was reduced in the present study.

It is possible that some analyses that rely on low base rates are particularly affected by the limitations to power. For example, 23% (14/61) of women in the control group

became pregnant as compared with 16% in the intervention group (7/45). This difference was not significant. Perhaps more importantly, the number of women who miscarried in the total sample was very low since there were only 22 pregnancies (ie. 4 women), however three of these were in the control group (3/7), compared to only one (1/14) in the intervention group. This result was unexpected and narrowly failed to reach significance on two-tailed tests ($p = 0.055$). However, this may indicate a difference between groups which, if confirmed, would be important. The only plausible reason for a difference between groups would be due to the effect of medications in the pre-conception or early pregnancy period. That is, it is possible that women in the control group did not realise that some medications needed to be stopped prior to conception and this may have affected their ability to carry the pregnancy to term. While this is speculative and the finding did not reach significance and hence may be due to chance, future research should investigate the effects of MS medications on early pregnancy and pre-conception health.

In the long-term, women from both the intervention and control groups became less conflicted in their decision over time, however, those in the intervention group became more certain more quickly. Further, those in the intervention group maintained their certainty in the longer term. Since all women in the study volunteered, it remains a possibility that participation in a study about motherhood in MS triggered women to begin thinking about their options and seek out information, regardless of which group they were allocated to. Therefore it is likely that this process may, at least partially, explain why women in the control group became more certain over time of their choices also.

These limitations notwithstanding, this is the first study to develop and validate, according to the C.R.E.D.I.B.L.E. criteria (O'Connor et al, 2003), a decision-aid to help women with MS make family-planning choices. This follow-up study provides additional support for the value of a decision-making tool to assist women with MS in making decisions about motherhood. Specifically, women found the decision aid to be useful in providing information, facilitating communication, helping to foster support in their environments and to plan for difficult periods. This decision-aid is currently being utilised by MS Australia and is distributed to women who have questions about child-bearing and child-rearing. Thus the motherhood decision-aid provides a relatively simple, potentially low cost intervention that can enhance women's ability to make informed decisions about motherhood in the face of a chronic illness, such as MS. Moreover, it seems that educating health-care professionals in these issues may also have positive consequences for women with MS who are making family-planning choices. This also represents a tool that could be adapted to suit other medical conditions where reproductive decision-making may be complicated, such as Lupus, HIV or whether to go through or continue IVF. Our results suggest that this would be an important direction for future research to take.

CHAPTER 6: SUMMARY AND CONCLUSIONS

6.1 Summary

The aim of this thesis was to develop and evaluate a decision aid for women with MS in making decisions about motherhood. The study was guided by the C.R.E.D.I.B.L.E. criteria, developed by the Cochrane Systematic Review of Patient Decision Aids (O'Connor et al, 2003) and consisted of four, related studies.

The first study was a needs assessment, which aimed to determine the need for a decision-aid for women with MS in New South Wales and Victoria. It was hypothesised that the motherhood decision would have relevance to a large percentage of women with MS between the ages of 20 – 40 and this was confirmed in chapter 2.

The second study was a qualitative study that sought to explore the themes associated with motherhood that are relevant for a heterogeneous sample of women with differing views regarding the motherhood choice. It was expected that by representing the views of women who have yet to decide or have decided against having children, new themes would emerge in addition to themes already represented in the literature from pregnant women with MS. It was found that there were some notable differences in women's concerns depending on their stage of decision-making (see chapter 3). At this point, a review of the draft materials was also conducted by both experts and consumers (reported in chapter 4). The preliminary decision aid was found to be clearly written, accessible, balanced and relevant by potential consumers and accurate by experts.

Minor additions were made to the decision aid based on feedback and individualised personal stories based on the experiences of group members were included. A randomised controlled trial was carried out to test the efficacy of the final decision aid. It was hypothesised that the decision aid would increase knowledge about pregnancy and child rearing in MS, reduce decisional conflict and increase decisional self-efficacy. This trial also aimed to ensure that the decision aid was free from any adverse effects on psychopathology. Increases in anxiety or depression were not expected in women with MS as a result of the decision aid. All hypotheses were confirmed (see chapter 4).

The final study was a follow-up phase conducted 12 months after participants' initial involvement in the research. The aim of this final phase was to assess the status of women's motherhood decisions since post-intervention and to determine whether the decision aid impacted on the way in which women approached their decision. It was also expected that some women would have become pregnant and those women who became pregnant were asked about what impacted on their decision-making process and their experiences of being pregnant, regardless of whether they were in the treatment or control group. It was also anticipated that receiving the decision aid may have had an impact on the motherhood choice for women with MS and that women would remain more certain of their choice compared to women in the control group. The results showed that over time, women in the intervention group did maintain their certainty, but women in the control group also became more certain of their choice. At follow-up the difference in certainty was no longer significant between the two groups. However, women did report that the intervention was useful in (a) providing access to information previously unavailable, (b) facilitating communication between

women, their partners and health care professionals, (c) aiding them in considering and utilising their networks of support, and (d) preparing them for potential difficulties.

6.2 Methodological Problems

Despite careful attention to the design and methodology of the study, there were some limitations that should be borne in mind in interpreting these findings. Firstly, the generalizability of the results may be limited. The initial recruitment process of the study consisted of a mail-out to all female members of the MS Society of NSW and VIC who were aged between 20 and 40 (1410 women). The initial recruitment rate was modest (33%) and may potentially indicate a response bias. That is, that this issue was more prominent for those women who did respond and therefore, participants may not be representative of the population as a whole. In subsequent studies this response rate could be improved by sending a reminder letter or conducting a telephone reminder (Silva, Smith & Bammer, 2002). However, since ethical requirements mean that the MS Societies had to send the information and we were unable to access names due to privacy laws, we were unable to organize to follow up these participants in order to increase the response rate. This is most problematic in chapter 2, which aimed to document the need for such a decision aid and as a result the need may be over-estimated. However, 46% of the sample did feel currently undecided and hence the motherhood decision was still relevant to at least 15% of the possible sample, that is 212/1410 representing a significant need. With regard to the qualitative study presented in chapter 3, generalizability is not an issue since it is the depth of the individual's experience that is important to capture in qualitative research. Further, clearly in clinical practice one would administer the decision aid

only to those who were currently considering motherhood. Therefore it would be these women who would be expected to receive the intervention in clinical practice, as they would be most likely to seek information about pregnancy in MS and other supplementary material regarding MS and motherhood. As such it is likely that women similar to those who took part in the study will be offered the decision aid in the future. Hence, we can be reasonably confident that the decision aid will be effective for those women requesting information regarding MS and pregnancy based on the results of chapters 4 and 5.

A second limitation was the failure of randomization to equalize the groups on all potential confounders in the randomized controlled trial. The intervention and control groups were significantly different on one measure, that being whether their decision to have children was complicated by the diagnosis of MS. Results showed that significantly more women in the intervention group found their MS diagnosis complicated their decision of whether to have children than women in the control group. However, in all other respects, randomization was effective and the groups had no significant differences on any of the demographic variables. That is, there were no differences for age, gender, type of MS, illness duration, and whether they wanted to have children before their diagnosis or any of the outcome variables. Given the number of outcomes where differences were not observed in a study that had good power to detect even small differences between groups, this suggests that the groups were nonetheless relatively well matched and the one difference observed would have been expected solely due to chance. Nonetheless, in order to be conservative in the analyses in chapter 4, whether MS was felt to complicate the decision was controlled for. To ensure that these results were robust with and without this covariate, the data

was analysed using ANOVA also and there were no changes in the pattern of responses. Hence, it is unlikely that this difference effected results.

The intervention comprised of the decision aid as well as a follow-up telephone call to ensure that participants had read the decision aid, understood it and were not upset by it. However, this also provided the opportunity to clarify any issues that women had after reading the decision aid. This raises the question of whether the significant results achieved in the randomised controlled trial of the decision aid, were from the decision aid itself, or from the discussion with an intern clinical psychologist following receipt of the decision aid. This procedure was chosen because the decision aid should be given in combination with a personal discussion with a health professional and it was the intent of the research team to mirror how the decision aid would best be used in clinical practice. Further, it should be noted that during the telephone discussions, none of the women reported feeling any distress as a result of receiving the decision aid. Thus, there was no need for the intern clinical psychologist to utilise any therapeutic techniques that may have influenced the participants in any way. Women tended to discuss their thoughts regarding the decision aid and how it alerted them to issues they may not have otherwise considered. They also spoke about their own situations and how they felt the information presented in the booklet was relevant to them. The length of the discussions was relatively short (5-20 minutes), which likely reflects a similar time that would be available in a medical consultation. Nonetheless, the fact that the discussion contributes to the efficacy of the decision aid cannot be excluded.

While results in the randomised controlled trial indicated that there were no adverse effects of the decision aid on depression or anxiety, this does not rule out the possibility that some individuals may have responded adversely. The outcome for people who failed to complete post-assessment measures remains unknown. However, women were provided with a phone number to contact if they felt distressed at any point from their participation in the study and no one accessed this number.

The maintenance of the gains achieved from the decision aid remains in question. Although women reported the decision aid to be useful, there was not a significant difference on the only measure re-administered. Ideally, all the measures administered in chapter 4 could have been re-administered at follow-up. While this may have confirmed the long-term efficacy of the decision aid more clearly, we decided against this for two main reasons. Firstly, the drop-out rate was already relatively high and it was feared that with greater demands, an increased attrition might be expected. Secondly, we wanted to explore outcomes in a qualitative fashion because we were particularly interested in the mechanisms of the decision aid, which was more suited to a qualitative methodology. However, as a result we cannot conclude that the decision aid has long term effects. This raises the issue of the timing of intervention or when would be most effective to provide women with the decision aid. However, with regard to ecological validity, this study was an efficacy study and not an effectiveness study and further research is needed to determine the decision aid's effectiveness in clinical practice.

Another possible issue may be that it appeared as though the decision aid was directed at women only. Obviously it is ideal to have both the mother and father involved in

the preparation for childbearing and for the childrearing itself. Therefore, particularly where there is the presence of a chronic illness such as MS, it would seem that it is even more necessary for partners of these women to be aware of the relevant issues so that they may be involved in the decision-making process. The decision aid was designed in such a way as to present information about the physical changes that occur with MS and when pregnancy occurs in a woman with MS. It also presented information about what women may experience psychologically, from pre-conception to the post-natal period. This included considering those people around them, such as partners and other family members and there were exercises in the decision aid to guide women through this process. Further, during the conversations with women in the randomized controlled trial, as well as at follow-up, women commented on how the decision aid actually helped them in discussing the relevant issues with their partners.

Secondly, some women were not in a relationship at the time of the study, but for them the decision aid was still helpful in enabling them to understand the facts so that when they are ready to have children they have a resource available and know how to approach health professionals for advice. The decision aid, therefore, is a tool for use by both parents in helping them become aware of the relevant facts, as well as helping them plan for any difficult times that may arise from MS. It also serves as a useful source of information for any woman with MS when learning about their illness generally, in preparation for future family-planning. Nevertheless, future research should explore the information needs of partners of women with MS, and ways in which the decision aid might be modified to include them more.

6.3 Strengths

Despite these limitations this study also has a number of strengths. The decision aid was developed according to the already well-established C.R.E.D.I.B.L.E. criteria, developed by the Cochrane Systematic Review of Patient Decision Aids (O'Connor et al, 2003) and the new International Patient Decision Aid Standards (Elwyn et al, 2006). Following the CREDIBLE criteria, the decision aid was Competently developed with the inclusion of components that promote quality decision-making and was developed by researchers with appropriate qualifications. Further, a needs assessment was conducted to determine whether this would be a useful resource. Once established that a decision-making tool would be useful to women with MS for the motherhood decision, a draft was written and a pilot study undertaken to gather further information to improve the decision aid and gain feedback from the potential users. The draft was also sent to neurologists who were given an opportunity to write back with their feedback on its accuracy. The data in the decision aid is Recently updated, such that it includes information that is based on evidence available within the past two years. Of course the decision would need to be updated every two years in order to remain accurate. The decision aid is Evidence-based in that the information provided is based on scientific studies following a thorough literature search. There were no conflicts of Interest and the decision aid had Balanced presentation of options, benefits and harms and this was confirmed by women during the pilot study and the results of the RCT where women in the decision-aid group were not pushed in one or another direction. The final criteria is that the decision aid is Efficacious at improving decision-making. This criteria was met as its evaluation included a large randomised controlled trial which demonstrated that the decision aid is effective in improving knowledge, reducing decisional conflict and increasing decisional self-

efficacy. Further, it was found to be free of adverse effects, such as depression and anxiety.

The decision aid developed in this study is unique in that it involves a lifestyle choice that is complicated by a medical illness, rather than a choice to either have or forego a particular intervention (e.g. screening in prostate cancer) (Volk & Spann, 2000), or a choice between two particular treatments (e.g. mastectomy versus lumpectomy in early breast cancer). This study has shown that decision aids may be useful for lifestyle choices, and thus other decision aids may be developed to empower people with the ability to get information for other lifestyle choices, such as in vitro fertilisation (IVF). A final strength of this study is that the MS Society of NSW is now using the developed decision aid as an evidence-based tool, thus it has been directly translated into practice.

6.4 Importance of the Motherhood Decision in Women with MS

The first study in this thesis was a needs assessment aiming to determine the proportion of women with MS in New South Wales and Victoria for whom the motherhood decision was relevant. The Cochrane Systematic Review of Patient Decision Aids (O'Connor et al, 2003) developed the C.R.E.D.I.B.L.E. criteria to guide the development of decision aids and advises that a needs assessment be undertaken in order for the decision aid to be competently developed. A second aim of this study was to identify subjects for the later stages of this research project. The main findings are summarized below.

The majority of women (80%) in this study had been diagnosed with relapse-remitting MS, and of these, 50% were undecided about their motherhood choice. In contrast, only 4% of women had been diagnosed with secondary-progressive MS and of these, 19% were undecided about their motherhood choice. This result is not surprising given that secondary-progressive MS begins its course with a relapse-remitting pattern and as this study targeted women between the ages of 20 and 40, intuitively it would seem that women with secondary-progressive MS would represent an older sample of women, and hence been excluded at the initial mail-out stage. This assumption was confirmed in this study, as the women with secondary-progressive MS were older than those with relapse-remitting MS by an average of three years. Further, in chapter 5, it was confirmed that even over a period of only one year women do become less inclined to want children.

Results showed that out of the 1410 women who were sent the initial mail-out inviting them to take part in the study, 461 (33%) women responded and of those, 212 (46%) were currently undecided about their motherhood choice. Another 158 (34%) women indicated that they already had their children and did not want to have anymore. It is unknown whether women in this group received their MS diagnosis before or after they had their children, however it is likely that a significant proportion of these women were making their decisions about motherhood after having received their diagnosis. Thus, the motherhood decision would have been a significant process that they had previously been through.

According to Access Economics (2005), which draws on data from the Australian MS Longitudinal Study (AMSLS), over 16 000 people in Australia have MS. It also

indicates that of this number, 74%, (ie 11, 200) are women. The National MS Society Sourcebook (2005) estimates that in the United States of America, approximately 400 000 people have been diagnoses with MS, and as in Australia, women are two to three times more likely to be affected than men. Therefore there are 280 000 women in the US and 11 200 women in Australia with MS. Even if we were to assume that the motherhood decision was irrelevant to those 949 (67%) women who did not respond to the initial mail-out and only to the 15% of the total sample who endorsed uncertainty currently, the DA would still be relevant to 4200 women in the US and 1680 women in Australia. These numbers are likely to be an underestimate since there are likely to be many reasons for the 949 women who failed to respond, not all of which are due to the irrelevance of the topic.

6.5 Issues Relating to Women in Decision-Making

The second study in this thesis was a pilot phase where a preliminary decision aid was drafted and sent to 20 women with MS in the four stages of the motherhood decision. These stages were those who had no children, those who had children and were unsure about having more, those who already had children and did not want to have anymore, and those who had decided against having children. Focus groups were run with an aim to capture the main concerns of women in each group, and those women who had already been through this decision-making process were able to share their experiences and insight with those who had not. These results were presented in chapter 2.

For women who were undecided about their choice of whether to start or enlarge their families, there were common concerns about their own health and well-being. A new

theme that emerged was that women were hesitant about having children at a time when their own illness was giving them trouble and that many women did not want to have children until they had experienced a period of stability in their illness. Their rationale was that they needed to be as sure as they could that they would be able to cope with the demands of looking after a baby, especially given that the post-natal period has been shown to be a time of increased relapse rates (Cook, Troiano, Bansil, & Dowling, 1994).

Another common concern found in this study amongst women who were undecided was the well-being of the child. Women who had no children were particularly concerned that the child may end up having to care for their mother and they were adamant that they did not want to have children if this would occur due to their beliefs that this would be an unfair situation for any child. Worries about coping with parenting itself were very strong amongst women in this group also. Many felt that it would be wrong to have to rely on other people, even if only in times of an exacerbation, and that if they could not handle the task for the majority of the time then they should not be taking it on at all.

Women from all groups commented that they felt increased pressure when it came to the timing of this decision. Some women who had had children spoke in retrospect of the advice they had been given that they should have children sooner rather than later and this sometimes posed dilemmas in relationships that perhaps were not ready for children. Women who had no children were worried that they were getting older and that it may be more difficult in the future to fall pregnant, which made them feel stressed that they should be having children now. Finally, women spoke about not

wanting to miss the experience of parenting, and this was supported by those who had had children, in the sense that having children brought a sense of meaning to them and they valued their roles as mothers.

During this phase, the decision aid was also sent to 15 neurologists who were given an opportunity to respond with their feedback on the decision aid's accuracy. The decision aid was very well received by the neurologists, and positive feedback was gained. They reported that it was an easy to read resource that contained the most relevant information for women with MS and that it would be a valuable tool to use in conjunction with their consultation. Indeed, we were approached by one neurologist prior to completing the study to see whether he could get a copy to use with his patients. Additionally, the women in the focus groups gave feedback on the preliminary decision aid. They reported that they found the resource very accessible and helpful in that it collated information in one place. Many women who had had children already spoke about how difficult it was to access this kind of information when they were going through their motherhood choice, and that a booklet such as this would have been a valuable thing to have had at the time.

The results of this phase are significant because they indicate that women with MS are not only unsure of the relevant issues when making family-planning choices, but that they have difficulty accessing this sort of information. Further, communication between health care professionals and these patients was highlighted as an area that needs improvement. Women in this pilot study found the preliminary decision aid to be a valuable resource because it collated the information that they would need to

seek out for themselves and for many it was useful as a tool to guide discussions between themselves and their health care professionals and partners.

6.6 Intervention

The third study in this thesis aimed to test the efficacy of the motherhood decision aid, by determining if it increased knowledge about pregnancy in MS, increased decisional self-efficacy, decreased decisional conflict, and made women more certain of their choice. It also aimed to ensure that the decision aid was unbiased towards any particular view and that it did not increase depression or anxiety amongst women allocated to either condition. The main findings are summarised below.

Women in the decision aid intervention group were significantly more certain of their choice than those in the control group at post intervention. Furthermore, there was no systematic change with regard to wanting a child or not wanting a child in either of the two groups, thus indicating that the decision aid did, indeed provide women with an unbiased view of all the relevant information. It was also confirmed that the decision aid increased decisional self-efficacy amongst women in the intervention group, and reduced decisional conflict. Both groups had a significant increase in their knowledge about pregnancy in MS at the post-intervention stage; however the increase in the intervention group was significantly greater than the change in the control group. Finally, levels of anxiety and depression remained unaffected with no significant main or interaction effects.

Knowledge about pregnancy in MS increased amongst women in the intervention group as compared to the control group. It is highly likely that merely participating in

the study itself may have triggered questions amongst participants motivating them to seek information about the issues involved in their motherhood decision. However, despite this likelihood, the interaction effect between group and time was highly significant.

6.7 Follow-Up

The mechanisms for change within the decision aid lie in several areas that would account for the significant improvement in decisional conflict and self-efficacy in the intervention group. Firstly, support has been identified as a key consideration for women with MS when making this choice (McNary, 1999; Smeltzer, 1994, 2002) and this decision aid required women to consider their sources of support should they fall pregnant. They were specifically asked to consider what each person could do to support them not just after the arrival of a new baby, but also in making the decision as well as in the future. Rather than making assumptions, women were able to gain a clear picture of what constituted support for each person whom they considered supportive.

Secondly, in participating in the decision aid intervention, a large proportion of this group of women ended up sharing decision-making with their health team. In accordance with the shared decision-making model, the patient received all necessary information regarding risks and benefits and alternative options available (Braddock, Edwards, Hasenberg, Laidley, & Levinson, 1999; Ford, Schofield, & Hope, 2003; Ruland & Bakken, 2002). The patient then also had the opportunity to communicate their values and preferences to their healthcare professional. In this case, communication between the individual and her partner or significant others as well as

family was encouraged. Additionally, shortly after receiving the decision aid, each woman was contacted by an intern clinical psychologist and given the opportunity to discuss any concerns or issues raised. The motherhood decision aid was designed with the intent of being used in conjunction with open communication between the individual with MS and her healthcare team. When women were contacted for discussion, many reported that they had read the information and subsequently decided to discuss their options further with their neurologist or GP.

Thirdly, the motherhood decision covers a range of issues, such as how many children to have and how much time to leave between children. Some people do not consider the additional decisions that are incorporated within the motherhood choice and the decision aid brings attention to these decisions as an additional way of enabling participants to plan for their futures. Women were asked to list the pros and cons of not having children, having a smaller family than they had originally planned, leaving a larger gap between children and proceeding as they had always intended, regardless of their medical condition. This method allowed women to consider their own personal values associated with each pro and con and rate how important this was to them.

A major concern for women was the possibility of passing on their MS to their baby. Despite research showing the small chance of this occurring, and the advice given to women who seek information about this possibility from their neurologists, it remains one of the main concerns in women with MS considering motherhood. The decision aid addressed by issue by means of a visual aid enabling the participants to compare the probability of inheriting MS if neither, one or both parents has it. Research has

suggested that the use of visual aids facilitates communication and assists with information-processing (Ahmed & Boisvert, 2003). Therefore this method directly addressed this concern for women and highlighted the realistic likelihood of this outcome in such a way that is easily comprehended.

6.8 Future Research

There is a need for future research to explore the relationship between stated preferences and actual behaviour to determine the effects of the decision aid in the longer term. Most decision aids refer to decisions that are taken immediately (Elwyn et al, 2006). Such decisions are usually one-off and when a decision is made, the treatment is pursued. Hence, there are few follow-up studies of decision aids, since the decision aid aims to affect the current decision. However, the motherhood choice is ongoing and can be revisited on numerous occasions. A number of follow-up studies, for example at two, five and ten years, would be useful for the purpose of discovering whether these women go on to have children or not. However, given the difficulty in accessing women in this study, drop-out is likely to be a problem.

In this study, women were asked during the pilot phase, the telephone conversations in the randomized controlled trial and at follow-up, about their hesitations in having children and what their associated concerns were. At follow-up, many women reported that they no longer saw their decision to have children as risky, and that the decision aid helped them to gain perspective. This idea of perceived risk is an important issue as it illustrates the importance of people having an accurate understanding of the issues related to their illness and highlights the need for consultation with health professionals about the relevant issues relating to specific

lifestyle choices. It would be useful for future research to look into the issue of perceived risk to determine if, after using the decision aid, women still attach the same level of risk to going through with pregnancy and child-rearing.

While this study has developed an evidence-based decision aid for women with MS that has been directly translated into practice, it also has wider applicability. Future research is needed to test the efficacy of decision aids in other areas. Systemic Lupus Erythematosus (SLE or Lupus) is another chronic autoimmune disease that is more prevalent in women than men (9:1 ratio) (Goodman, Morrissey, Graham & Bossingham, 2005). Like MS, it too is progressive in nature and treated symptomatically with corticosteroids and immunisuppressants, though it is more likely than MS to be associated with early mortality. Research has found that patients with Lupus experience similar psychological symptoms to those with MS, such as depression, anxiety and adjustment problems and also experience feelings of uncertainty and fear of death (Goodman, Morrissey, Graham, & Bossingham, 2005). Lupus also tends to onset prior to age 40 when child-bearing decisions are usually made. Since there is an established need for a decision aid for women with MS in making family-planning choices, it would seem likely that a similar need would exist for other chronic illnesses that onset relatively early in women, such as Lupus, where family-planning choices are likely to be compromised.

Another example is infertility, which is a medical condition that affects the ability of some couples to conceive (Peterson, Newton, Rosen & Schulman, 2006). One treatment option is in vitro fertilization (IVF) (Hjelmstedt, Widstrom & Collins, 2006). However, IVF is not without side-effects and disappointments and up to 40%

of couples fail to conceive (Eugster & Vingerhoets, 1999). The results of this study suggest that such complicated lifestyle choices can be adequately addressed in a decision aid and future research could develop a resource for couples contemplating IVF.

This decision aid provides a direct model for other conditions where reproductive choices may be complicated (e.g. Lupus, infertility, HIV, etc). Clearly, the educational components of the decision aid will be different, but the sections that relate to values and different options could form the basis of future applications. Clearly, information will need to be targeted to the particular illness. For example in Lupus the risk of miscarriage is greater than in healthy people. However, the issues of values and expectations are similar across illnesses. Obviously the efficacy of modified versions would need to be assessed.

There also exist broader implications for the development of the motherhood decision aid. For example, it represents a tool that may be adapted to suit other illnesses, for example HIV/AIDS or Sickle Cell Disease. With these illnesses there is a higher risk of genetic transmission (Asgharian, Anie, & Berger, 2003; Heard, Sitta, & Lert, 2007) than with MS so while we cannot generalize from these findings as to whether the diagnosis creates ambiguity regarding reproductive choices, it is likely that providing these patients with information to enable informed decision-making, as well as to facilitate communication between patients, their carers and their treating team would be of significant benefit.

6.9 Clinical Implications

The development of a decision aid for women with MS in helping them to make decisions about motherhood, raises several considerations for its use clinically. Firstly, there is the question of when the right time is to administer the decision aid. Given that the majority of women are diagnosed with MS between the ages of 20 and 40, it may be valuable to provide this resource to women when they are newly diagnosed, allowing them to be already equipped with information when they come to making family-planning choices. However, given that a diagnosis of MS has been found to be associated with depression, anxiety and symptoms of post-traumatic stress disorder (Beatty, 1993; Chalfant, Bryant, & Fulcher, 2004; Feinstein, 2002), raising the issue of motherhood may, in reality, be an additional stressor for women at this already distressing time. It could, for example, increase women's perception that this is a decision with a pressure of time.

Further, this study has assessed efficacy and not effectiveness, therefore whether health care professionals would need additional training to administer it is an empirical question. Linked to this issue is the question of who would be an appropriate health care professional to administer the decision aid. Given that a large number of women in this study took the decision aid to their general practitioners or neurologists, it would seem that they would be appropriate health care providers to introduce patients to this resource. What may be even more helpful would be for general practitioners and neurologists to liaise with clinical psychologists and clinical nurse consultants who could support the patient with some of the more emotional aspects of the decision, such as who they would be able to turn to for support, as well as for facilitating discussions between these women and their partners and families.

An issue of relevance raised in this study is the pressure women have felt, either from their health care providers, partners or themselves, in coming to a decision. This is particularly significant given the current trend of delaying motherhood by having babies later and the related issue of conception sometimes taking up to a year (Chapman, Driscoll, & Jones, 2006). For example, at age 40, it takes on average 9-10 months for a couple to conceive in comparison to 4 months for women at age 30 (Sydney IVF, 2007). This is particularly difficult for women with MS needing to be without medication while contemplating pregnancy, that is, trying to conceive. Intuitively it would be stressful for any woman trying to establish their career and financial security to receive a diagnosis of an illness which may cause them to feel like they must have children sooner than they would prefer out of fear of it being more difficult later or because of the possibility of increased disability. This decision aid would be helpful in this instance because it could help to create realistic expectations of outcomes regarding future disability as well as assisting women to plan with their partners for times of difficulty. Additionally, providing information to women that educates them about the impacts of MS on pregnancy and vice versa should also ease some distress because it allows them to have a clearer picture of what to expect.

6.10 Conclusion

In conclusion, this thesis has developed and evaluated a decision aid for women with MS to help them make decisions about motherhood. This study found that a decision aid is needed for these women, and when it was trialed, it was found to be effective without adverse effects. That is, the decision aid reduced decisional conflict, increased decisional self-efficacy and knowledge, made women more certain of their choice and

did not bias them towards a particular choice. Further, the decision aid did not cause increased levels of anxiety or depression. This decision aid therefore, represents a useful tool that not only helps women obtain relevant information but also equips them with a resource that guides discussions with their health care providers and aids communication with their partners. While evidence of the long term impact is not conclusive, the motherhood decision aid has been put into practice as an available resource used by the MS Society of New South Wales, Australia. It is the first decision aid of this kind, and may be easily adapted by other countries.

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APPENDICES

APPENDIX A:
ETHICAL APPROVAL



Department of Psychology
The University of Sydney, N.S.W. 2006

PARTICIPANT INFORMATION SHEET

“The Motherhood Decision: Designing a Decision Aid for Women with MS”

This information sheet has been prepared to provide you with information about the study you have been asked to take part in. This study aims to develop a decision-making aid with the function of assisting women with MS in making their decision of whether to become a parent.

Researchers at the University of Sydney are interested in how the use of a decision aid will provide information that increases knowledge about pregnancy in MS as well as facilitating decision-making in women with MS who are in the process of deciding whether or not to have a child. Research suggests that women with MS are often faced with decisional conflict regarding the motherhood decision as the already physically and emotionally demanding time of pregnancy and childbearing may be exacerbated by a potentially disabling illness. Women with MS considering pregnancy and childrearing are faced with an exaggerated experience of the already stressful concerns associated with bringing a child into the world. However at this time little is known about how such distress and conflict may be alleviated with the provision of an aid to assist woman in engaging in the decision making process.

If you agree to take part in this study, you will be asked to attend a one hour focus group at the MS Society in order to give feedback on the decision aid and provide some information on your experiences. We are interested in the views of women who have already contemplated this decision, regardless of whether they chose to have children or not, and of women who are currently making this decision.. We have developed a decision aid to help women currently making this decision and will ask you to read through the decision aid and complete a brief questionnaire giving us your opinion about it, which will take approximately five minutes. We will also ask you some questions in the group about your own experiences of this decision and ask you to share any other information that you think might be helpful to women in this situation. All information collected during this study is strictly confidential, except as required by law. The information will not be stored with your name, but will be stored against a number. Only the investigators for this project will have access to this information.

We hope that you will agree to take part in this study, but you do not have to. Your participation is completely voluntary. If you do agree to take part, but later

change your mind you may withdraw at any time. Whatever you decide about the study, your decision will not in any way affect you or your future treatment at the MS Society, which will continue exactly as before. We will provide you with a copy of this information sheet if you agree to take part.

If you have any questions about the study, please do not hesitate to ask Ms. Martine Sponiar, the intern clinical psychologist collecting information from participants. She can be contacted on (02) 9351 5952. For further information, you can contact Dr. Louise Sharpe on (02) 9351 4558 who is the chief investigator for this project.



Department of Psychology
The University of Sydney, N.S.W. 2006

CONSENT FORM

“The Motherhood Decision: Developing a Decision Aid for Women with MS”

I,, have been told about this study and have been allowed the opportunity to ask questions about it. I understand what is involved in taking part and I agree to do so.

Signed:

Date: / /

Name:

Witness:

Date: / /

Name:

<p>If you have any concerns or complaints about your involvement in the study, please contact the Manager of the Human Research Ethics Committee, University of Sydney on (02) 9351 4811.</p>
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Department of Psychology
The University of Sydney, N.S.W. 2006

PARTICIPANT INFORMATION SHEET

“The Motherhood Decision: Designing a Decision Aid for Women with MS”

This information sheet has been prepared to provide you with information about the study you have been asked to take part in. This study aims to develop a decision-making aid with the function of assisting women with MS in making their decision of whether to become a parent.

Researchers at the University of Sydney are interested in how the use of a decision aid will provide information that increases knowledge about pregnancy in MS as well as facilitating decision-making in women with MS who are in the process of deciding whether or not to have a child. Research suggests that women with MS are often faced with decisional conflict regarding the motherhood decision as the already physically and emotionally demanding time of pregnancy and childbearing may be exacerbated by a potentially disabling illness. Women with MS considering pregnancy and childrearing are faced with an exaggerated experience of the already stressful concerns associated with bringing a child into the world. However at this time little is known about how such distress and conflict may be alleviated with the provision of an aid to assist woman in engaging in the decision making process.

If you agree to take part in this study, you will be asked to fill out a number of questionnaires related to your own feelings and beliefs, which will take approximately forty minutes. You will be asked to complete these same questionnaires again after one month and twelve months. With your permission, the researchers will contact your treating neurologist or GP to confirm the type of MS that you have. We will also ask you whether you would be happy for the researchers to contact you in the future to see whether you did start a family or not.

After you have completed the questionnaires, you will be randomly assigned (like the toss of a coin) to one of two groups. One group will be given a copy of the Motherhood Decision Aid to read through. If you are allocated to this group, you will receive a telephone call from Martine Sponiar to talk through that information and clarify anything that is unclear. This telephone conversation would take no more than half an hour. The other group will not receive the decision aid, but will be asked to complete the questionnaires. All information collected during this study is confidential, except as is required by law. The information will not be

stored with your name, but will be stored against a number. Only the investigators for this project will have access to this information.

We hope that you will agree to take part in this study, but you do not have to. Your participation is completely voluntary. If you do agree to take part, but later change your mind you may withdraw at any time. Whatever you decide about the study, your decision will not in any way affect you or your future treatment at the MS Society, which will continue exactly as before. We will provide you with a copy of this information sheet if you agree to take part.

If you have any questions about the study, please do not hesitate to ask Ms. Martine Sponiar, the intern clinical psychologist collecting information from participants. She can be contacted on (02) 9351 5952. For further information, you can contact Dr. Louise Sharpe on (02) 9351 4558 who is the chief investigator for this project.

If you have any concerns or complaints about your involvement in the study, please contact the Manager of the Human Research Ethics Committee, University of Sydney on (02) 9351 4811.



Department of Psychology
The University of Sydney, N.S.W. 2006

CONSENT FORM

“The Motherhood Decision: Developing a Decision Aid for Women with MS”

I,, have been told about this study and have been allowed the opportunity to ask questions about it. I understand what is involved in taking part and I agree to do so.

Signed:

Date: / /

Name:

Witness:

Date: / /

Name:

I give permission for the researchers to contact my treating physician to confirm my MS-type and medications prescribed for its management.

Signed:

Date: / /

Name:

Witness:

Date: / /

Name:

Name of Physician:

Address:

.....

.....

Phone:

I would be happy for the researchers to contact me after the end of the study to find out
whether or not I decided to start a family: YES / NO

APPENDIX B:
RECRUITMENT MAIL-OUT



Department of Psychology
The University of Sydney, N.S.W. 2006

Motherhood Choice: Developing a Decision Aid for Women with MS

The University of Sydney, in conjunction with MS Society, is recruiting women into a study to evaluate a decision aid to help women with MS make decisions about whether to become a parent

We have mailed all women, who have agreed to take part in research, and are between the ages of 20-40 to invite them to take part.

Even if you would prefer not to take part in the study, we would be really interested to know which of these situations best describes your current situation with regard to your decision about motherhood. If you would be happy to provide this information (either anonymously or with your name), simply tick the statement below that best describes your current situation, answer the few questions below and return this sheet in the reply-paid envelope.

- ☐ I am currently unsure about whether I want to start a family but am considering these issues at the moment.
- ☐ I am currently unsure about whether I want to start a family at some time in the future, but am not ready to think about that now.
- ☐ I already have a child (or children), but am not sure whether I will have another child and am thinking about this at the moment.
- ☐ I already have a child (or children), but am not sure whether I will have another child, but am not ready to think about that now.
- ☐ I already have children and do not want to have any more.
- ☐ I do not have children and have decided not to have any.

Age: _____
How long have you had MS: _____
Type of MS (if known): _____
Age of any children: _____

If you are interested in participating in this study to evaluate a decision aid for the motherhood decision for women with MS, please complete your details below and we will contact you.

Name: _____
Address: _____

Phone No: _____
Mobile: _____
Email (if available): _____

APPENDIX C:
STATISTICAL OUTPUT CHAPTER 2

**APPENDIX D: QUESTIONNAIRE & FEEDBACK ON
DECISION AID FROM WOMEN IN FOCUS
GROUPS**

Acceptability Questionnaire

My thoughts on the decision-aid for the motherhood choice after an MS diagnosis

We would like to know what you think about the decision-aid you have just reviewed.

1. Please rate each section, by circling 'poor', 'fair', 'good', or 'excellent' to show what you think about the way the information was presented on:

Impact of MS	Poor	Fair	Good	Excellent
Medication	Poor	Fair	Good	Excellent
Types of MS	Poor	Fair	Good	Excellent
Pregnancy	Poor	Fair	Good	Excellent
Postnatal period	Poor	Fair	Good	Excellent
Breastfeeding	Poor	Fair	Good	Excellent
Disability	Poor	Fair	Good	Excellent
Concerns of women with MS	Poor	Fair	Good	Excellent
Stories about other women	Poor	Fair	Good	Excellent
Values clarification	Poor	Fair	Good	Excellent
Decision making steps	Poor	Fair	Good	Excellent

2. The length of the booklet was (*tick one*)

- ☐ Too short
- ☐ Too long
- ☐ Just right

3. The amount of information was (*tick one*)

- ☐ Too much information
- ☐ Too little information
- ☐ Just right

4. I found the information

- ☐ Slanted towards having children
 - ☐ Slanted towards not having children
 - ☐ Balanced
5. Would you have found this decision aid useful when you were making the motherhood choice?
- ☐ Yes
 - ☐ No

Comments:

6. Did you find the values-clarification exercise made options:
- ☐ Easier to contemplate
 - ☐ More difficult to contemplate

Comments:

7. Did the decision-making steps make your decision:
- ☐ Easier
 - ☐ More difficult

Comments:

8. Do you think we included enough information to help a woman make this decision?
- ☐ Yes
 - ☐ No

Comments:

9. What did you like about the decision aid and worksheet?

10. What suggestions do you have to improve the decision aid or worksheet?

Table 7.1

Mean ratings of the way the information was presented in the decision aid

Item	Mean	Descriptor
Impact of MS	3.1	Good
Medication	3.25	Good
Types of MS	3.15	Good
Pregnancy	3.1	Good
Postnatal	3.2	Good
Breastfeeding	3.3	Good
Disability	3.1	Good
Concerns	3.2	Good
Values	2.6	Good
Decision steps	2.7	Good
Length	1.7	Just right
Amount	2.25	Just right
Slant	2.05	Balanced
Useful	1	Yes
Values	1.55	Easier → unchanged
Decision steps	1.45	Easier → unchanged
Enough information	1.25	Yes

APPENDIX E:
DECISION AID

APPENDIX F:
MEASURES USED IN RANDOMISED
CONTROLLED TRIAL

ID #

Demographic Questionnaire

1. Age:
2. How old were you when you were diagnosed with MS?
3. What type (course) of MS have you been diagnosed with?
 - ☐ Relapse-remitting
 - ☐ Secondary-progressive
 - ☐ Primary-progressive
 - ☐ Progressive-relapsing
 - ☐ Benign
 - ☐ Other
4. Please list any medication you are currently taking to manage your MS:

5. Do you already have children?
 - ☐ Yes, 1 child
 - ☐ Yes, more than 1 child
 - ☐ No
6. Before you were diagnosed with MS, did you want to have children?
 - ☐ Yes
 - ☐ No
 - ☐ Unsure
7. Did your MS diagnosis complicate your previous decision of whether or not to have children?
 - ☐ Yes
 - ☐ No
8. When was your last relapse/exacerbation?

9. Please circle where you see yourself on the following continuum:

-5	-4	-3	-2	-1	0	1	2	3	4	5
Definitely will not have children now or in the future					Unsure					Definitely will have children now or in the future

KNOWLEDGE QUESTIONNAIRE

1. The majority of studies on pregnancy in MS have been carried out on women with which course of MS:
 - ☐ Benign MS
 - ☐ Relapse-Remitting MS
 - ☐ Secondary-Progressive MS
 - ☐ Primary-Progressive MS
 - ☐ Progressive-Relapsing MS
2. The risk of passing on MS when one of the parents has it is:
 - ☐ 20%
 - ☐ 0.1%
 - ☐ 50%
 - ☐ 3-5%
 - ☐ 10-12%
3. When trying to become pregnant, how early should a woman stop taking their Immunotherapy medication?
 - ☐ 1 month before trying to conceive
 - ☐ 3 months before trying to conceive
 - ☐ 2 weeks before trying to conceive
 - ☐ Once pregnancy is confirmed
 - ☐ After the first trimester
4. Which childbirth aesthetic procedure is not advised for women with MS?
 - ☐ Spinal anaesthesia
 - ☐ General anaesthesia
 - ☐ Epidural
 - ☐ All of the above
 - ☐ None. All are considered safe.
5. Which of the following statements is true?
 - ☐ Pregnancy accelerates a woman's course of MS
 - ☐ There is a higher risk of stillbirth in women with MS
 - ☐ Pregnancy does not affect the course of MS
 - ☐ MS symptoms remain unchanged during the pregnancy period
 - ☐ There is a lower risk of stillbirth in women with MS
6. Which are the most reliable predictors of relapse during the postpartum period?

- ☐ A higher number of previous pregnancies, increase disability at pregnancy onset & an increased relapse rate during pregnancy
- ☐ An earlier age at MS onset, lower number of previous pregnancies & an increased relapse rate during pregnancy
- ☐ Breastfeeding, a progressive-relapsing course of MS & a decreased relapse during pregnancy
- ☐ A progressive-relapsing course of MS, higher number of previous pregnancies & a later age at MS onset
- ☐ An increased relapse rate in the pre-pregnancy year, an increase relapse rate during pregnancy & increased disability at pregnancy onset

7. Which of the following statements is true?

- ☐ Breastfeeding is not recommended for women with MS
- ☐ Relapse rates generally decrease during pregnancy
- ☐ Becoming pregnancy increases relapse rates in the long term
- ☐ MS can be passed on to a baby through breast milk
- ☐ Managing relapse rates with medication is vital during pregnancy

8. Which of the following statements is *not* true?

- ☐ MS can result in sexual dysfunction
- ☐ There is an 80% risk of increased relapses in the postpartum period
- ☐ Fatigue is worse in the postpartum months
- ☐ MS medication can reduce the effectiveness of oral contraceptives
- ☐ Parenting may be impractical for some women with MS

9. How might engaging in physical affection with your child be affected by your MS?

- ☐ Fatigue
- ☐ Sensory problems
- ☐ Motor problems
- ☐ A & C only
- ☐ All of the above

10. Women with MS commonly express concern about not being able to perform tasks to the social standard expected of a mother. These worries can become worse when a mother perceives social doubt of her ability to parent her children, and can increase the risk of depression. What factor significantly mediates the negative effects of these concerns?

- ☐ Less obvious disability
- ☐ A longer period of time since diagnosis, thus there is a higher chance that the woman will have come to terms with her illness.
- ☐ Being on effective medication
- ☐ Social support
- ☐ Having a well behaved child

Decisional Conflict

My difficulty making this choice

Please look at the following comments made by people when making decisions.

Please show how strongly you agree or disagree with these statements by circling the number from 1 (strongly agree) to 5 (strongly disagree), which best shows how you feel about the choice you will be making.

- | | | | | | |
|--|---------------------|------------|---------------------------------|---------------|------------------------|
| 1. This decision is easy for me to make | 1
Strongly Agree | 2
Agree | 3
Neither Agree Nor Disagree | 4
Disagree | 5
Strongly Disagree |
| 2. I am sure what to do in this decision | 1
Strongly Agree | 2
Agree | 3
Neither Agree Nor Disagree | 4
Disagree | 5
Strongly Disagree |
| 3. It is clear what choice is best for me | 1
Strongly Agree | 2
Agree | 3
Neither Agree Nor Disagree | 4
Disagree | 5
Strongly Disagree |
| 4. I am aware of the options I have in this decision | 1
Strongly Agree | 2
Agree | 3
Neither Agree Nor Disagree | 4
Disagree | 5
Strongly Disagree |
| 5. I feel I know pros of each option | 1
Strongly Agree | 2
Agree | 3
Neither Agree Nor Disagree | 4
Disagree | 5
Strongly Disagree |
| 6. I feel I know cons of each option | 1
Strongly Agree | 2
Agree | 3
Neither Agree Nor Disagree | 4
Disagree | 5
Strongly Disagree |
| 7. I know <u>how important</u> the pros are to me in this decision | 1
Strongly Agree | 2
Agree | 3
Neither Agree Nor Disagree | 4
Disagree | 5
Strongly Disagree |
| 8. I know <u>how important</u> the cons are to me in this | 1
Strongly | 2
Agree | 3
Neither | 4
Disagree | 5
Strongly |

decision	Agree		Agree Nor Disagree		Disagree
	1	2	3	4	5
9. I know which is more important to me (the pros or the cons)	Strongly Agree	Agree	Neither Agree Nor Disagree	Disagree	Strongly Disagree
10. I am making this choice without any pressure from others	Strongly Agree	Agree	Neither Agree Nor Disagree	Disagree	Strongly Disagree
11. I have the right amount of support from others in making this choice	Strongly Agree	Agree	Neither Agree Nor Disagree	Disagree	Strongly Disagree
12. I have enough <u>advice</u> about the options	Strongly Agree	Agree	Neither Agree Nor Disagree	Disagree	Strongly Disagree
13. I feel I will make an informed choice	Strongly Agree	Agree	Neither Agree Nor Disagree	Disagree	Strongly Disagree
14. My decision will show what is important to me	Strongly Agree	Agree	Neither Agree Nor Disagree	Disagree	Strongly Disagree
15. I expect to stick with my decision (when I make it)	Strongly Agree	Agree	Neither Agree Nor Disagree	Disagree	Strongly Disagree
16. I think I will be satisfied with my decision (when I make it)	Strongly Agree	Agree	Neither Agree Nor Disagree	Disagree	Strongly Disagree

Decision Self-Efficacy

My confidence in making an informed choice

Below are listed some things involved in making an informed choice. Please show how confident you feel in doing these things by circling the number from 0 (not at all confident) to 4 (very confident) for each item listed below.

I feel **confident** that I can:

1. Get the facts about the medication choices available to me	Not at all confident	0	1	2	3	4	Very confident
2. Get the facts about the benefits of each choice	Not at all confident	0	1	2	3	4	Very confident
3. Get the facts about the risks and consequences of each choice	Not at all confident	0	1	2	3	4	Very confident
4. Understand the information enough to be able to make a choice	Not at all confident	0	1	2	3	4	Very confident
5. Ask questions without feeling dumb	Not at all confident	0	1	2	3	4	Very confident
6. Express my concerns about each choice	Not at all confident	0	1	2	3	4	Very confident
7. Ask for advice	Not at all confident	0	1	2	3	4	Very confident
8. Figure out the choice that best suits me	Not at all confident	0	1	2	3	4	Very confident
9. Handle unwanted pressure from others in making my choice	Not at all confident	0	1	2	3	4	Very confident
10. Let my partner/family know what is best for me	Not at all confident	0	1	2	3	4	Very confident
11. Delay my decision if I feel I need more time	Not at all confident	0	1	2	3	4	Very confident

CENTER FOR EPIDEMIOLOGIC STUDIES DEPRESSION SCALE (CESD)

Name: _____

Date: ____/____/____

(day/month/year)

CES-D *Instructions:* Below is a list of the ways you might have felt or behaved. Please tell me how often you have felt this way during the past week.

Responses: A. Rarely or none of the time (*less than 1 day*)
B. Some or a little of the time (*1-2 days*)
C. Occasionally or a moderate amount of time (*3-4 days*)
D. Most or all of the time (*5-7 days*)

	A	B	C	D
1. I was bothered by things that usually don't bother me.	ف	ف	ف	ف
2. I did not feel like eating, my appetite was poor.	ف	ف	ف	ف
3. I felt that I could not shake off the blues even with the help of my family or friends.	ف	ف	ف	ف
4. I felt that I was just as good as other people.	ف	ف	ف	ف
5. I had trouble keeping my mind on what I was doing.	ف	ف	ف	ف
6. I felt depressed.	ف	ف	ف	ف
7. I felt that everything I did was an effort.	ف	ف	ف	ف
8. I felt hopeful about the future.	ف	ف	ف	ف
9. I thought my life had been a failure.	ف	ف	ف	ف
10. I felt fearful.	ف	ف	ف	ف
11. My sleep was restless.	ف	ف	ف	ف
12. I was happy.	ف	ف	ف	ف
13. I talked less than usual.	ف	ف	ف	ف
14. I felt lonely.	ف	ف	ف	ف
15. People were unfriendly.	ف	ف	ف	ف
16. I enjoyed life.	ف	ف	ف	ف
17. I had crying spells.	ف	ف	ف	ف
18. I felt sad.	ف	ف	ف	ف
19. I felt that people dislike me.	ف	ف	ف	ف
20. I could not "get going".	ف	ف	ف	ف

STATE-TRAIT ANXIETY INVENTORY FOR ADULTS

SELF-EVALUATION QUESTIONNAIRE - Part 1

Please read each statement below and then place a cross over the appropriate number to the right of the statement to indicate how you feel ***right now***, that is, ***at this very moment***. There are no right or wrong answers. Do not spend too much time on any one statement but give the answer that seems to describe your feelings best.

	Not at all	Some- what	Moder- ately so	Very much so
1. I feel calm	1	2	3	4
2. I feel secure	1	2	3	4
3. I am tense	1	2	3	4
4. I feel strained	1	2	3	4
5. I feel at ease	1	2	3	4
6. I feel upset	1	2	3	4
7. I am presently worrying over possible misfortunes	1	2	3	4
8. I feel satisfied	1	2	3	4
9. I feel frightened	1	2	3	4
10. I feel comfortable	1	2	3	4
11. I feel self-confident	1	2	3	4
12. I feel nervous	1	2	3	4
13. I am jittery	1	2	3	4
14. I feel indecisive	1	2	3	4
15. I am relaxed	1	2	3	4
16. I feel content	1	2	3	4
17. I am worried	1	2	3	4
18. I feel confused	1	2	3	4
19. I feel steady	1	2	3	4
20. I feel pleasant	1	2	3	4

ADDPENDIX G:
STATISTICAL ANALYSES FOR RANDOMISED
CONTROLLED TRIAL

Table 7.2

T-test baseline differences pre-intervention

	t	Df	Sig.
Age	-1.093	149	0.276
Depression	-0.088	147	0.930
Anxiety	-0.194	147	0.846
Self-Efficacy	1.809	149	0.072
Decisional Conflict	-1.684	148	0.094
Knowledge	0.380	148	0.704
Certainty	0.895	148	0.372
MS Type	-0.677	149	0.498
Whether subjects wanted kids before their diagnosis	-0.604	149	0.546
Whether diagnosis complicated decision of children	-1.998	149	0.046

Table 7.3

Correlations of outcome measures

	Balance	Depression	Anxiety	Self- Efficacy	Decisional Conflict	Knowledge	Certainty
Balance	-	-0.128	-0.062	0.069	-0.152	0.063	0.454**
Depression		-	0.658**	-0.299**	0.127	-0.245**	-0.067
Anxiety			-	-0.283**	0.250**	-0.256**	-0.159
Self- Efficacy				-	-0.278**	0.012	0.038
Decisional Conflict					-	-0.007	-0.352**
Knowledge						-	0.103
Certainty							-

** Correlation is significant at the 0.01 level (2-tailed)

* Correlation is significant at the 0.05 level (2-tailed)

Table 7.4

ANOVA Table

Measure	Source	df	Mean Square	F	sig	Partial Eta Squared
Certainty	Time	135	10.135	8.739	0.004	0.061
	Group	135	2.859	0.493	0.484	0.004
	Time x group	135	3.310	2.854	0.047	0.021
Balance	Time	135	2.496	0.863	0.355	0.006
	Group	135	41.167	2.513	0.115	0.018
	Time x group	135	0.228	0.079	0.779	0.001
Self-Efficacy	Time	135	295.580	2.486	0.117	0.018
	Group	135	2.605	0.006	0.936	0.001
	Time x group	135	995.818	8.375	0.002	0.058
Decisional Conflict	Time	133	1.902	11.997	0.001	0.083
	Group	133	0.692	1.211	0.273	0.009
	Time x group	133	1.715	10.820	0.001	0.075
Knowledge	Time	133	19.402	8.215	0.005	0.058
	Group	133	35.884	6.774	0.010	0.048
	Time x group	133	46.961	19.883	0.001	0.130
Depression	Time	133	81.654	1.744	0.189	0.013
	Group	133	0.566	0.003	0.954	0.001
	Time x group	133	0.052	0.001	0.973	0.001
Anxiety	Time	133	17.477	0.291	0.590	0.002
	Group	133	4.049	0.019	0.890	0.001
	Time x group	133	59.988	0.001	0.991	0.001

Table 7.5

Chi-square table of clinical significance

Measure	Chi-Square	Df	sig.
Self-Efficacy	4.475	2	0.107
Decisional Conflict	6.467	2	0.018
Pre-intervention Knowledge	0.123	2	0.940
Post-intervention Knowledge	11.147	1	0.001

Table 7.6

Clinical significance cross-tabulation

Measure	Clinical significance			Total
	Worse	Same	Better	
Self-Efficacy				
Control	4	53	4	61
DA Intervention	1	66	11	78
Decisional Conflict				
Control	4	52	4	60
DA Intervention	0	68	10	78
	Below 7/10	7/10 or Above		Total
Pre-Intervention				
Knowledge				
Control	61	6		67
DA Intervention	75	8		83
Post-Intervention				
Knowledge				
Control	48	12		60
DA Intervention	41	37		78

APPENDIX H:
STATISTICAL ANALYSES FOR FOLLOW-UP

Table 7.7

ANOVA table comparing those who completed the study with those who did not

Variable		Df	Mean square	F	Sig
Group	Between groups	1	0.144	0.575	0.449
	Within groups	192	0.250		
Age	Between groups	1	5.012	0.297	0.586
	Within groups	159	16.860		
Time since Diagnosis	Between groups	1	33.080	2.279	0.133
	Within groups	150	14.514		
MS Type	Between groups	1	1.898	3.025	0.084
	Within groups	150	0.627		
Whether they wanted kids before their diagnosis					
	Between groups	1	0.070	0.248	0.619
	Within groups	150	0.283		
Whether diagnosis complicated their motherhood decision					
	Between groups	1	0.229	1.090	0.298
	Within groups	149	0.210		

Table 7.8

ANOVA table

Measure	Source	Df	Mean square	F	Sig.	Partial Eta squared
Certainty	Time	1	21.604	6.464	0.013	0.060
	Group	1	2.532	1.041	0.310	0.010
	Time x group	102	2.844	0.851	0.358	0.008
Balance	Time	1	43.270	4.101	0.045	0.039
	Group	1	12.984	1.483	0.226	0.014
	Time x group	102	0.091	0.009	0.926	0.001

APPENDIX I:
PUBLISHED PAPERS