An Exploration of the Information and Decision Support Needs of People with Multiple Sclerosis

Abigail Jane Eccles

Green Templeton College

Nuffield Department of Primary Care Health Sciences

Medical Sciences Division

University of Oxford

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I. Statement of contribution

I, Abigail Jane Eccles, hereby declare this thesis is my own work. I confirm that when others' work is referred to, this is referenced and acknowledged appropriately.

II. Acknowledgements

Firstly, I would like to thank all the participants with MS who gave up their time, welcomed me into their homes, and shared experiences with me so candidly during interviews. Thank you also to the consultant neurologists, not only for taking part in the interviews, but also for showing engagement and interest in the topic. And thank you to all the members of the patient and public involvement group, who provided very useful advice throughout all stages of this research.

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III. Thesis abstract

Recent decades have seen increasing recognition of the importance of patient involvement during patient-professional interactions and promotion of preventative and long term approaches to healthcare for those with long-term conditions. The concepts of 'shared decision making' and ‘personalised care planning’ have both been advocated by patient groups, policy-makers, professional bodies and academia as best practice. During shared decision making, patients and healthcare professionals work in equal partnership to decide the best course of action. Shared decision making is a central tenet of personalised care planning, as it aims to foster partnerships between patients and healthcare professionals when making decisions, but personalised care planning also describes an overall approach to healthcare that is forward-planning and preventative, rather than episodic and reactive. Despite the breadth of support for such approaches, in reality they are not routinely adopted. Multiple Sclerosis (MS) is a heterogeneous neurodegenerative long term condition, which is unpredictable with limited treatments available. Such uncertainty and complexity position MS as an interesting long term condition to explore decisional and information needs.

This doctoral research comprises of three methods stages. Firstly, two systematic reviews assessing the effectiveness of personalised care planning for people with long-term conditions and people with MS were carried out. Secondly, 22 in-depth semi-structured qualitative interviews were carried out with people with MS across the UK to explore experiences of decision making and interactions with healthcare professionals. Purposive sampling was carried out and data saturation determined sample size. A modified grounded theory approach was used and thematic analysis of interview data was carried out. Lastly, a series of structured qualitative interviews were carried out with 6 consultant neurologists. This stage was iterative in that problematic areas

identified during analysis of interview data from stage 2 were presented to neurologists in infographic form to further examine issues raised. Framework analysis was carried out on neurologist interview data to examine their interpretations and potential solutions.

Although there appears to be some evidence demonstrating that personalised care planning is effective for people with long term conditions, such favourable effects were not demonstrated in the context of MS. Based on the findings from the systematic reviews it is unclear whether personalised care planning is effective for people with MS and there is a clear gap in the literature examining this. Findings from the interview stages suggest there are key areas which are lacking in terms of information and decisional support. Such areas included the type and amount of information around the time of diagnosis, support when choosing disease modifying drugs and discussions about approaches outside mainstream medicine. Findings from neurologist interview data corroborated those from MS interview data, but through examination of issues raised it also highlighted some of the complexities and challenges of involving patients and enacting shared decision making in reality.

This research identified key areas that require improvement for people with MS in terms of provision of the information and decisional support. Although in theory personalised care planning and shared decision making are positioned as best practice, in reality it is unclear whether they are effective or appropriate for people with MS. The way in which such approaches are enacted are complex and require careful consideration. Potential barriers and pitfalls identified within this study suggest a lack of clarity in how to respond to challenges and further investigation into how patient involvement is enacted is needed.

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VI. List of abbreviations

ABN	Association of British Neurologists
CAM	Complementary and alternative medicine
CASP	Critical Appraisal Skills Programme
CCSVI	Chronic cerebrospinal venous insufficiency
CIS	Clinically isolated syndrome
CNS	Central nervous system
CNS	Central nervous system
CPPF	Canadian Practice Process Framework
DIP	Disability and impact profile
DMDs	Disease modifying drugs
EBM	Evidence based medicine
ECGP	Evaluation of client-centred process “ECGP” (sic)
IPA	Impact on participation and autonomy
ITT	Intention to treat
LDN	Low Dose Naltrexone
LTC	Long term condition
LTNC	Long term neurological condition
MND	Motor neurone disease
MRI	Magnetic resonance imaging

MS	Multiple Sclerosis
NPT	Normalisation Process Theory
NRS	Non-randomised studies
OT	Occupational therapist
PAL	Personal Activity Logs
PCP	Personalised care planning
PML	Progressive multifocal leukoencephalopathy
PP	Per protocol
PPI	Patient and public involvement
PPMS	Primary Progressive Multiple Sclerosis
RCT	Randomised controlled trails
RRMS	Relapsing remitting Multiple Sclerosis
SDM	Shared decision making
SPMS	Secondary Progressive Multiple Sclerosis
WHO	World Health Organisation

Section one
Introduction and Methods

Chapter 1

Patient Involvement, Shared Decision Making and Personalised Care Planning: Changing Trends in Current Healthcare Systems

1.1. Chapter Summary

Recent decades have seen increasing recognition of the value of patients' involvement in their own healthcare, as well as planned approaches to prevent ill-health and promote wellbeing as opposed to services that react to ill-health. Shared decision making (SDM) and personalised care planning have been proposed as ways to involve patients in collaboration with their healthcare professionals (SDM) in an approach that is preventative and forward-planning. This chapter outlines previous thinking around patient-doctor relationships and how this has changed with increasing recognition of the value of patient involvement. It summarises current support for SDM and personalised care planning from official bodies and academia, and how they have been reflected in government policies. Lastly, this chapter explores the barriers to implementing such approaches, despite overwhelming support from official perspectives.

1.2. Rise of patient involvement

1.2.1. Conceptualisations of the Patient-Doctor Relationship

Reflected in the Latin origins of the terms, *docēre (to teach)* and *pati (to suffer),* doctors and patients historically have held – and still do hold – unequal positions of power. Even before a consultation has begun, doctors (or healthcare professionals) are positioned as more knowledgeable, powerful and privileged than the average person (or patient). This inequality is often reinforced by education, income (Charles et al., 2004)

and simply because patients often require access to healthcare, and healthcare professionals act as gatekeepers to this (Lupton, 2004). Today's Oxford English Dictionary definitions also allude to unequal power: 'doctor' is defined as "someone who is qualified in medicine and treats people who are ill" and 'patient' is defined as a "person receiving or registered to receive medical treatment." These suggest an active doctor *treating* and passive patients *receiving* treatment. Although contemporary definitions, they do not encapsulate more current thinking, which recognises patients may have more active roles and the importance of their input – positioning patients and doctors more equally, than perceived historically.

One of the earliest and most cited theorists to write about doctor and patient roles was Talcott Parsons (1951). In the 1950s, he conceptualised illness as a form of societal deviance inhibiting normal functioning within society. He described the 'sick role': patients who become ill are expected to, and given permission to, stop performing their normal roles and activities, be in need of care and seek medical advice from doctors. He said doctors are a source of expertise and objective knowledge and patients are expected to follow doctors' advice as prescribed. The doctor was positioned as authoritative and the patient as unquestioning of the medical profession. He considered such unequal roles as unproblematic and a functional part of societies. Later, Szasz and Hollender (1956) argued that Parson's conceptualisation was not always appropriate and that different illnesses and symptoms may shape clinical interactions . They defined and described three basic models of patient-doctor relationship as:-

"(a) Activity-passivity. The comatose patient is completely helpless. The physician must take over and do something to him. (b) Guidance-cooperation. The patient with an acute infectious process seeks help and is ready and willing to cooperate. He turns to the physician for guidance. (c) Mutual participation. The patient with a chronic disease is aided to help himself. The physician's own inner needs (and satisfactions) form a complementary series with those of the patient." (Szasz and Hollender, 1956, p.592)

Szasz and Hollender's conceptualisations did not contradict Parsons', but further scrutinised roles in terms of how they may change according to patients' circumstances. They also interpreted the relationship as functional.

Later, Ivan Illich (1975) challenged the functionality of unequal power positions and authoritative status of the medical profession. He argued power was gained through 'medicalisation', a process where phenomena become understood in medical terms and thus *controlled* by the medical gaze. He considered the unequal power dynamic between medical institutions and society as problematic and even harmful. Also challenging the functionalist interpretation of the patient-doctor relationship, Stewart and Roter (1989) expanded conceptualisations of patient-doctor relationships further and argued that in reality there might be conflict during the consultation. Pointing to the importance of power dynamics between patients and doctors, they described four basic styles of consultations according to levels of patient and doctor control:-

Table 1.1. Stewart and Roter's Spectrum of control in patient-clinician relationships

		Physician Control	
		Low	**High**
Patient Control	**Low**	Default	Paternalism
	High	Consumerist	Mutuality

(Stewart and Roter, 1989, p.21)

Box 1.1. Stewart and Roter's (1989) descriptions of patient-clinician relationships

1) Paternalism:

"If there are high physician and low patient control, the physician will dominate, making decisions in what he or she perceives to be the patient's best interest. These decisions entail access to information and services. The patient's job is to cooperate with medical advice, that is, to do what he or she is told. This is the traditional form of the doctor-patient relationship and is still most common."

2) Consumerist:

"In the reversed power relationship between doctor and patient, a patient's demands for information and technical services are accommodated by a cooperating physician. This type of relationship is consumerist in nature and redefines the consultation as a marketplace transaction […] When the patient's requests are within the parameters of good medical judgement – and for most consultations these parameters are quite wide – little conflict need emerge. In acceding to demands that are contrary to standard practice, however, the physician is acting outside of his professional expertise and training, skirting the mainstream of medicine and risking criticism from medical colleagues. On the other hand, the physician may lose the patient's goodwill and business"

3) Mutuality:

"The mutuality model stills stresses patient control, but is based on a more participatory alternative, […] Each participant brings strengths and resources to the relationship on a relatively even footing. Because the power in the relationship is balanced, decisions are the results of what is being called a meeting between experts (Tuckett, 1985). The patient's job in this model is to become part of a joint venture."

4) Default:

"In this case, a patient may drop out of care completely because of failed expectations, concluding that all doctors are incompetent, insensitive, or otherwise inadequate. Most often the physician will be unaware of the reasons for the loss of this patient; the patient will simply not come back." (1989, p.20-23)

Stewart and Roter's descriptions broadly encapsulate the spectrum of interaction styles and the first three (paternalism, consumerism and mutuality) are regularly referred to in the bioethics and medical decision making literature. Mutuality is referred to as 'shared decision making' in more recent literature.

1.2.2. Changing attitudes to patient involvement and evidence-based practice

Recent decades have seen a change in attitudes towards patient involvement during consultations. Moving away from traditional paternalistic styles of interaction, there has been increasing recognition that patients ought to be involved in their healthcare and decisions made. Not only was it argued that their input would lead to best outcomes, there was also recognition that patients had the *right* to be involved (Morgan, 2008, Tuckett, 1985, Edwards and Elwyn, 2009), driven by key factors such as the Women's Health Movement (Nichols, 2000), introduction of the Mental Health Act (UK Government, 1983) and activism from patient and disability groups demanding more control, better care and independence (Foot et al., 2014).

Additionally, the increasing prevalence of long term conditions (LTCs) has led to a change in healthcare in terms of space and time. Rather than discrete decisions about acute illness made in during a consultation, people with LTCs are frequently carrying out on-going daily negotiations between different internal perspectives, such as social and medical, (Audulv et al., 2010) as well as decisions which are considered over a long period of time (Holman, 2004). Decisions are often made outside the consultation setting, with patients holding decisional power on how they self-manage. People with LTCs gain unique knowledge about their own condition through experience and have personal preferences relevant to the best course of action and outcomes.

There have also been immense changes in information use with the advent of the internet. This not only enabled societies to become interconnected but also allowed the general public (or patients) access to an abundance of health and medical information which previously would have been – in comparison – vastly limited. Thus, the knowledge divide between patients and healthcare professionals has potential to be reduced (Horton, 2003), as patients not only have knowledge about their preferences and illness experiences but may also gain clinical understandings too. Additionally, choice increased as medicine and technology advanced and more viable treatment options became available (Coulter, 2011). Increasing recognition of the importance of patient input (in terms of ethics and outcomes), reduced the knowledge divide between patients and doctors and increased uncertainty about best course of action (due to increased choice), meaning that paternalistic styles of interaction are considered less appropriate for current healthcare consultations.

A new disciplinary field, evidence based medicine (EBM), was also gaining increasing support around the same time increased patient involvement was. In the early 1990s, the term evidence based medicine first appeared in medical journals (Evidence-Based Medicine Working Group, 1992) and since then there has been exponential growth in its use alongside dedicated journals and conferences (Sackett, 2000). EBM is defined as "the conscientious, explicit, and judicious use of current best evidence in making decisions about the care of individual patients" (Sackett et al., 1996, p.71). Clinical information was positioned as something which should be scrutinised and assessed for effectiveness and appropriateness, rather than accepted without critical appraisal (Charles et al., 2004).

1.2.3. Shared decision making

In response to arguments for increased patient involvement and EBM, the concept of shared decision making[1] (SDM) gained increasing attention during the 1990s. SDM as an approach aims to involve patients alongside healthcare professionals whilst ensuring that information used to make decisions is based on reliable evidence. It is defined as:-

> "a process in which clinicians and patients work together to clarify treatment, management or self-management support goals, sharing information about options and preferred outcomes with the aim of reaching mutual agreement on the best course of action." (Coulter and Collins, 2011, p.2)

Although, SDM and EBM evolved from separate disciplinary backgrounds, there is much overlap between them and unity in their principles. Both call for access to - and critical appraisal of – best evidence, and both recognise that patient preferences should factor into the decision making process. Expanding further than their initial definition of EBM, Sackett et al. (1996) also explained that EBM in practice should incorporate patients' perspectives:

> "The practice of evidence based medicine means integrating individual clinical expertise with the best available external clinical evidence from systematic research. By individual clinical expertise we mean the proficiency and judgment that individual clinicians acquire through clinical experience and clinical practice. Increased expertise is reflected in many ways, but especially in more effective and efficient diagnosis and in the more thoughtful identification and compassionate use of individual patients' predicaments, rights, and preferences in making clinical decisions about their care." (Sackett et al., 1996, p.71)

However, EBM (as a disciplinary field) has received criticism for focusing on critical appraisal and synthesis of evidence, whilst giving little attention to how to apply it alongside patient values (Hoffmann et al., 2014). SDM focuses more on the

[1] termed as 'mutuality' by Stewart and Roter (1989)

interactional style during healthcare consultations and how best evidence alongside patients' perspectives are incorporated when making health related decisions.

The key principles of SDM have been conceptualised as involving at least two participants; the patient and healthcare professional, and sometimes more (wider networks of patients' friends and family and healthcare colleagues). Both parties take steps to participate in the decision making process, information is exchanged and a treatment decision is made (no action may be a decision) to which both parties agree (Elwyn et al., 2000). Patients' expertise is in: their experience of illness; social circumstances; attitude to risk; values and preferences. Whereas, clinicians' expertise is in: diagnosis; disease aetiology; prognosis; treatment options; and outcome probabilities (Coulter and Collins, 2011, Morgan, 2008). Various justifications have been put forward advocating SDM, which are summarised in the points and table below. The SDM approach is argued to:-

1. Value and recognise the importance of patient input alongside clinicians'.
2. Consider a broad range of circumstances.
3. Enable patient access to evidence-based, unbiased, full information.
4. Be responsive and reflective to context.
5. Be applicable to most scenarios.

Box 1.2. A summary of arguments made for Shared Decision Making

1. **Value and recognise the importance of patients' input alongside healthcare professionals' input**

The logic of paternalism is based on an assumption that clinicians are best positioned to judge trade-offs and risks because of their expertise and experience (Charles et al., 2004). In reality, consultation settings, knowledge and decision making processes are often more complex. For example, misinterpretation of patients' preferences has been argued to be *as* damaging as clinical misdiagnosis, but such misinterpretation is more difficult to recognise and so is continually overlooked (Mulley et al., 2012). The SDM approach recognises the importance of patient input and positions this as equal to healthcare professionals'. In valuing patient input, whilst recognising the importance of healthcare professionals' responsibility, patients are less likely to feel burdened with decisions (as they might with the consumerism model).

2. **Consider a broad range of circumstances**

Discussions about interaction styles are often focused on the communication exchange within consultations. A SDM approach recognises that decision making is not an isolated event but is affected by – and takes account of – a broader range of external and temporal circumstances (Entwistle, 2009). The focus is not limited to discrete decision points and ownership of these; rather it is concerned with processes and interactions throughout the decision making process.

3. **Enable patient access to evidence-based, unbiased, full information**

The ways in which clinical evidence and outcome probabilities are presented are potentially open to manipulation, leading to biased and unrealistic assessments (Gigerenzer and Gray, 2011). Such manipulation may be used to persuade a patient to make (or agree to) a particular choice (Elwyn et al., 1999). Despite healthcare professionals' best intentions, this would be considered unethical by most bioethicists because it prioritises healthcare professionals' opinions over patients' (Holm and Davies, 2009). SDM ideals are that information provided by healthcare professionals is "reliable, balanced, evidence-based" in a manner that is clear and understandable (Coulter and Collins, 2011, p.2).

4. Be responsive to, and reflective of, context

During SDM, ideally patients and healthcare professionals are both engaged and work as "active partners" during consultations (Coulter and Ellins, 2009, p.31). However, in reality, this level of involvement may conflict with patients' preferred level of involvement. How a person will react or respond to illness and the options available, is not known until they actually experience it. Thus, realistic assessments of preferred levels of involvement and information beforehand may be impossible (Elwyn et al., 2000, Lupton, 2004). Generally, patients' preferences for levels of involvement and information have been shown to vary according to demographic factors and health status. Furthermore, they are likely to change for each person throughout time and illness trajectory (Coulter and Ellins, 2009). Within the SDM model, healthcare professionals can be sensitive and responsive to the patients' preferred roles, levels of involvement and information needs. Also, where practical tailor communication and information accordingly.

5. Be applicable to most scenarios

Health related decisions are diverse and expansive. It has been highlighted that certain circumstances are more appropriate for SDM and patient involvement (Edwards and Elwyn, 2009). Contexts where biomedical knowledge is limited and patients' values and experiences affect the best outcome are frequently cited as most suitable for SDM. These include: "major health care decisions where there is more than one feasible option"; "screening tests and preventative strategies"; and "self-management for people with long-term conditions" (Coulter et al., 2011, p.2). As medical technologies advance, feasible options are ever-increasing and rates of chronic illness are increasing, the SDM approach is becoming ever more relevant (Coulter, 2011). Scenarios where patients absolutely cannot be involved in decision making (i.e. emergency scenarios) are less common, thus SDM is arguably a suitable approach to most consultations (Gulland, 2011).

1.2.4. Personalised care planning

As well as arguments made for SDM, it has also been suggested that planned, preventative and proactive approaches to healthcare (rather than healthcare that is unplanned, reactive and episodic) are most suitable. Increasing level of LTCs has led to increased use of healthcare services. On a global level, the management and treatment of LTCs has been identified as the greatest challenge to current healthcare systems (World Health Organization, 2013). In order to overcome such strains, it is argued that patients need to become more engaged in self-care and ill health ought to be prevented, rather than reacted to (UN Secretary General, 2011). In the UK specifically, the

Wanless Report (2002) identified the need for preventative approaches, engaged patients and partnerships with healthcare professionals in order to overcome economic strains.

Throughout recent decades there have been efforts to empower and encourage patients to become involved in care planning and self-management in a preventative rather than reactive manner. One of the most cited examples is Wagner's (1998) Chronic Care Model. This community-based model positions policies and resources as supportive of self-management for people with LTCs. Complementing SDM ideals, the model promotes care that is consistent with research evidence, incorporates patient preferences and prepares and empowers people to be involved in, and to manage, their health and healthcare (Wagner, 1998).

Taking this chronic care model forward, in 2012 the Department of Health advised that all patients with LTCs should have a personalised care plan (Department of Health, 2012). NHS England (2017b) describe personalised care planning as follows:-

> "This [personalised] care planning process changes the focus of how patients are cared for by looking at their current and future needs. This creates a more helpful proactive service – centred on the needs of each individual, rather than a service that comes into effect when something goes wrong. It helps to focus on what is important to the person and puts them on an equal footing with professionals. It goes beyond clinical treatment, taking a whole-life approach and includes things like peer and community support and support for self-management." (NHS England, 2017b)

This approach incorporates SDM, in that patients and healthcare professionals are expected to play equal roles and work together, but also expands further by incorporating a preventative and forward planning approach.

1.3. Policy and official support for Personalised Care Planning and Shared Decision Making

1.3.1. Developing support in recent decades

Developing support for both SDM and personalised care planning has also been reflected in UK policy documents and official guidance. In 1982, Margret Thatcher wrote about "extend[ing] patient choice" and "put[ting] the needs of patients first" in the foreword for the White Paper 'Working for Patients' (Department of Health, 1989). This White Paper proposed to be more responsive to local-level needs and provide increased choice to patients. A year later, the Mental Health Act (1983) legislated that patients must provide consent for treatment, except for specified circumstances. Patient involvement was also promoted by official bodies such as the General Medical Council. In 1995, their guide to Good Medical Practice stated that in order to maintain trust, doctors must "listen to patients and respect their views […] respect the right for patients to be fully involved in decisions about their care" (General Medical Council, 1995, p.4)

With a New Labour Government in 1997, a White Paper 'The new NHS. Modern Dependable' was published (Department of Health, 1997). There was emphasis on overcoming unequal access to healthcare across the UK and for primary care to coordinate services. In order to overcome variation, it aimed to provide national consistency based on clinical evidence and cost-effectiveness, yet remain responsive to individual patients. The paper also acknowledged patients' increasing access to information and predicted that this would result in "a powerful alliance between knowledgeable patients advised by knowledgeable professionals as a means of improving health and healthcare." (Department of Health, 1997, p.27). In 1998, the government introduced an annual national patient survey to gather information about

the performance of NHS services from patients' perspectives, which has continued to this day (Care Quality Commission, 2016)

In 1999, movements toward engaging patients in their own health behaviours were reflected in the White Paper, 'Saving Lives: Our healthier nation' (Department of Health, 1999) which emphasised individual patients' responsibility in maintaining good health through healthy behaviours, such as exercise, sensible diet and not smoking. It positioned the Government's role as providing evidence-based information on how to improve health and patients' as making informed decisions based on this. The White Paper also introduced NHS Direct as an initiative to provide health advice (via phone and internet) in order to empower members of the public in their own healthcare. In recognition of the knowledge people with LTCs have, the Expert Patients Programme was also proposed. In this programme, people with LTCs attended a patient-led programme and were supported in managing their own health (Department of Health, 1999). In 2000, the White Paper 'The NHS Plan' (Department of Health, 2000) continued to focus on self-care and preventative approaches and also proposed that over the next 10 years the NHS would be "redesigned to be patient centred – to offer a personalised service" (Department of Health, 2000, p.17). It recommended that patients should have access to their own health records, be copied into letters about their care and lay summaries of NICE guidelines would be initiated.

In 2002, the UK government-funded Wanless report (2002) examined factors which were likely to affect healthcare service provision in the long term. Patients' engagement in self-care, use of technology and preventative approaches were identified as crucial factors predicted to improve outcomes, such as the population's life expectancy and health status, and in overcoming healthcare system strains during the subsequent ten years. Wanless recommended that in order to achieve favourable outcomes, effective

partnerships between patients and healthcare professionals were needed. In 2009, The NHS Constitution was devised (Department of Health, 2009) and reemphasised the rights of patients to be involved in their healthcare. However, in addition to commitments to enabling patient involvement, providing evidence-based accessible information and providing decisional support, The NHS Constitution also outlined expectations that patients take responsibility for their own health status and well-being.

The UK government's White Paper, Equity and Excellence: Liberating the NHS (2010) cited the concept of SDM as an argument for consumerism and enabling patients to choose their healthcare provider. However, some have noted that the paper did not sufficiently engage in the concept of SDM, causing further confusion of (and potential resistance to) the concept (Coulter, 2012). Taking heed of recommendations for proactive and preventative care which engages patients in taking responsibility, personalised care planning was put forward as the best approach for those with LTCs. In 2012 policy makers mandated that *all* patients with LTCs should be offered a personalised care plan and be given the option to control their own personal health budgets (Department of Health, 2012).

1.3.2. Current continued support

After decades of development, SDM and personalised care planning as approaches continue to be advocated by official bodies. Content of the approaches have not changed, rather their imperatives have been reasserted in subsequent government documents (NHS England, 2014, NHS England, 2016, NHS England, 2017a). Focusing on SDM specifically, a 'Shared Decision Making Collaborative', made up of various high-profile government, quality standard, professional, academic, patient-led groups

and charities (listed in box 1.3) has formed to support the encouragement of SDM in UK healthcare systems (NICE, 2017).

Box 1.3. Members of the Shared Decision Making Collaborative (2017)

- Advancing Quality Alliance
- British Medical Journal
- Cardiff University
- Coalition for Collaborative Care
- Dartmouth College
- General Medical Council
- Health Education England
- The Health Foundation
- Imperial College, London
- Keele University
- King's College London
- National Voices
- Newcastle University
- NHS Choices
- NHS England
- National Institute for Health and Care Excellence
- Patient and public representation
- Patients Involved in NICE (PIN)
- Royal College of General Practitioners
- Royal College of Obstetricians and Gynaecologists
- Royal College of Physicians
- Royal College of Psychiatrists
- Royal College of Surgeons
- UCL Partners
- University of Leeds
- University of Liverpool
- University of Oxford

As a collaborative, they have endorsed a consensus statement that outlines recommendations to encourage, implement and embed SDM. Such a broad range of high profile organisations emphasises the weight of support behind SDM.

1.4. Barriers to Shared Decision Making and Personalised Care Planning

Despite broad consensuses and official support positioning SDM and personalised care planning as best practice, in reality these approaches do not seem to be commonplace. Many have argued that most healthcare professionals still do not involve patients in SDM processes during consultations or even incorporate their preferences (Couet et al., 2015, Elwyn et al., 2013, Lloyd et al., 2013). For example, only half of patients surveyed reported being involved in decisions to their preferred level (NHS England, 2014). There is also little evidence that personalised care planning is being carried out. Although most patients with LTCs in England do report having care planning discussions in some form, they rarely experience systematic, proactive personalised care planning support (Burt et al., 2012, Newbould et al., 2012) and many patients are completely unfamiliar with these methods of care (Newbould et al., 2012). There are various barriers that appear to work against the adoption of SDM and personalised care planning approaches.

1.4.1. Cultural inertia

In a context where healthcare interactions are traditionally paternalistic and services are designed to provide reactive and episodic responses to acute ailments, then enacting SDM and personalised care planning may be slow. As Foot el al (2014, p.7) put it:-

"[I]nvolvement is challenging. It challenges orthodoxies, vested interests and established ways of doing things. It requires professional communities to do things differently and to let go. It requires unwieldy administrative systems to become more responsive and human. It requires some patients and citizens themselves to think about their health and their care in different ways. In short, it poses a fundamental cultural and behavioural challenge to our systems of health and social care. Technocratic responses to this challenge – for example, the introduction of patient decision aids and feedback questionnaires – are not in themselves sufficient. There must also be a will to do things differently"

Thus it is argued that the normative position paternalism holds works against adoption of a new approach to interaction and decision making (Elwyn et al., 1999). A recent narrative review detailing patients' views as to why patients are reluctant to be involved, found patients often believe that healthcare professionals *do not want* them to be involved and those patients who adopt more involved roles would be considered "troublesome", "inflexible" or "difficult" and potentially cause the quality of their care to become compromised. There was also a view that to ask questions may "undermine the status of the professional" or "indicate distrust" (Joseph-Williams et al., 2014, p.13). Additionally, other reasons behind patients' reluctance in being involved detailed in the review focused on timing and resources. Some patients found that the timing of the decision (e.g. soon after diagnosis) could make it difficult to be involved. Awareness of healthcare professionals' heavy workloads and time pressures were also identified as a barrier to involvement, as patients assumed that increased involvement would imposition healthcare professionals further. There was also a perception that patients do not have the *right* to choice, when services are publically-funded (Joseph-Williams et al., 2014).

Traditional unequal power positions may mean patients assume a passive role by default. Adoption of more passive roles, means patients typically have less power to effectively shape and negotiate their roles and relationships with healthcare

professionals and many patients may be unaware of the possibility of increased involvement. Thus, healthcare professionals specifically would need to be the ones to instigate and encourage patient involvement (Stewart and Roter, 1989, Stiggelbout et al., 2012, Joseph-Williams et al., 2014). If healthcare professionals do not recognise the value of involving patients, it may not be executed effectively (Stiggelbout et al., 2012) and so in order to enable SDM, both patients and healthcare professionals need to recognise its value and be willing to act as partners (Stiggelbout et al., 2012, Coulter, 2011, Coulter and Ellins, 2009), which means patients need to feel supported and willing to adopt a more involved role.

Both Wagner's chronic care model (Wagner, 1998) and the 'House of Care' model (NHS England, 2015) highlight the importance of having organisational support when enacting a personalised care planning approach. A recent randomised controlled trial testing the effectiveness of a very comprehensive personalised care planning intervention found that the implementation was poor. In a subsequent evaluation, authors found that healthcare professionals did not consider the personalised care planning intervention a priority compared with other ways of carrying out care. This exemplifies how adoption of new approaches can be problematic if such approaches compromise other priorities or do not complement traditional approaches (Kennedy et al., 2014). In 2011, 'The Year of Care' programme piloted an intervention for diabetes based on Wagner's model; their report found that personalised care planning was effective, but its success was dependent on having organisational features set up to support this approach. These features included that all staff (from leadership to frontline) subscribed to personalised care planning and roles and responsibilities were clarified; a framework was used to guide consultations and relevant services were made available through commissioning and links with third sector organisations (Year of

Care, 2011). So, although personalised care planning is promoted by official bodies, without cultural change, health professionals' backing and organisational support, it seems likely to fail. Currently, UK healthcare systems are not set up accommodate personalised care planning. They have been described as being organised to provide reactive and acute care (NHS England, 2014) rather than the preventative long-term approaches personalised care planning demands. Some argue that separate healthcare teams have led to lack of coordinated care, as different teams have differing priorities and drivers (Eaton et al., 2015), most of which concern short-term financial gains, overlooking patients' priorities (Chew-Graham et al., 2013, Eaton et al., 2015). In such settings, personalised care planning may not be realistic until there is significant reorganisation of healthcare systems and cultural changes, alongside coherent strategies and incentives to prioritise it.

1.4.2. Potential for conflict

Ideally during SDM, patients (and/or caregivers) and healthcare professionals will consider clinical expertise, patient values and the best available evidence when making decisions (Stacey et al., 2009). However, the way in which these sources of information are prioritised can be a challenge and an ethical dilemma, especially when they come into conflict (Beauchamp and Childress, 2009, Stiggelbout et al., 2012, Holmes-Rovner and Rovner, 2009). Healthcare professionals' and patients' priorities can differ; during SDM healthcare professionals and patients may deliberate potential options together before reaching agreement. They do not have to both agree that the choice opted for is the *best* course of action, but they do have to agree that it is the action they will implement (Charles et al., 1997). However, although the deliberation process is often referred to, potential conflict and how such tensions are dealt with or overcome, appear

to be overlooked in much of the literature (Labrie and Schulz, 2014). Charles et al. (1999) address disagreement briefly, suggesting that when agreement is not reached, patients have to consult another healthcare professional if they wish to persue their preferred course of action (if healthcare professionals hold access to this). This points to how healthcare professionals often act as gatekeepers to treatment and services and therefore they *literally* do hold more power. How this factors into patients and healthcare professionals becoming equal partners as proposed by those theorising SDM (Coulter, 1999) appears to be unaddressed.

1.4.3. Confused and narrow conceptualisations

Patient-centred care has been conceptualised in different ways and motivated by multiple political positions. Various other concepts are related to, but are not the same as SDM and different descriptions of SDM have been used within the literature (Moumjid et al., 2007, Da Silva, 2012). Terminologies have been used interchangeably and authors are not always consistent and clear about which concept they are referring to, leading to confusion, ambiguities and misunderstandings of SDM (Moumjid et al., 2007).

The precise meaning of such concepts is often unclear for patients and healthcare professionals. For example some patients have been shown to mistake healthcare professionals' politeness for patient-centred care, (when in fact it may act to conceal lack of patient involvement) (Agledahl et al., 2011, Holmes-Rovner and Rovner, 2009). Additionally, the concepts of SDM and 'informed choice' are often confused and used interchangeably (Moumjid et al., 2007) but there is an important distinction to be made. Both promote patient autonomy and although SDM *involves* 'informed choice', it expands further than this to describe partnership between patients and healthcare

professionals. Whereas 'informed choice' *alone* can be interpreted as adoption of a consumerist approach, where healthcare professionals provide information and patients make decisions independently (Charles et al., 1999, Gafni et al., 1998, Charles et al., 1997, Entwistle, 2009). This confusion has led some to argue against SDM (Moumjid et al., 2007) due to concern that SDM (when confused with consumerism) relinquishes healthcare professionals' responsibilities, thus abandoning patients to deal with complex information and unwanted responsibility alone. But SDM theorists assert this is a misunderstanding, because whilst it is true that SDM ideals aim for patients to take more responsibility, this does not mean healthcare professionals take less. Responsibility is not passed from healthcare professionals to patients, but both take responsibility for the decision, resulting in an overall increase of responsibility (Holm and Davies, 2009).

SDM conceptualisations often focus only on exchange of information within consultations, the patients' understandings of information and the influence patients have over the option selected. Some have argued such focus is too narrow, as it overlooks the attitudes patients and healthcare professionals hold towards one another, their feelings when making decisions and their levels of engagement (Entwistle, 2009). Such oversimplified conceptualisations are also reflected in SDM intervention studies that tend to focus on information giving (rather than exchange) and do not explore patients' values and expectations with regards to potential outcomes (Stacey et al., 2009).

1.4.4. Practicalities and efficacy

SDM theorists argue it is a flexible and broad approach that responds to complexities within medical consultations. But this means enacting it is challenging and its simple

application is not appropriate. For example, although SDM aims to foster equal partnership, it also aims to accommodate preferred levels of involvement (in recognition that patients' preferred levels of involvement differ). Thus healthcare professionals need to be aware of their patient's preferred level of involvement in decision making, but the way this is assessed is an area of some contention. Some have argued that they should have explicit conversations about preferred levels of involvement (Towle and Godolphin, 1999) whereas others have suggested patients' desired levels can be instead be assessed intuitively from non-verbal signals (Elwyn et al., 2000). But using intuition may result in ambiguities, which go against SDM communication ideals of unbiased and clear communication. Additionally patients may not be aware of the option to be more actively involved and assume a passive role as default (Stewart and Roter, 1989).

It has been suggested that SDM is impractical as it requires longer consultation times due to increased communication during consultations (Holmes-Rovner and Rovner, 2009). In response others have suggested that increased consultation times do not necessarily mean the process is less efficient as potential SDM outcomes of less overtreatment and improved quality of care justifies this investment of time (Stiggelbout et al., 2012). Also, it is often assumed that implementation of SDM will encourage patients to make impractical and excessive demands for treatment that conflict with public health goals and spending (Coulter and Ellins, 2009, Holm and Davies, 2009). However, there is evidence suggesting that patients are in fact more likely to opt for more conservative treatment options when fully informed and involved in the decision making process (Stacey et al., 2014). This implies that SDM can prevent overtreatment rather than cause it.

The SDM model requires evidence to be communicated and reviewed during consultations and efforts to enact this have been made in recent years with the

introduction of decision aids. In order to tailor information to patients and practise SDM effectively, clinicians need access to reliable information quickly and to be proficient in evaluating and communicating all the legitimate options (Elwyn et al., 2000). But this may be unrealistic; for example, it has been estimated that 627.5 hours each month are required to keep up to date with current evidence in primary care (Alper et al., 2004) and healthcare professionals may not always be proficient in understanding the evidence (Gigerenzer and Gray, 2011). Therefore, access to the best evidence may not always be feasible in the reality of healthcare consultations. Furthermore, clinicians may lack sufficient training and confidence in explaining complicated information concerning risk and statistical inference to patients and avoid this process altogether (Holmes-Rovner and Rovner, 2009). Therefore, the role of the healthcare professional in communicating up-to-date evidence in a clear and unbiased manner may be unrealistic and expecting too much. Healthcare professionals are not always well trained in SDM and some report feeling ill equipped to take account of patient values (Elwyn et al., 2000). This suggests there may be gaps in medical training in communication skills and effective ways to support and empower patients to become more involved during decisions about their healthcare (Petrova et al., 2006, Stiggelbout et al., 2012).

There is a lack of consensus as to whether SDM is favourable in terms of efficacy, practicalities and efficiency. The very complexity it aims to address has resulted in SDM being a complex approach and its adoption challenging. Although existing evidence shows SDM is associated with positive affective and cognitive outcomes (e.g. knowledge and attitude), these are often measured via patient self-reports using scales which have not previously been validated, with less evidence demonstrating its effects on behavioural or physiological outcomes (Shay and Lafata, 2015).

1.5. Summary

In summary, increasing recognition of the importance of patient involvement alongside EBM has led to promotion of SDM. Additionally, increasing rates of LTCs has not only provided further support for patient involvement, but has also led to arguments for forward-planning, long-term, preventative approaches in the form of personalised care planning. Support for these approaches has been demonstrated throughout recent decades in government policies, academia and guidance from professional bodies. However, various barriers work against the adoption of these and in reality such approaches do not appear to be common place. At this stage, the way in which SDM and personalised care planning is effectively implemented, how barriers are overcome and whether such approaches are feasible or effective in reality remains unclear.

Chapter 2
An Introduction to Multiple Sclerosis

2.1. Chapter summary

This chapter is an introduction to Multiple Sclerosis (MS); it summarises widely understood theories about biological mechanisms and aetiology, its prevalence and some of the uncertainties that exist. It explores the services people with MS may access and potential decisions they may face. It then considers healthcare experiences and the prospect of personalised care planning and shared decision making (SDM) in the context of MS.

2.2. Note on terminology

In a recent survey, people with MS showed preference for the term 'MSer', followed by 'people with MS', but indicated little support for the term 'patient' (Baker et al., 2014). Throughout the thesis, the term 'people with MS' is used as it is considered acceptable by people with MS, but is more formal than 'MSer' (which is frequently and colloquially used on social networking sites). Two terms, 'disease modifying drugs' and 'disease modifying therapies', are both general terms used to refer to a group of treatments for MS. For the purpose of this thesis, the term 'disease modifying drugs' (DMDs) will be used throughout (unless using verbatim quotes from participants or extracts from literature).

2.3. Brief background on features of MS

MS is a neurodegenerative condition which typically manifests as gradual long term impairment, alongside episodes of symptoms (relapse) and recovery (remission) (Rae-Grant et al., 2011). There is a range of symptoms associated with MS. An international

survey from the World Health Organisation (WHO) reports the most common presenting symptom is motor weakness, spasticity or dysfunction (experienced by an estimated 50% of people with MS), then sensory problems and fatigue (40%), visual disturbances (31%), balance problems (22%), incontinence (17.5%), pain (15%), cognitive or behavioural problems and sexual dysfunction (10%) (Dua et al., 2008). There are also secondary issues such as obesity, osteoporosis and cardiovascular damage which may be affected by the way in which MS is managed (Döring et al., 2011). Estimates of the prevalence of MS ranges from 1-2.5 million people diagnosed worldwide (Noseworthy et al., 2000, Kantarci and Wingerchuk, 2006, Dua et al., 2008). In the UK specifically, there are no centrally collected data of neurological condition prevalence, but it has been estimated that around 100,000 people have been diagnosed with MS (National Audit Office, 2011) and it appears to be more prevalent in women than men with a ratio of 2:1 (Dua et al., 2008). After diagnosis, people with MS tend to live with the condition for much of their adult life as the majority are diagnosed between the ages of 20 and 40 years (Ontaneda et al., 2012); there is no known cure and treatments for symptoms are limited (Dua et al., 2008). MS, however, is not a terminal condition. Life expectancy does not differ greatly to population average (Loren and Rolak, 2003, Freedman, 2011) and those who have less severe or stable forms are more likely to die of other causes than MS-related complications (Freedman, 2011).

2.4. Aetiology of MS

Despite considerable research, the pathogenic mechanisms have not yet been conclusively identified (Diaconu et al., 2012) and the various conflicting theories have led some to argue that MS is better understood as a syndrome (various diseases with common overlapping symptoms), rather than a distinct disease (Lucchinetti et al., 2000, Rejdak et al., 2010). However, more commonly MS is understood as one disease and its

pathogenesis as an autoimmune response which affects the central nervous system (CNS); causing inflammation and progressive degeneration of myelin sheaths (connective white matter) which surround neuronal axons (nerve fibre) in the brain and spine. This is thought to disturb nerve connections and lead to MS symptoms and relapses (NICE, 2016). Magnetic resonance imaging (MRI) scans are used to identify CNS damage caused by MS. There is some evidence that the number of lesions is positively correlated with the severity or amount of symptoms (Kappos et al., 1999) but evidence demonstrating how patterning directly translates to symptoms is lacking (Lublin et al., 2014). So even with the aid of MRI scanning, MS symptoms and its clinical course are unpredictable.

Epidemiological research and demographic mapping suggest that a combination of environmental and genetic factors may contribute to MS risk (Rosati, 2001, Summerday et al., 2011, Ramagopalan et al., 2010). Although there is a large body of literature supporting associations of genetic factors (Ontaneda et al., 2012), vitamin D deficiency (Rosati, 2001, Summerday et al., 2011) and viral infections (Ramagopalan et al., 2010, Pohl, 2009, Ontaneda et al., 2012) with MS susceptibility, the exact causal links and mechanisms are unknown. MS aetiology appears to be multifactorial as research suggests effects are modest and combinations of these may predispose individuals to MS (Ramagopalan et al., 2010).

Certain triggers have been associated with MS relapses and long-term impairment, and therefore people with MS may adapt their lifestyles and behaviours accordingly. Diet (D'Hooghe M et al., 2012) and stress (Mohr et al., 2004) have been linked to relapse rates. Again, associations have been demonstrated, but causal links remain unknown and triggers vary between and even within individuals (D'Hooghe M et al., 2012, Summerday et al., 2011, Mohr et al., 2004). For example, a meta-analysis of studies

about stress and MS demonstrated that stress is associated with MS exacerbations, but the link is highly varied across different people, within individuals and according to the type of stress experienced (everyday and traumatic) (Mohr et al., 2004). Such variation means that people with MS may be likely to learn which factors act as relapse triggers for them personally from experience.

2.5. Disease course

The course of MS is highly variable with the frequency, pattern and type of symptoms differing widely between individuals and at different stages of an individual's disease trajectory. To make sense of this complexity, various definitions have been developed to classify different patterns of MS. The three types[2] most commonly referred to are:

- **Relapsing remitting MS (RRMS):** It is estimated that around 85% of people experience this pattern in the initial stages of the disease (Confavreux et al., 2003), which is characterised by interchanging periods of remission (good health) and relapse (symptoms) (NICE, 2016).
- **Secondary progressive MS (SPMS):** This stage develops after having RRMS at onset. It is characterised by gradual worsening of symptoms with fewer remissions, resulting in gradual deterioration (NICE, 2016). Some people are not diagnosed with MS until they are in this stage (Milne et al., 2012).
- **Primary progressive PPMS:** It is estimated that around 15% of people have this type at onset (Confavreux et al., 2003) which is characterised by symptoms which worsen over time without any remission (NICE, 2016).

2 Other types of MS, such as 'relapsing progressive', 'progressive relapsing', 'benign' and 'malignant', have also been proposed; however, these are less commonly used (Lublin and Reingold, 1996)

Although these descriptions are frequently used, they are somewhat limited and not recognised in the current edition of the World Health Organization's (WHO) 'International statistical classification of diseases and related health problems' (World Health Organization, 2016). These broad descriptions often do not sufficiently account for individual patterns of disease course and clinicians lack consensus on precisely which terms should be used to identify and describe different stages and patterns of MS (Lublin and Reingold, 1996)[3]. For example, according to an international survey most MS experts include patients with *some* residual effect after a relapse in the relapsing remitting category, and patients with *some* remission after a relapse in the secondary progressive category, with baseline deterioration being the defining feature discriminating relapsing remitting MS from secondary progressive MS (Lublin and Reingold, 1996). However, the rate of baseline deterioration or the amount of residual effect or remission remains undefined. A further limitation is that assessments of disease severity and pattern are often based on visible motor symptoms (particularly walking). This leads to less visible symptoms being overlooked and, as a consequence, flawed assessments of disease severity and overestimations of proportions of people with less severe MS (Correale et al., 2012). A cohort study of 163 people diagnosed with 'benign MS', (assessed as having no symptoms since initial diagnosis), reported that 45% had in fact experienced symptoms such as cognitive dysfunction, fatigue and depression, and neurological damage was visible on MRI scans. However, their symptoms appear to have been overlooked because they were less visible, resulting in diagnosis of 'benign MS' (Amato et al., 2006). Variation in symptoms and potentially

[3] The Advisory Committee on Clinical Trials of New Agents in MS surveyed 215 members of the international MS clinical research community. Respondents allocated pattern descriptions to MS type, this led to over 215 pattern descriptions being allocated to some description types (Lublin and Reingold, 1996), illustrating the variability of clinical course patterns.

flawed assessment methods mean that records on proportions, according to pattern or severity, are problematic (Edmonds et al., 2007).[4]

Expanding further than the physical symptoms, as in most chronic conditions, people with MS may experience 'biographical disruption' as their experience of illness will affect (or disturb) other more social elements, such as relationships and material resources (Bury, 1982). Typically the average age of MS diagnosis (20-40 years) is a period associated with the initiation of a career, establishment of spousal relationships and child rearing (McLaughlin and Zeeberg, 1993). At the point of diagnosis, people with MS have reported re-evaluation of their future expectations, reprioritising aspects of their lives and making life decisions which accommodate their MS (Thorne et al., 2004a). These life decisions may include: where to live; whether to adapt your home; whether to change or end employment; whether to have children; whether to end or change relationships; whether to take on different relationship roles.

2.6. Treatments available

There is a range of interventions and treatments available to manage specific MS symptoms (Dua et al., 2008). In treating MS more broadly, steroids are used to aid rehabilitation from relapses, and disease modifying drugs (DMDs) are used to prevent frequency and severity of relapses. DMDs have limited effectiveness, potential side effects and not everyone is eligible (only those with more active relapsing remitting forms are eligible). People with MS often seek more complementary and alternative approaches to manage their MS; most popular of which include: diet and nutrition, acupuncture, herbal medicine, massage and homeopathy (Dua et al., 2008). There are

4 Estimates of the prevalence benign MS range from as little as 5% to as much 64% of PwMS (Correale et al, 2012) Potentially every person with MS has this type after diagnosis, until they experience subsequent symptoms.

also everyday self-management strategies people may adopt, such as pacing oneself to manage fatigue or avoiding stress to reduce potential relapses.

2.6.1. Disease modifying drugs

DMDs are used to prevent relapse severity and frequency, but their effects are limited. As the Association of British Neurologists (ABN) guidelines explain: "disease-modifying treatment efficacy can only be partial, moderate and not curative" (Scolding et al., 2015, p.3). Globally, around half of people with MS who are eligible for DMDs receive them, ranging from a median percentage of 10% in lowest income countries and 75% in highest income countries (Dua et al., 2008). The main reasons why people eligible for DMDs do not receive them are due to funding policies (57.7%) and patient choice (61.1%) (Dua et al., 2008). The various types of DMDs are stratified on two levels: people with MS typically begin with basic DMDs (interferons or glatriamer acetate). Then if, or when, these appear to be ineffective, people with MS may use immunosuppressives which have more severe risks (Rejdak et al., 2010). On-going DMD trials and increasing numbers of licensed DMDs available has led to increasing decision-making complexity around risks and benefits (Heesen et al., 2011). Evidence demonstrating that DMDs slow the overall progression of MS is weak, and difficulties of carrying out longitudinal clinical trials mean long term efficacy and side effects are unclear (Scolding et al., 2015). Their effectiveness differs substantially between individuals and at different stages of one person's MS disease progression and so there are no standardised markers to identify when and which DMDs are appropriate (Kachuck, 2011).

Decisions about DMDs are not limited to deciding whether and which type to take, but another key decision is *when* to start using DMDs (Thompson, 2006). As the course of

MS is unpredictable, it is difficult to assess the optimum time to start taking DMDs. For example, initiating DMDs at pre-diagnosis stage has been shown to delay the development of clinically definite MS (Comi et al., 2001, Jacobs et al., 2000) and patients may have a better chance of any potential (but unproven) long term benefits. However, on an individual level, such early treatment would be less beneficial for those with a benign or slowly progressing form of MS (Miller et al., 2005a). Choosing the best time to *stop* taking, or *change* DMD treatment is equally challenging. The ABN's guidelines recommend people start DMDs when eligible, but do not provide generic criteria for stopping or changing treatment, and highlight the difficulty of concluding that a DMD is ineffective for individual cases (Scolding et al., 2015). Such complexities have led the ABN to recommend that assessments are made on an individual case basis and that decisions are shared between patients and neurologists:

"The complexities and uncertainties that we have outlined, combined with the now significant range of treatments available for relapsing-remitting MS, make treatment decisions complex for both patient and neurologist. We stress the importance of shared conversations about disease activity, risk and benefit, to make a choice that is right for the individual and their circumstances." (Scolding et al., 2015, p.2)

The guidelines also highlight the importance of on-going monitoring of the effectiveness and side effects of DMDs on individual case basis (Scolding et al., 2015). The way in which such guidance is applied in reality has not been extensively explored in the literature thus there is a need to examine interactions between healthcare professionals and people with MS, and how they are involved and informed during decisions about DMDs.

2.6.2. Complementary and Alternative Medicine

Survey data has shown that people with MS commonly use complementary and alternative medicine (CAM) strategies to manage their condition (Apel et al., 2005, Nayak et al., 2003). CAM may be defined in various ways, but for the purpose of this thesis, it is defined by what it is not; CAM is an intervention that is not considered a conventional approach or formally recommended by the medical profession. There is an array of such approaches that may be adopted by people with MS. These range from diet change, acupuncture and reflexology, to more controversial approaches such as the use of parasitic worms (Correale, 2014) and vascular surgery to treat chronic cerebrospinal venous insufficiency (CCSVI) (Laupacis et al., 2011) (explored further below). In a condition such as MS where often there are no conventional approaches suitable, people may find CAM particularly appealing. A survey in the USA into CAM use by people with MS found that those who adopted these approaches were more likely to have had MS for longer and be dissatisfied with conventional medicine (Nayak et al., 2003). Reported benefits from using CAM often go beyond physical effects. Some people with MS have reported feelings of empowerment and perceived practitioners as being particularly caring and good at listening (Shinto et al., 2005). A UK based study that combined a questionnaire and follow-up workshop found that people with MS reported benefits such as "enhancing relaxation, reducing specific physical symptoms and inducing general well-being" (Esmonde and Long, 2008, p.178).

2.6.3. Chronic cerebrospinal venous insufficiency

There are many controversial MS theories and treatments, but one procedure worth noting is vascular surgery (liberation procedure) to treat CCSVI, as it has gained much attention in recent years. CCSVI is controversial because it proposes an alternative

theory for MS pathogenesis (Diaconu et al., 2012). The chief proponent of this theory, Prof Zamboni argued MS is *not* caused primarily by an immune malfunction as widely believed, but by CCSVI (or abnormalities in circulatory systems) causing a build-up of iron deposits in veins, resulting in a secondary immune response (Zamboni et al., 2008, Singh and Zamboni, 2009). Although largely disregarded by healthcare professionals, for a while the evidence available neither supported nor refuted the theory (van Zuuren et al., 2012). More recently, evidence discrediting the theory appears to be increasing (Kugler et al., 2015, Tsivgoulis et al., 2015) with some warning of adverse events (Ghezzi et al., 2013). CCSVI is therefore typically not acknowledged in usual practice (Ontaneda et al., 2012, Diaconu et al., 2012). However, claims made about the effects of having the liberation procedure by advocates are dramatic, even curative (Mazanderani et al., 2013). When people are experiencing a debilitating (or potentially debilitating) condition with limited conventional strategies available, it is easy to see the appeal of such an approach.

2.6.4. Self-management strategies

As with most long term conditions (LTCs), people with MS self-manage their condition on a daily basis. Self-management not only has the potential to benefit management of symptoms, but may also enable people with MS to gain a sense of control over the disease and reduce feelings of anxiety (McLaughlin and Zeeberg, 1993). Self-management strategies may change daily and at different stages of the condition due to its fluctuating course, and these may include scheduled rest periods, dietary changes, exercise routines and life changes to reduce stress. Thorne et al.'s interview study found that as time went on people with MS described a process of taking responsibility for management of both the clinical and personal aspects of the disease, and appreciated

clinicians who adapted their relationship in light of this (Thorne et al., 2004a). A national survey of people with long term neurological conditions (LTNCs) reported that the majority of respondents (94%) felt that they had received enough information about management of medication; however, only a minority (27%) reported that they had been given support in developing strategies for self-management (Fitzpatrick et al., 2010). This suggests there may be missed opportunities for clinicians to provide more support in this area.

2.7. Healthcare experiences

2.7.1. Diagnosis

The period of diagnosis has been identified as particularly stressful for people with MS (Dua et al., 2008, Milne et al., 2012, Methley et al., 2015). The process of diagnosis is often lengthy; symptoms suspected to be the first sign of MS defined as clinically isolated syndrome (Miller et al., 2005b) are not enough to confirm diagnosis. Additional evidence, such as subsequent symptoms, tests to exclude other conditions and MRI scans, are required to confirm the presence of MS (McDonald et al., 2001). The WHO reported the median reported length of time to confirm a diagnosis is between 4 and 48 weeks; however, the range was broad, spanning from 1 week to 480 weeks (9.2 years) (Dua et al., 2008) and a recent review of MS healthcare services in UK reported that some diagnoses took over 17 years to be confirmed (Methley et al., 2015). This diagnostic period can be very distressing as people with suspected MS undergo various tests and are faced with the uncertainties of potentially serious unexplained symptoms (Milne et al., 2012, Dua et al., 2008).

2.7.2. Coordination of healthcare

Before diagnosis, people with MS typically have contact with a GP and then, during diagnosis, a neurologist. However, as their disease progresses and they experience different symptoms, they may encounter a diverse range of healthcare services (NICE, 2014), which is typical for most people with LTNCs (Fitzpatrick et al., 2010). Such services may include: "GP; neurologist; hospital inpatient; hospital outpatient; community specialist nursing; physiotherapy; speech and language therapy; dietician; continence advisor; special clinic (pain etc.); mobility and home nursing equipment (e.g. wheelchair); specialised neurological services (e.g. neurosurgery, neuro-rehabilitation, neurophysiology); general and community services (e.g. pharmacy services)" (Fitzpatrick et al., 2010, p.15). Alongside healthcare, a range of additional services may be accessed in relation to MS. These may come from social services, charities, the private sector, financial and employment advisors, and transport, housing and education services (National Audit Office, 2011).

In 2005, the Department of Health identified various problems with healthcare services for people with LTNCs living in the UK including: lengthy diagnosis; poor information on the condition and services available; variable access and little integration of health and social services; and poor quality of care in hospitals. The Department of Health introduced the National Service Framework for LTCs; its priorities addressed these problems by aiming to put individuals at the centre of care with personalised care plans, increased access to services, and reduced waiting times (Department of Health, 2005). However, despite extra public spending[5], a subsequent review by the National Audit Office (2011) demonstrated that overall the framework's priorities were not achieved.

[5] Between 2006 and 2010 there was a disproportionate 38% increase in spending on healthcare for neurological conditions.

The review stated that the framework's failure was due to poor execution and lack of accountability (National Audit Office, 2011). This suggests the policy may have been impractical given the reality of healthcare services, highlighting the difficulties of changing established healthcare systems, cultures and routines, and the challenges of implementing policy (explored further in Chapter 1).

People with MS, and more broadly LTNCs, have reported a lack of coordinated care as a major difficulty (National Audit Office, 2011, Fitzpatrick et al., 2010, Thorne et al., 2004a, Edmonds et al., 2007). The National Audit Office (2011) advise that all people with LTNCs should have a central point of contact coordinating their care and a personalised care plan. A national survey of people with LTNCs reported that only 36% of people with MS, Parkinson's Disease and motor neurone disease felt there was a central point of contact overseeing coordination of their care, and just 22% had a care plan (only three quarters of which were kept up to date) (Fitzpatrick et al., 2013).

2.8. Provision of information

Access to current relevant information is argued to be particularly important for people with MS because it is a disease with high levels of uncertainty over its causes and patterns (Edmonds et al., 2007). Access to current information may allow people with MS to manage their condition, maintain independence and enable empowerment during decision making (Dua et al., 2008). In addition to manging symptoms, accessing information and gaining knowledge may alleviate fear and uncertainty (Thorne et al., 2004a). The National Audit Office advises that all people with MS are given an MS information pack along with contact details for charitable organisations, local and national services (National Audit Office, 2011). However, a survey found that of those newly diagnosed with MS in the UK, only 64% received written information and 61% were given details of national MS organisations (Royal College of Physicians, 2011).

Furthermore, a recent review of healthcare services for people with MS identified a lack of timely information as a key concern (Methley et al., 2015).

2.9. The prospect of shared decision making and personalised care planning for people with MS

As explored in chapter one, arguments have been made for the value of patient involvement alongside healthcare professionals' in an SDM process, as well as preventative long term approaches for people with LTCs in the form of personalised care planning (an approach which incorporates SDM). In this chapter, MS has been shown to be a complex condition that is unpredictable, highly variable between and within individuals with limited treatment options available. Some have argued such uncertainty means that SDM is particularly suitable for people with MS. As Heesen et al. (2007a) put it:

> "MS is a prototypic condition for a shared decision process. […] This is the "equipoise-condition" which asserts scientific evidence alone can never lead to an individual treatment decision" (Heesen et al., 2007a, p.109-111)

In other words, the best course of action is often unclear from a medical perspective, therefore patients' own judgements, values and preferences may factor more pivotally during the decision making processes.

There is also evidence suggesting involvement in decision-making is received positively. Koudriavtseva et al. (2012) surveyed people with MS in an Italian cohort study and demonstrated that patients who assessed their treatment as useful were also more likely to feel they were involved in decision making. A German survey of people with MS and their decisional preferences found the majority (79%) preferred to be actively involved in decision making; the remainder mostly preferred a 'professional-as-

agent' approach, where healthcare professionals make decisions whilst factoring in patients' personal circumstances and preferences. Only a small minority preferred to leave decisions to healthcare professionals entirely (paternalistic) (Heesen et al., 2004). Hamann et al.'s (2007) research into a decision aid intervention for people with hypertension, depression, breast cancer, schizophrenia, minor traumas and MS, recorded patient preferences for involvement in decision making at baseline. They found that there was little difference in patient preferences for taking part in decisions according to whether the condition was acute or chronic. However, participants with MS had significantly higher preferences for involvement than those with other conditions (Hamann et al., 2007).

Successful management of a varied and fluctuating condition such as MS can require coordination of care due to potential for a varied range of health services to be used, and such services may be changed repeatedly as the condition changes. Also, fluctuations mean that assessments made during periods of temporary better health may not be comprehensive or realistic (National Audit Office, 2011)**.** Therefore, there is a need for effective communication, personalised long-term monitoring of the condition and coordination of care. Such requirements may be fulfilled by adopting a personalised care planning approach, however as mentioned above (in 2.7 Healthcare experiences), although mandated by policy, people with MS do not appear to experience this approach.

2.10. Summary

MS is a highly complex and unpredictable condition, with triggers, symptoms and patterns that vary widely between, and within, individuals. The varied nature of MS and lack of instrumental disease measures means that people with MS learn about their MS through experience. People with MS make treatment decisions in a context of

uncertainty. There is no known cure and medically recommended treatments available are relatively few. It is unclear whether or when interventions are required due to its unpredictability. Those who are eligible may start DMDs, however their effectiveness and side effects (some of which are severe) vary between individuals and it is unclear when to start, stop or change DMDs. Many people with MS opt for CAM, ranging from diet change, acupuncture and hyperbaric oxygen therapy, to more controversial approaches which include vascular surgery to treat CCSVI. However, when opting for CAM there may be little official advice available and there is unlikely to be much evidence demonstrating effectiveness.

Studies suggest that the amount and timing of information provided to people with MS is suboptimal and can be improved. Some have argued that information provision is key in alleviating uncertainty, but the impact of accessing information that is in itself uncertain is unclear. Some areas of healthcare have been identified as lacking for people with MS; these include self-management support, diagnostic processes, information provision and coordination of care. The National Service Framework aimed to address these areas by recommending people with LTNCs have personalised care plans and a central contact coordinating care. Personalised care planning would allow people with MS and healthcare professionals to carry out SDM, whilst forward planning, monitoring condition and coordinating care. However, these aims do not appear to have been achieved in practice. Although in theory, personalised care planning and SDM appear to be the best approaches, it is unclear whether they are feasible or effective for people with MS in reality.

Chapter 3

Methods

3.1. Research aims

As explored in chapter 2, Multiple Sclerosis (MS) is a long term condition (LTC) which is unpredictable, with limited treatments available and uncertainty around the best course of action to follow. Existing literature has highlighted the lack of coordinated care and timely information for people with MS, and uncertainty in MS positions patients' input as particularly relevant when making decisions. This thesis explores the information and decision support needs for people with MS. In doing so, it aims to identify problematic areas for people with MS and consider how these may be improved with information provision and decisional support. The study also examines the usefulness and potential application of shared decision making (SDM) and personalised care planning in an MS context, as these approaches have been advocated by government policies, official bodies and supported in much of academic literature consistently throughout recent decades.

3.2. Research questions

One overarching research question followed by four sub-questions were developed in order to examine the research aims.

Overarching research question

What are the decision support and information needs for people with MS?

Research sub-questions

1. What does the literature tells us about the effectiveness of a personalised care planning approach for people with MS?
2. In which contexts are decision support and information needs particularly relevant for people with MS?
3. How can healthcare professionals most effectively support people with MS?
4. What elements are important to consider in personalised care planning for people with MS?

3.3. Overview of methods

3.3.1. Methods stages

In order to answer the research questions three main stages of methods were used:

A. Two systematic literature reviews assessing effectiveness of personalised care planning for people with long term conditions and more specifically people with MS
B. In-depth semi-structured qualitative interviews with people with MS
C. Focused qualitative interviews with consultant neurologists

The table below indicates which methods will be used to answer each question:

Table 3.1. Research questions and corresponding methods

Research question	Methods used
1. What does the literature tells us about the effectiveness of a personalised care planning approach for people with MS?	A
2. In which contexts are decision support and information needs particularly relevant for people with MS?	B
3. How can clinicians most effectively support people with MS?	B and C
4. What elements are important to consider in personalised care planning for people with MS?	A, B and C

These methods are described in more detail under the relevant subheadings below.

3.3.2. Ethical considerations

As stages B and C were qualitative, I adhered to the British Sociological Association's statement of ethical practice (2002) to ensure good ethical practice. Participants' personal information was kept confidential and interview data is anonymised in all research outcomes. It was made clear in information sheets that participants were under no obligation to take part, and they could withdraw at any point leading up to, and during, the interviews. Audio recordings and transcripts of the interviews are stored securely on a password protected hard drive.

Specific risks involved in taking part in the interviews were assessed as minimal. However, it was possible that taking part may have exacerbated fatigue (common MS-related problem) or caused distress if there was discussion about previous stressful experiences. These risks were detailed in the information sheet, and participants were verbally reminded before the interview started that they did not have to answer interview questions (if they preferred not to) and they could stop the interview at any

time without providing explanation. All participants signed a consent form before the interview began and interview data copyright form at the end of their interviews. (See appendix A for participants' documents).

This research gained ethical approval from the Medical Sciences Inter-Divisional Research Ethics Committee, University of Oxford (MSD-IDREC-C1-2013-020).

3.3.3. Funding

This doctoral research was funded through an NIHR School for Primary Care Research studentship.

3.4. Systematic reviews

To assess the effectiveness of personalised care planning for people with MS, two systematic reviews were carried out. One review assessed the effectiveness of personalised care planning for people with LTCs and was published as a Cochrane review in 2015 (Coulter et al., 2015). Developing on from this initial review, the protocol was adapted and a subsequent systematic review on the effectiveness of personalised care planning for MS specifically was carried out. Findings from both reviews contributed to answering research sub-questions 1 and 4. For ease of reading and in order to keep the processes, methods and results of the systematic reviews together, the methods used to carry out the reviews are described further in chapter 4.

3.5. In-depth qualitative interviews with people with MS

Qualitative research methods are particularly suitable when exploring complex subjects (Ritchie, 2008). As explored in chapter 1, the processes of using information, making decisions and patient-healthcare professional interactions are complex and mediated by norms shaped by history and tradition. This stage of the research was theory *generating*

rather than theory *testing*, as I aimed to understand and identify contexts particularly relevant to people with MS in terms of their information use, decision making and interactions with healthcare professionals. Therefore, using qualitative methods, an inductive approach was adopted in order to draw conclusions from explorative findings (Bryman, 2012). Through a series of in-depth semi-structured interviews with people with MS, I aimed to gain insights into their experiences and perspectives on accessing information, making decisions and interactions with healthcare professionals.

3.5.1. Sampling, recruitment and analysis - MS interviews

Originally proposed by Glaser and Strauss (1967), grounded theory is an approach to social research that focuses on theory generation from data, without previous assumptions that may limit analytical processes. In this stage of the research, although I aimed to generate theory from interview data allowing participants to discuss areas important to them, I also had some idea about some key areas to explore based upon research aims. Therefore, I took a modified grounded theory approach to the collection and interpretation of interview data. Methods of sampling, recruitment and analysis are discussed together due to their interdependence and iterative nature of the processes.

In qualitative research, using a sample made up of purposefully selected participants is considered good practice, which aims to optimise the relevance of data analysed (Morse, 2010). Initially, participants were recruited using a broad purposeful sampling approach (which was later refined). The inclusion criteria were that participants had an MS diagnosis, were over 18 years old and lived in the UK. Recruitment was carried out via MS charity websites and newsletters; social networking websites (Twitter, Facebook and Shift.MS); university college newsletters and word-of-mouth. Posters were put up in communal areas of staff rooms (e.g. schools, office workplaces and university),

support groups and libraries. Members of the patient and public involvement (PPI) group were asked to distribute posters amongst mailing lists and groups they were members of. (Further information about PPI activities are detailed below).

After initial interviews were analysed purposeful sampling was refined; recruitment was guided by the need to explore emergent themes further and with an aim to recruit participants with a range of backgrounds. For example, when it emerged that 'uncertainties' was a key theme, I wrote a guest blog post about this on a social networking website (Shift.MS) with a link recruitment poster and when initial participants were all white British young adults, I aimed to purposely recruit participants who were older and/or from ethnic minorities. This was done by prioritising these volunteers to be interviewed first and using taglines (calling those who were old and from ethnic minorities to volunteer) posted on Twitter. (See appendix B for recruitment documents). Some snowball sampling also took place, as participants spontaneously suggested others they knew with MS who would have interesting perspectives to share. Participants were given information sheets and an opportunity to ask questions about the study. Informed consent was given before each interview.

As it is not possible to predict the amount of data needed to explore emergent themes and the content within each interview, sample size was guided by the principle of data saturation. This approach means that data is collected until "there are fewer surprises and there are no more emergent patterns in the data" (O'Reilly and Parker, 2012, p.192). Data saturation is a challenging principle to follow and there is little practical guidance available in how to determine when data is saturated (Bowen, 2008). The approach relies on researcher judgement, and some argue we can never know whether data is truly saturated, as there is always the potential for subsequent data to be discovered and additional interpretations to be made (Wray et al., 2007). In reality,

practical constraints such as time and resources may also inhibit potential for data saturation. However, failure to reach saturation does not mean findings are invalid, rather it means they could be explored further (O'Reilly and Parker, 2012).

Following grounded theory principles, data analysis began alongside data collection. I continued to analyse and collect data simultaneously, and scrutinised emergent themes through constant examination and comparison (Charmaz, 2006). Interviews were audio recorded and transcribed verbatim. As a first step in analysis I coded interview transcripts thematically using NVivo10 software. The coding process involved preparing the interview data for later analytical stages by assigning codes to each datum (or section of interview text). A code is defined as:

> "a word or short phrase that symbolically assigns a summative, salient, essence-capturing, and/or evocative attribute for a portion of language-based or visual data" (Saldana, 2013, p.3).

Categories, themes and theoretical ideas developed from the on-going analyses of the coded data. Emerging themes were tested and explored further during subsequent interviews, and data was coded again in response to developing codes and categories (Saldana, 2013). Code descriptions were created and frequently revisited and edited to enable on-going systematic reflection of the development of codes and categories. This was considered alongside more general reflections about individual interviews and theoretical reflections (Charmaz, 2006), which were recorded in the form of memos in NVivo. Coding audits were carried out at various stages of the analysis, whereby my supervisor (Dr Sara Ryan, an experienced qualitative researcher) coded several interviews and members of the PPI advisory group coded sections of interview transcripts. I compared their interpretations with mine in order to clarify my understandings and identify areas overlooked.

Guided by grounded theory, I took a fine coding approach to begin with; rather than lumping data together into broader codes, I coded interview data in very fine detail. Through constant revisiting of developing the coding framework and familiarisation with the data, themes began to develop. With time and through experience, it became apparent that the level of detail I initially took to coding was inefficient and unnecessary, as the analysis process became unwieldly and unmanageable. This was a useful learning process that allowed me to develop a more practical approach to analysis and illustrated the practical difficulties in following grounded theory principles. Throughout the coding phase, I had regular meetings with my supervisor (SR) to discuss the data, coding framework and emerging themes. The point where new codes stopped being created and potential patterns were identified in the data provided justification for data saturation. At 22 interviews, data collection stopped as nothing new was emerging from the key themes identified during analysis. During the next stage of analysis, I scrutinised patterns within the data. This was carried out by collating data assigned to relevant codes, and mapping the content (Ziebland and McPherson, 2006).

3.5.2. MS participants

Twenty-two participants from a broad range of backgrounds were interviewed between February and September 2013. (Individual participant demographic information and interview summaries can be found in appendix C).

Table 3.2 Interview participant demographics

Sex:	13/22 (59%) Female
Age:	Mean: 43.7 years old Range: 27 – 63 years old
Ethnicity:	18 (81%) white British, 1 (4.5%) Black Caribbean/African, 1 (4.5%) white Czech, 1 (4.5%) British Italian.
Years since diagnosis:	Mean: 7.9 years since diagnosis Range: 1 – 23 years
Employment:	5 retired/welfare support, 1 unemployed, 4 freelance/self-employed, 2 managers, 10 employed as follows: actor; architect; wholesale supervisor; parliamentary research; sales assistant; public relations; journalist; secretarial; community support assistant and bank project support officer

3.5.3. Interview technique during interviews with MS participants

I developed an interview schedule informed by background reading, the research questions and pilot interviews. Three pilot interviews were carried out but not included in the data analysis. They informed the content and organisation of the interview schedule. During the pilot phase, I trialled a 'minimalist passive interviewing' approach, which uses very broad and open questions, allowing the participant to recount their experiences uninterrupted (Jones, 2003). Pilot interviewees were initially asked to talk about their experiences of MS, with no other direction. This approach aims to encourage participants to control the direction and content of the interview, revealing aspects that are considered important to them (Mruck and Mey, 2007). Such a technique is consistent with grounded theory principles where data content comes from participants who identify aspects important to them, rather than being restricted to produce certain topics. However, pilot interviews indicated that this approach was too broad and much of the data yielded was irrelevant to the research question. Considering this, I decided to adapt the minimal passive approach. As my research questions focused

on decision-making and interactions, (rather than broad experiences of having MS), I began the interview with a very open question asking participants to talk about their experiences of MS, but also asked them to focus on any decision-making experiences and interactions with healthcare professionals that they felt were particularly relevant. The modified technique meant that participants could still lead the content of the interview, but could also provide more explanation about the areas relevant to the research questions i.e. it enabled the interview data to remain relevant, whilst being shaped by what participants considered important. In the later stages of the interview schedule, I introduced an 'active interview' technique. This technique recognises and reflects on the effects the interviewer can have on interview content. Using this approach, an interviewer can probe further and even challenge participants' responses (Holstein and Gubrium, 1995). So, during the initial (modified minimalist passive) stage of the interview, where participants talked uninterrupted, I took notes about areas that were pertinent. Once the participant stopped speaking and looked to me for guidance, I probed further in order to explore these areas, again taking notes for subsequent probing. Probing allowed me to gain more comprehensive understandings and explore areas specifically relevant to the research questions. Then, in the final stage of the interview, if there was time, and based on areas explored in the first stages of the interview, I mentioned any topics I felt were missing from their interview, to allow participants to discuss further if they felt it was important. It is considered good qualitative practice to reflect upon and revise interview schedules during data collection period (Ziebland and McPherson, 2006) in order to generate high quality data. The interview schedule was continuously reflected upon throughout interviewing and modified accordingly.

3.5.4. Researcher's role during interviews with people with MS

Researchers cannot be entirely neutral during the interview process, in the same way participants cannot, because researcher and participant "provide a 'unique' stimulus for each other" which shapes what is and is not disclosed and how information is presented (Mruck and Mey, 2007, p.522). It was important to consider how my presence as the researcher may have influenced the interviewees' responses (Creswell, 2009). To address this, a fieldwork diary was kept throughout the research. Keeping such a diary is considered useful in understanding emotional and personal effects, and to reflect on how these may shape theory development and analysis (Arber, 2006).

During interviews, participants seemed very open to sharing their experiences with me. I initially anticipated that interviews would last around an hour; however, they were typically closer to 2 hours. At times, some participants appeared to feel judged during interviews; this is something that I did not anticipate. This may have been because much of the content was about decision-making and the role they played during healthcare consultations. Talking about previous decisions, or their behaviour during consultations, meant that participants were not only telling me about an experience they had had, but also explaining their rationale or justifying their actions to me. Considering this, I tried to be particularly sensitive if they appeared uncomfortable or regretful.

I studied sociology at university and worked as a research assistant for the Health Experiences Research Group prior to starting my doctoral research. This may mean that I am particularly mindful of social power inequalities and find it easier to see situations from patients' perspective rather than healthcare professionals'. This may have allowed participants to feel comfortable in recounting times when they felt unfairly treated, as I could demonstrate empathy, allowing participants to talk about difficult interactions. It may have also influenced my interpretation of the data, as it is possible that more

positive interactions were overlooked. My supervisors (SR and LL) noted that there were substantially fewer positive compared to negative examples of interactions with healthcare professionals in the interpretations of data. In view of this, I revisited findings, but this time distancing myself from participants' accounts and ensuring that I considered potential healthcare perspectives when interpreting findings. This allowed a more rounded interpretation of data, which was examined further during structured qualitative interviews with consultant neurologists (see below).

3.6. Structured qualitative interviews with consultant neurologists

Grounded theory methodology recommends that implications of theories developed should be further explored (Bryman, 2012). Presenting findings from earlier stages of research to a subsequent group of participants can facilitate further analysis by providing insights into areas which are lacking and the translation of findings into reality (Morse, 2010).

3.6.1. Structure of interviews with consultant neurologists

In this stage of this research, findings judged to be particularly problematic from analysis of the MS interview data[6], were presented to and discussed by consultant neurologists during structured qualitative interviews. These findings were judged to be problematic because they posed specific dilemmas faced in provision of patient-centred care. These interviews were structured, in that infographics were used to aid discussion, and they were limited to approximately 30 minutes (to accommodate neurologists' availability). The infographics represented two themes, 'imposed consumerism' and 'feeling dismissed' with vignette examples in the form of quotations from the MS

[6] The abbreviated term 'MS interview data' will be used to refer to the data obtained from the in-depth qualitative interviews with people with MS and 'neurologist interview data' will be used to refer to data obtained from structured qualitative interviews with consultant neurologists.

interview data (see appendix D). In using vignettes, I aimed to problematise 'everyday' or routine behaviour, thus allowing careful considerations of aspects which are normally 'unquestioned' and not typically volunteered in normal discussion (Jenkins et al., 2010). Participants were shown the infographics, and discussed their interpretations, solutions and barriers to these.

3.6.2. Researcher's role during interviews with neurologists

Before commencing interviews, I carefully considered my role and how this may affect neurologist participants' perceptions and their responses. The content of these interviews was likely to be contentious, because the very aims were to discuss interview findings judged to be particularly problematic. When designing infographics that posed dilemmas, I ensured this was sensitive to neurologists' position. For example, I used the term 'ethical' in an infographic to position neurologists' stance; before each interview I explained that the content was selected due to the problematic nature; and, when neurologists put forward potential justifications for situations, I restated these to demonstrate my understanding of their perspective.

3.6.3. Sampling and recruitment for neurologists' interviews

Upon commencing fieldwork, I planned to present findings from MS interview data to healthcare professionals; however I had not identified which healthcare professionals would be most relevant to gain feedback from. Based on analyses of MS interview data, and discussion with supervisors (SR and LL), we identified consultant neurologists as being well positioned to provide further insights into some of the problematic areas identified from analysis of the MS interview data. Twenty consultant neurologists were approached and invited to take part in a 30-minute interview at a time and place

convenient to them. Six consultant neurologists were recruited from 3 distinct locations across the UK and were interviewed between August and November 2016. Participants were given information sheets and an opportunity to ask questions about the study. Informed consent was given before each interview. (See appendix A for participant documents).

3.6.4. Analysis of neurologist interview data

Interviews were audio recorded and transcribed verbatim. Using an iterative approach, I aimed to examine neurologists' perspectives on findings from analysis of MS interview data and therefore framework analysis was carried out. Framework analysis is particularly suitable for exploring differing views of key topics and "making comparisons within and between groups" (Gale et al., 2013, p.7). During framework analysis a matrix is created, with 'cases' along one axis and 'codes' along the other. Within this matrix, data is systematically organised in preparation for analysis and interpretation (Ritchie, 2008). When organising the neurologist interview data, the data from each interview participant acted as a 'case' and the structured interview content was used to guide the 'code' axis. Once the data was mapped out, comparisons were made and patterns were identified and interpreted in reference to research questions and findings from previous stages.

3.7. Patient and public involvement

Patient and public involvement (PPI) can enhance the quality of research and ensure that it is relevant and appropriate for members of the public and people with the condition researched (Brett et al., 2014). A PPI group was organised to provide input throughout my research with aims to enhance its quality, applicability and credibility.

3.7.1. Organisation of the patient and public involvement group

Eleven members of the public volunteered to be take part in the PPI group, 7 of whom had MS and 3 had long-term conditions (LTCs). Various PPI activities were organised throughout the research and not all PPI members took part in every activity. Having 11 members allowed a broad range of perspectives, and for members to take part when it was convenient or suitable for them to do so.

There are few reported adverse effects of PPI, but some have reported feeling frustrated when processes and training are lengthy and inconvenienced by face-to-face meetings and difficulties in arranging transport (Shah and Robinson, 2007). In order to enable access (especially for those who have mobility issues), allow members to work at their own pace, reduce inconveniences and reduce costs, the PPI group was organised to allow tasks to be carried out remotely, at a time and place convenient to PPI members. A purpose-built website hosted tasks for PPI members to complete, and I corresponded with them via email and phone. PPI members were paid £75 for each task completed and were expected to spend 3.5 hours on each task.

Development of mutual respect between researchers and PPI members can encourage PPI members to be more forthcoming and reduce frustrations (Domecq et al., 2014). I aimed to gain a good rapport with members early on in their involvement and maintain good communication. In addition, there is evidence that studies face increased challenges (in success of PPI) when it is sporadic and roles are unclear (Brett et al., 2014). Before commencing PPI activities, I sent a role description to those interested in volunteering (see appendix E). This described potential ways they may be involved; it clarified what they could expect and what I expected from them. Being clear about the levels of involvement can allow PPI members to "maximise the usefulness of their

contributions" (Snow et al., 2015, p.12), as they are aware of limits to involvement and can provide advice relevant to the research. Once enrolled, I organised a phone call with each member. These informal conversations allowed us to introduce ourselves, talk about our backgrounds and the research. Although potential future tasks were scheduled, the nature of research work meant that timeframes were changeable and there were periods with reduced and increased PPI input. Therefore, involvement may have been perceived as sporadic. To overcome this I provided members with updates via email throughout.

3.7.2. Patient and public involvement activities

PPI has developed and grown substantially in recent years, and researchers have taken varied approaches when involving the public in their research; best practice is unclear and many have identified the need to review and assess the impact of varied approaches (Snow et al., 2015, Domecq et al., 2014). In order to maximise potential influence on the research, PPI members were involved early on (Brett et al., 2014). They then went on to provide interpretations, advice and feedback at various stages of the research. There appears to be some consensus that PPI is valuable when designing research, recruiting participants and disseminating to lay audiences; however, there is less certainty over (or fewer examples of) the value of involving patient and public more actively in carrying out and translating research (Domecq et al., 2014). Despite this lack of certainty over the value of PPI in this area, tasks were also designed for PPI members to interpret interview data (as a coding audit) and scrutinise findings (as a relevance check). However, although they were involved in these areas, it was in addition to, not instead of, the analysis I carried out i.e. such activities were to complement not replace the research tasks I was carrying out. Organising PPI in such areas aimed to broaden

interpretations, identify gaps and establish credibility and relevance of findings (Brett et al., 2014). The PPI activities that were carried out for this research are summarised in the table below:

Table 3.3. Patient and public involvement for this doctoral study

Task	**Rationale**	**Organisation and input**
A. Familiarisation with research	This allowed members to become familiar with the research and provided opportunity to comment on its overall design, aims and priorities.	As an induction, PPI members read a lay summary of the research background, aims and design in PDF form. This was sent via email and available online website. They were asked specific questions and gave general feedback.
B. Assistance with recruitment processes	PPI members helped with recruitment and provided feedback on participant documents.	PPI members provided feedback on recruitment documents: including adverts, patient information sheets, consent forms and copyright forms. They were asked to assess them in terms of: comprehensiveness and accessibility; attractiveness and ease of use. They also advised on areas to aim recruitment and distributed posters within groups they were members of.
C. Prioritisation of outcomes	This ensured outcomes demonstrating effectiveness of personalised care planning in the systematic reviews were relevant and important to people with long-term conditions and MS.	PPI members were presented with background to the systematic reviews and the outcomes identified (by the review team from literature). They were then asked for feedback about these outcomes and ranked them in order of importance. These were then collated, and two outcomes were identified as primary, and two as secondary. For the review focused on MS only, one of the secondary outcomes was adapted, because it was ranked as low priority by PPI members with MS, and one member provided insight into why it would not be relevant to people with MS.
D. Coding interview data	PPI members interpreted data and identified codes which could be compared to the ongoing analysis. This audit process aimed to improve the robustness of analysis by identifying misinterpretations or areas overlooked.	PPI members with MS were given instructions on how to code, and extracts from 3 interview transcripts on two sides of paper to code themselves. Codes identified by PPI members were compared to the coding framework to ensure areas were not overlooked or misinterpreted.

Task	Rationale	Organisation and input
E. Feedback on lay summaries of systematic review	To aid public engagement, PPI members provided feedback on the lay summary of the systematic review.	PPI members were presented a lay summary of the systematic review, and asked to comment on the accessibility of the language and suggest edits.
F. Scrutinise interview findings	PPI members provided feedback on interview findings in order to test and examine them, discuss their relevance and identity areas overlooked or missing.	PPI members took part in an online discussion. 3 blog posts were displayed online. PPI members provided feedback and discussed the findings in the comments boxes for each blog post.

In addition to the tasks detailed in the table above, PPI members will also be given opportunity to provide feedback on lay summaries of the overall research findings, and help to disseminate these amongst their own networks. (See appendix E for PPI activities).

Section two
Systematic Reviews

Chapter 4

A Summary of, and my Contributions to, a Cochrane Review of Personalised Care Planning for Adults with Long Term Conditions

4.1. Chapter summary

As part of this DPhil I worked with a team to carry out a systematic review into personalised care planning (PCP) for adults with long term conditions (LTCs), which was published as a Cochrane Review in March 2015 (Coulter et al., 2015). The findings from the review will contribute to the overall findings of my thesis when considering the effectiveness of PCP for LTCs and comparing this to people with MS specifically. The protocol developed for the Cochrane review was adapted and followed to carry out the first methods stage; a systematic review of PCP for adults with MS specifically (presented in chapters 5 and 6). This chapter outlines my contributions to, and briefly discusses the findings of, the Cochrane review.

4.2. Brief background

As explored in previous chapters, PCP for adults with LTCs has been identified as a priority by policy makers (Department of Health, 2011) and the World Health Organisation have promoted this approach for developed countries (Singh, 2008). However, in reality, few people with LTCs seem to experience this approach and evidence of its effectiveness is unclear (Burt et al., 2012, Newbould et al., 2012). This systematic review was carried out to assess the effectiveness of PCP interventions, and identify elements and contexts that aid or hinder its implementation.

4.3. Protocol development

The lead author Dr Angela Coulter (AC) and I drafted an initial protocol for the systematic review. We designed and ran a search strategy through relevant databases and independently screened these. We identified 30 (out of 1573) studies which were potentially eligible. This pilot stage allowed us to identify and overcome potential issues with the review; aided further development of the protocol and concepts; and confirmed that the review was manageable and practical.

After the pilot stage, the review was registered with the Cochrane Collaboration and additional authors joined the review team. During review team discussions and in accordance with Cochrane Collaboration requirements, we made three key developments to the initial protocol design, which were to; 1) expand the number of LTCs included, 2) refine our definition and conceptualisation of PCP, and 3) confirm which outcomes should be assessed as important to PCP. These key developments are described further below.

4.2.1. Expand the number of Long Term Conditions

The original review was commissioned by QORU (Quality and Outcomes of person-centred care policy Research Unit). QORU specified a list of LTCs that were considered most common and clearly long term (arthritis; asthma; heart disease; chronic obstructive pulmonary disease; depression; diabetes; epilepsy; MS; stroke; chronic pain; schizophrenia and bipolar disorder). Using this as a basis, in the original protocol we planned to only include studies that tested PCP for this pre-determined list of LTCs to ensure the review was practical, relevant and manageable. However, after further consideration and in response to advice from the Cochrane Collaboration, we decided not to use a predetermined list and expanded the protocol to include all conditions

which may be considered long term, bearing in mind our intention to make comparisons between types of LTCs during the analyses.

4.2.2. Defining and conceptualising personalised care planning

During various discussions with co-authors and in light of the relevant literature, we refined our definition and conceptualisation of PCP as follows:

> "We define personalised care planning as: an anticipatory (forward-looking), negotiated discussion or series of discussions between a patient and a health professional (perhaps with other professional or family members present) to clarify goals, options and preferences and develop an agreed plan of action based on this mutual understanding." (Coulter, 2012, p.2)

Expanding upon this definition, we outlined the processes involved in PCP (see Box 4.1). This acted as a tool to assess interventions and potentially consider effects of different stages of the model on outcomes.

Box 4.1 Processes involved in personalised care planning

"**A. Preparation:** The patient may be invited to a preliminary appointment to check their progress and undergo relevant clinical tests. Information (printed, electronic, written, or verbal) may be provided prior to or during the care planning appointment to encourage the patient to reflect on their condition. This might include test results, information about treatment options, or about health-related behaviours such as diet, exercise or smoking.

B. Goal setting: Aimed at agreeing treatment or management goals, the goal setting process involves eliciting and clarifying patients' understanding of their condition, their values, outcome preferences and priorities. Patients may be encouraged to talk about their experience of living with the condition, their beliefs and concerns, and their comprehension of, and reactions to the information provided. The discussion of what matters to them may cover treatment or management options, desired outcomes, lifestyle or behaviour changes, practical, social and emotional challenges and problem solving strategies. In personalised care planning patients have scope to influence the agenda for discussion, and the choice of goals and priorities is not restricted to a pre-specified list of professionally determined options.

C. Action planning: A plan is jointly developed for working towards agreed goals. This may include identifying practical ways in which the patient can achieve their behavioural goals (for example how and when to take more exercise), referring the patient to external sources of support, either within formal health services (e.g. health coaching or rehabilitation services), or in the community (e.g. exercise or cookery classes) or peer support. The plan may also include clinician-ordered tests or treatments, referral to other clinical specialists or other professionals, educational materials or courses, access to aids or appliances, care assistance or domestic help.

D. Documenting: The agreed actions are usually documented in a specially designed record (printed, electronic, or written) for use by clinician(s) involved in the patient's care and/or for use by the patient as an aide memoire.

E. Supporting: Patient and clinician agree a schedule for regular systematic follow-up that may involve a number of contacts (face-to-face, telephone, or electronic) to provide appropriate support to help the patient achieve their goals. This may take the form of health coaching or motivational support, problem-solving, or simply checking and reinforcing progress in implementing the agreed plan.

F. Reviewing: A meeting (face-to-face or remote) during which patient and clinician jointly review progress and plan next steps." (Coulter, 2012, p.4)

We identified two core stages, B. Goal setting and C. Action planning as defining processes within PCP. Therefore, studies included in the systematic review had to involve both of these stages.

This carefully considered conceptualisation guided further amendment of the inclusion and exclusion criteria within the protocol. For example, we decided to exclude studies with predetermined actions prescribed to participants (e.g. if participants were instructed to increase medication in response to specific test results), or those studies which were limited to specific goals (e.g. if interventions only focused on increasing participants' levels of physical activity). These exclusion criteria were justified because in our conceptualisation of PCP, the goal-setting process demands that "patients have scope to influence the agenda for discussion, and the choice of goals and priorities is not restricted" (Coulter, 2012, p.4).

4.2.3. Prioritisation of outcomes considered important in the review

Determining which outcomes are important to PCP was scrutinised at length during review team discussions. Guided by the literature and consultation with the patient and public involvement (PPI) group (described in chapter 3), four potential outcomes of PCP were identified as relevant and ranked in order of importance:-

Box 4.2 Relevant outcomes ranked in order of importance
"Changes in health and well-being. This may include changes in physical or mental health measured objectively (for example blood pressure, blood lipids, body mass index, HbA1c, urinary albumin, etc.) and/or subjectively (using validated patient-reported measures, including symptom scales, pain scores, depression or anxiety scores, fatigue, self-esteem, coping, quality-of-life, activities of daily living, etc.). Change in the patient's capabilities for managing their condition (assessed by themselves or by others), including knowledge of their condition, general health literacy, self-efficacy, self-management behaviours, activation, perceived competence, and ability to access relevant support. Change in health-related behaviours, including diet, exercise, smoking, use of relaxation techniques, condition-relevant self-monitoring, attainment of personal goals. Change in use of formal health services, including numbers and length of hospital admissions, and numbers of outpatient, emergency department, or primary care visits." (Coulter, 2012, p.7)

Based upon PPI members' feedback, 'changes in health and well-being' and 'change in the patient's capabilities for managing their condition' were identified as the primary outcome measures; and 'change in health-related behaviours' and 'change in use of formal health services' as secondary outcome measures within the review protocol.

4.4. Carrying out the systematic review

In accordance with the protocol changes, I redesigned the search strategy (see appendix H) and ran searches through the following databases from earliest records to July 2013: Cochrane Central Register of Controlled Trials, Embase, Medline, PsycInfo, Proquest, clinicaltrials.gov and WHO International Clinical Trials Registry Platform. Database search results were collated and de-duplicated, resulting in 9890 studies. Authors AC, Dr Sara Ryan (SR) and I sifted through the articles and independently identified studies for inclusion according to the study checklist (see appendix H). Any disagreements were discussed and in some cases other authors were consulted until agreement was

reached. 43 articles describing 19 unique studies were identified and included in the review. The risk of methodological bias of the included studies was assessed and data were extracted independently by myself, AC and SR then compared for accuracy. The data extracted were entered into RevMan software. Various analyses were carried out on the data by co-author Dr Rafael Perera.

4.5. Discussion of Findings

The systematic review found that PCP does moderately improve indicators of physical and psychological health, and people's abilities to self-manage compared with usual care. However, there was no clear improvement in subjective health status. Interestingly, the review also found that the interventions which were more intensive, comprehensive and better integrated into routine practices appeared to be most effective (Coulter et al., 2015). This emphasised the interdependence of components and importance of organisational structures in enacting a PCP approach. Batalden et al. (2015) outlined the importance of organisational features in enacting successful patient clinician partnerships:-

> "Our central tenet is that healthcare services are always coproduced by patients and professionals in systems that support and constrain effective partnership. We do not improve healthcare services by *adding* coproduction" (Batalden et al., 2015, p.4).

Supporting this statement, the review findings suggest that if PCP is to be successful, it needs commitment at organisational levels and activation of all stages of the process. The majority of studies yielded by the review included participants with diabetes (12 out of 19), followed by studies which included participants with mental health problems (3) and the remaining studies included participants with heart failure (1), end-stage renal disease (1), asthma (1) and various conditions (1). Elsewhere, PCP has shown

promising impacts for people with diabetes and there is much literature discussing the value of patient involvement, education programmes and self-management in diabetes (Davies et al., 2008, Snow et al., 2013, Year of Care, 2011). The strong evidence demonstrating the positive effects of patient involvement in diabetes suggests that PCP approaches may be particularly successful in diabetes care or positive impacts may be particularly measurable. Alternatively, it may reflect that PCP approaches for diabetes happen to be better studied compared to other conditions, thus there is more evidence available. Interestingly, the review yielded no eligible studies which included participants with MS, nor any other neurodegenerative conditions, suggesting this may be an under-explored area in the PCP intervention literature.

The skills and knowledge I developed when carrying out this Cochrane review and the gaps in evidence demonstrating the effectiveness of PCP for people with MS, provided a context where I went on to organise and lead the subsequent systematic review to assess PCP for people with MS specifically (see chapters 5 and 6).

Chapter 5

Personalised Care Planning for Adults with Multiple Sclerosis – Systematic Review Methods

5.1. Chapter summary

Starting with a brief background into the prospect of personalised care planning (PCP) for people with MS, this chapter then presents how the protocol for the previous Cochrane review (outlined in chapter 4) was adapted and used to carry out this systematic review into PCP for adults with MS.

5.2. Background

As explored in chapter one, recent decades have seen growing support for increased patient involvement and long-term preventative approaches to healthcare for those with long-term conditions (LTCs). In light of this, PCP has been put forward as a suitable approach for those with LTCs. There has been some promising evidence indicating favourable effects of PCP (see chapters 1 and 4). Focusing on MS specifically, there are features worth noting when considering the prospect of PCP. In chapter 2, it was demonstrated that MS is a condition that is uncertain and unpredictable with limited conclusive clinical information available (in terms of progression, treatments, triggers and causes), or reliable instrumental measures of disease activity. In this context, people with MS may draw upon their own experiences to assess effectiveness of interventions, identify relapse triggers and adapt their lifestyles accordingly. Despite the breadth of MS symptoms, official assessments of disease activity are often based on visible motor symptoms (particularly walking), while less visible symptoms (such as fatigue and cognitive difficulties) are overlooked in research literature (Correale et al., 2012). The presence of less visible symptoms highlights the importance of eliciting patients' input

in order to gain understandings about experiences of such symptoms and their effects on well-being. Equal partnerships between patients and healthcare professional brought about through PCP could potentially provide tools to value patients' input and gain mutual understandings.

As mentioned above, there is some evidence of positive outcomes from PCP for LTCs, however whether this approach is effective for people with MS specifically remains unknown. Several studies have shown people with MS tend to prefer to collaborate with healthcare professionals and be actively involved during healthcare decision making (Heesen et al., 2004, Hamann et al., 2007). Barlow et al. (2009) explored experiences of a 'lay-led chronic disease self-management programme' for people with MS. Participants reported positive outcomes such as learning new self-management techniques and improvement in feelings of self-efficacy. PCP may enable people with MS to communicate their priorities and experiences of their condition, but the usefulness of a forward planning preventative approach in a condition that is essentially unpredictable is less clear. As explored in chapter 2, the UK government has invested funding into and endorsed PCP for people with long term neurological conditions. But it reality such approaches do not appear to have been adopted, and it is unclear as to whether they are effective for people with MS.

5.3. Aims and objectives

The objective of this review was to assess if PCP interventions for people with MS are effective. It also aimed to examine reasons why PCP is effective or ineffective, and how people with MS experience PCP interventions.

5.3.1. Review questions

The two questions underpinning the systematic review are:-

1. Is personalised care planning effective for people with MS?
2. Why, and how, does personalised care planning for people with MS work or not work?

5.4 Protocol adaptations

A protocol was developed in order to carry out in this systematic review. This protocol was adapted from the Cochrane review of PCP interventions for people with LTCs (Coulter et al., 2015) (see chapter 4). The Cochrane review included randomised controlled trials (RCTs) that compared PCP interventions for people with LTCs with usual care. The adapted protocol for this systematic review is narrower in that it focuses on MS only in order to examine PCP in the context of MS specifically, but it is also broader in that it allowed inclusion of non-randomised studies (NRSs) to include more evidence about experiences of PCP.

As RCTs are considered the highest quality evidence when assessing the effectiveness of interventions, they were included in the review. In addition to RCTs, any relevant NRSs were also included for two reasons. Firstly, the expectation that there would be few RCTs available, as the Cochrane review found no RCTs testing PCP interventions for people with MS specifically. Secondly, due to additional evidence NRSs may provide. The designs of RCTs aim to eliminate all confounding factors which may affect results, but in doing so this also limits some aspects of their findings. The advantages of NRSs are that they can provide evidence about the long-term and unintended effects of an intervention and consider an intervention's applicability to different settings (Reeves et al., 2013). PCP is a complex approach; strategies devised to

enact it and settings in which it is performed may be varied and multifaceted components may result in unintended consequences. PCP is also a forward planning preventative approach with effects that may be long-term and missed within the limits of RCTs. By including NRSs I aimed to consider the way in which interventions are used, the unintended effects of complex interventions and longer term or undetermined outcomes through inclusion of NRSs. There are various study designs that come under definition of NRSs, however varied interpretations means there is a lack of consensus on how these ought to be described (Higgins and Green, 2011). Due to inconsistencies within study type descriptions, Cochrane's 'Non-randomised Studies Methodology Group' recommend that eligibility criteria are based upon assessment of explicit study design features of the studies, rather than study design labels (Higgins and Green, 2011). In addition to explicit study design features, eligibility criteria for NRSs may be determined by either a quality threshold, or guided by the amount other good quality evidence available (Reeves et al., 2013, Higgins and Green, 2011). For the purpose of this systematic review the eligibility criteria for NRSs was broad. Included NRSs were defined as:-

> "Any quantitative study estimating the effectiveness of an intervention (benefit or harm) that does not use randomization to allocate units to comparison groups. This includes studies where allocation occurs in the course of usual treatment decision or peoples' choices, i.e. studies usually called observational. In the scientific literature, there are many types of named non-randomized intervention study, including cohort studies, case-control studies, controlled before-and-after studies, interrupted-time-series studies and controlled trials that use inappropriate randomization strategies (sometimes called quasi-randomized studies)." (Reeves et al., 2013, p.2)

Qualitative studies were also included in this review. Qualitative methods alone cannot be used to assess effectiveness of interventions, however they may be used alongside effectiveness studies in order to examine processes of implementation and explore the ways in which interventions are used or received by participants and deliverers (Lewin

et al., 2009). PCP interventions are likely to be complex involving social and behavioural processes due to the various stages they may comprise and interactions between individuals in social settings. Quantitative methods are limited in assessing complex interventions that involve social and behavioural processes (Campbell et al., 2000), therefore any qualitative studies which exist alongside effectiveness studies were also included in order to explore these aspects. Inclusion criteria was not limited to specific qualitative methods, but was limited to studies that exist alongside effectiveness studies. The qualitative data included aimed to answer question:

2. *Why and how does PCP for people with MS work or not work?*

Inclusion of qualitative data may allow evaluation of the ways in which interventions work (or do not work) in varied settings. By exploring the experiences of individuals using (or receiving) interventions and examining factors that shape these, can enable deeper understandings of the mechanisms and practicalities of interventions (Mays et al., 2005).

5.5. Eligibility criteria

RCTs, NRSs and qualitative studies accompanying intervention studies were eligible for inclusion if they adhered to the eligibility criteria outlined below. Using these criteria, my supervisor (SR) and I independently assessed studies for inclusion in the review.

5.5.1. Participants

- Participants must be adults (aged over 18) with MS[7]. Studies where the age is not stated or the majority of participants are adults will be included.

[7] This may be any type of MS i.e. RRMS, SPMS, PPMS

- Studies must include participants with MS, however studies which additionally include participants with other conditions are also included.

5.5.2. Interventions

- Included interventions must include one-to-one PCP (not decision aid only, patient education only, psychological treatment only).
- Included interventions must have an explicit aim to actively involve patients in a collaborative or shared decision-making process when planning care or treatment with clinician, coach or other health professionals.
- Included interventions must include both collaborative goal setting and collaborative action planning.
- Included interventions must allow and encourage patients to have meaningful influence on their own goals or priorities, rather than being constrained by predetermined options.
- The PCP must be about current ongoing care, rather than 'end of life' care.

5.5.3. Comparison

- The experimental group will be compared to usual care.

5.5.4. Outcomes

Four outcomes were identified as being important in demonstrating the effectiveness of PCP interventions for MS. These are: 1) improvement in health and wellbeing; 2) improvement in capabilities to self-manage; 3) improvement in formal healthcare service use; and 4) attainment of personal goals. Further details are as follows:

1. Improvement in health and wellbeing: Changes in health and wellbeing may include objective measures of physical or mental health (e.g. Expanded Disability Status Scale and Cambridge Cognitive Examination) and/or subjective measures physical or mental health (e.g. pain scores, fatigue, self-esteem, coping, quality-of-life, activities of daily living, etc.).
2. Improvement in capabilities to self-manage: Changes in capabilities to self-manage assessed by themselves or by others. Examples include: MS knowledge, health literacy, self-efficacy, self-management behaviours and ability to access relevant support.
3. Improvement in formal healthcare service use: The PPI group agreed that although they did not consider this a primary concern, it was important. They made clear that improvement did not necessarily translate as *reduction* in use of healthcare services. Instead, it might be more efficient and coordinated use of healthcare services.
4. Attainment of personal goals: This replaced 'Changes in health-related behaviours' which was used as an outcome of the Cochrane review. This is because members with MS in the PPI group rated 'changes in health-related behaviours' as being a low priority compared to other outcomes. In the additional PPI comments, it was argued that the complexity of PCP and MS mean outcomes will be personal to each individual. Therefore, in this review attainment of personal goals has been identified as an outcome.

5.6. Information sources

In January 2015, electronic searches of databases listed below for all years were carried out-

- Cochrane Central Register of Controlled Trials (CENTRAL), *The Cochrane Library*, (Issue 12, 2014).
- Cochrane Database of Systematic Reviews. *The Cochrane Library*, (Issue 1, 2015)
- Database of Abstracts of Reviews of Effects. *The Cochrane Library*, (Issue 4, 2014)
- Dissertations & Thesis. (ProQuest) (1743-January 2015)
- Embase (OvidSP). (1974 to 2015 January 20)
- Medline & Medline In-process. (OvidSP). (1946-January 2015)
- PsycINFO. (OvidSP). (1967 to January Week 2 2015)
- Science Citation Index, Social Science Citation Index and Conference Proceedings Citation Index – Science (Web of Science, Thomson Reuters) (1945-January 2015)
- Clinicaltrials.gov (22 January 2015)
- WHO ICTRP (22 January 2015)

In order to identify additional papers from included studies and additional studies potentially eligible for the review, reference scans of relevant papers identified from the search were performed. When deemed necessary, I contacted study authors to retrieve missing data or clarify discrepancies.

The search strategy (see Box 5.1) is based upon the search strategy I devised for Cochrane review, but adapted to include studies focused on MS only (rather than LTCs more generally) and non-randomised and qualitative studies (rather than RCTs only):-

Box 5.1 Medline search strategy for MS systematic review

exp Multiple Sclerosis/
Demyelinating Autoimmune Diseases, CNS/
multiple sclerosis.ti,ab.
(demyelinat* adj3 (disease? or disorder? or condition? or illness*)).ti,ab.
1 or 2 or 3 or 4
Patient Care Planning/
((care or action or treatment) adj plan*).ti,ab.
decision making/
Choice Behavior/
(patient* adj7 (decision* or choice*)).ti,ab.
(patient* adj3 (preference* or priorit* or value*)).ti,ab.
Patient Preference/
(treatment adj (option* or choice*)).ti,ab.
Goals/
(goal* adj2 (set* or plan*)).ti,ab.
Patient-Centered Care/
(patient adj (cent*red or focus*ed or oriented)).ti,ab.
Individualized Medicine/
(individuali#e? or individuali#ing or personali#e? or personali#ing or tailor or tailored or tailoring).ti,ab.
6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19
(patient* adj3 (participat* or involv*)).ti,ab.
(negotiat* or agreement or concordan* or cooperat* or co-operat* or collaborat* or partnership).ti,ab.
21 or 22
5 and 20 and 23
Patient Participation/
(patient* adj2 (empower* or activat*)).ti,ab.
((shared or joint or informed or collaborative) adj2 decision making).ti,ab.
((involv* or participat*) adj3 (choice* or decision*)).ti,ab.
(decision adj (aid* or support or tool*)).ti,ab.
patient provider agreement*.ti,ab.
decisional self efficacy.ti,ab.
(personal budget* or direct payment*).ti,ab.
(record access or patient held record*).ti,ab.
((self management or self care) adj2 support*).ti,ab.
25 or 26 or 27 or 28 or 29 or 30 or 31 or 32 or 33 or 34
5 and 35
24 or 36

5.7. Data extraction

An experienced systematic reviewer Dr Ben Fletcher and I independently extracted data from included studies into an extraction table (see appendix I). This table recorded details about the participants; intervention details; study methods; outcome measures used; quality assessment according to the Cochrane's risk of bias tool; and results data.

We then compared and clarified methodological quality assessments and the data extracted to ensure accuracy.

5.8. Quality assessment

Biases within studies are confounding factors that can cause under or over exaggeration of true intervention effects. Thus, in comparison studies, variation in results between experimental and control groups may be due to differences between groups, rather than the intervention itself. Therefore, it is essential to consider potential biases within studies when assessing the true effects of an intervention (Higgins et al., 2011).

NRSs are at increased risk of confounding factors compared with RCTs due to the allocation process not being randomised, therefore it is important confounding factors are described and considered when including NRSs in systematic reviews (Reeves et al., 2013). Additionally, susceptibility to biases between NRSs vary substantially due to the broad variety of types of NRSs (Reeves et al., 2013), which means development of a generic tool to assess risk of bias in NRSs is problematic (Higgins and Green, 2011). Currently, the Cochrane Collaboration are in the process of developing a generic tool ACROBAT-NRSI in order to assess the quality of NRSs, however this tool is still in its pilot stage (Sterne et al., 2014). In the absence of a finalised suitable tool, the standard methods to assess risk of bias in RCTs set out by Cochrane Collaboration was followed (Higgins et al., 2011). This tool was also used to assess quality of RCTs included.

Approaches taken to appraise qualitative studies are different to those used to assess quantitative studies. A good quality quantitative study will eliminate confounding factors, with an aim to prevent results being systematically affected by other factors, whereas qualitative studies explore social and behavioural elements, which are normally examined in their context. There are disagreements about the best way to assess

qualitative studies and whether generic tools are useful (Seers and Toye, 2012), as such tools treat qualitative research as a "unified field" and overlook differences between, and complexities within studies (Dixon-Woods et al., 2004). However, there are some broad common features to most qualitative research and it has been argued that these features may be used as "cues to think about issues rather than explicit criteria" (Seers and Toye, 2012, p.1). In order to guide quality assessment of evidence from qualitative studies the Critical Appraisal Skills Programme (CASP) Qualitative Research Checklist (CASP, 2013) was used.

5.9. Data analysis

Meta-analyses are typically used in systematic reviews to pool data from RCTs to gain more reliable understandings of the effects of interventions. However, due to the likely heterogeneity of potential relevant outcome measures, the breadth of the inclusion criteria for study type and the lack of RCTs on MS identified in the Cochrane review, it was anticipated in the protocol that a meta-analysis would be unrealistic in this systematic review. If included in a meta-analysis, the allowance for bias within NRSs widens the confidence interval to an unknown extent. Therefore the inclusion of NRS in a meta-analysis is problematic and risks producing exaggerated effects (Reeves et al., 2013). Due to NRSs' increased susceptibility to confounding factors, it is recommended NRSs and RCTs are analysed separately and not incorporated in a meta-analysis together (Higgins and Green, 2011). Primary outcomes (subjective health status and capabilities for self-management) are vulnerable to heterogeneity, thus tools used to collect and measure intervention effects are likely to vary substantially between studies. Inclusion of studies with highly heterogeneous outcomes in meta-analysis may be impossible due to the difficulties of pooling dissimilar data (Higgins and Green, 2011). If enough RCTs with data judged to be homogenous were identified, then meta-analysis

would have been performed. However due to the inclusion of both a randomised and a non-randomised study, along with the varied outcome measures, a narrative synthesis was carried out (see chapter 6).

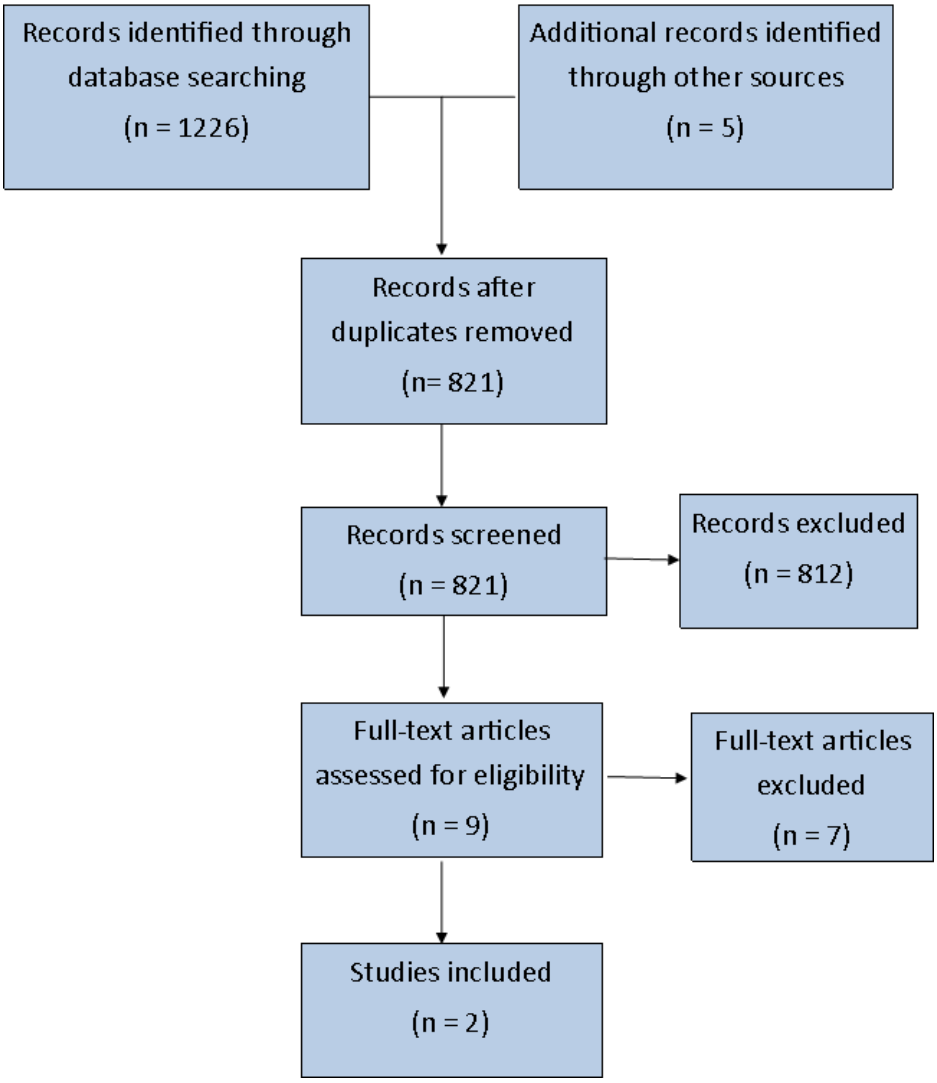
Chapter 6

Personalised Care Planning for Adults with Multiple Sclerosis Systematic Review Results

6.1. Results of the search

The database searches yielded 821 studies (after duplicates were removed). My supervisor (SR) and I independently screened titles and abstracts of these studies for inclusion. Nine studies were identified for potential inclusion and full texts of these studies were obtained and simultaneously assessed by myself and SR for inclusion. We then compared our assessments and any disagreements were resolved through discussion, or referred to another supervisor (AC) for a third opinion. Two studies were identified as eligible for inclusion and 7 were excluded. Reasons for exclusions are described in table 6.2 below. A colleague (who is experienced in carrying out systematic reviews) (BF) and I independently assessed the quality of – and extracted the data from – the two included studies (see appendix I). Any discrepancies were discussed and subsequent agreements made. Figure 6.1 is a PRISMA flow diagram (PRISMA, 2017) mapping the number of records identified, included and excluded:

Figure 6.1. Flow diagram mapping the number of records identified, included and excluded in the MS systematic review



6.1.1. Included studies

Two studies referenced below were included in the systematic review:

Block, P., Vanner, E. A., Keys, C. B., Rimmer, J. H. & Skeels, S. E. 2010. Project Shake-It-Up: using health promotion, capacity building and a disability studies framework to increase self efficacy. Disability and rehabilitation, 32, 741-54.

Eyssen, I. C., Steultjens, M. P., de Groot, V., Steultjens, E. M., Knol, D. L., Polman, C. H. & Dekker, J. 2013. A cluster randomised controlled trial on the efficacy of client-centred occupational therapy in multiple sclerosis: good process, poor outcome. Disability and rehabilitation, 35, 1636-46.

The Block 2010 study was a non-randomised trial of a community based personalised care planning (PCP) intervention for people with MS, spinal cord injury and other related neurological impairments. Thirty-three participants took part in the study. The Eyssen 2013 study was a cluster RCT of a PCP intervention for people with MS receiving occupational therapy. The intervention involved occupational therapists attending training and was set in outpatient clinics in hospitals and rehabilitation centres. The study included 269 participants. Further details about the included studies are summarised below:-

Table 6.1 Characteristics of included studies

Study	**Block 2010**	**Eyssen 2013**
Intervention dates	August 2002 - December 2003	May 2007 – March 2009
Deliverers of intervention	Researchers working with local Centre for Independent Living (a non-profit organisation providing community based recreational opportunities for people with disabilities).	29 Occupational therapists (OT); mean age 36.3; and 90% female. OTs in the intervention group had received client-centred training which adhered to components of the personalised care planning model.

Study	Block 2010	Eyssen 2013
Participants	People with neurological impairments: spinal-cord injury, n=16; multiple sclerosis n=12; traumatic brain injury n=2, stroke n=2; and cerebral palsy n=1, spina bifida n=1, and aneurism n=1 Mean age: 44.2 and 37% female n= 35. First intervention group n=13, second intervention group/wait list control n=13, comparison group: n=9	People with MS receiving occupational therapy. Mean age: 45.5 and 68% female. n=269, Intervention group: n=156 (51/207, 25% excluded), Control group: n=113 (63/176, 36% excluded). Lost to follow up at 4 mos: I = 6 , C = 4, at 8mos: I= 2, C=2
Recruitment setting	Organisational newsletters, mailings from State Department of Health and word-of-mouth at a local Centre for Independent Living and other local disability organisations	OTs aimed to select consecutive patients for recruitment who were already attending outpatient clinics
Intervention setting	Various community based settings, such as parks, libraries and university campuses. New York, USA	Outpatient clinics in hospitals (H) and rehabilitation centres (RC), Netherlands. I: 6 centres (4 H & 2 RC). C: 7 centres (3 H & 4 RC). 1 centre withdrew.
Comparison	Usual care	Usual care
Outcomes	Self-management: Self-efficacy	Health and wellbeing: Self-reported outcomes disability and fatigue, participation and autonomy, generic health-related QoL, upper extremity function, effects of pain on everyday life, cognitive functioning. Self –Management: Self-care is one of the domains measured in disability outcome.

Study	Block 2010	Eyssen 2013
Method of measurement	Self-efficacy: General Perceived Self-efficacy Scale which assesses participants' perceived sense of resourcefulness, ability to deal with unexpected events, and capacity to find solutions to problems using a 10 items Likert scale	Disability: disability and impact profile (DIP) a 39-item self-administered questionnaire. DIP domains include symptoms, mobility, self-care, social activities, communication and psychological status, with an additional two questions about fatigue. Impact on participation and autonomy (IPA) assessed impact of condition on perceived participation and autonomy of outdoor and indoor autonomy, family role, work and social relations. Upper extremity function: a nine hole peg test. Fatigue: fatigue was added to the DIP (above), and a modified fatigue impact scale was also used. Pain and disturbing sensations effect on everyday life: the pain effects scale. Cognitive functioning: the Perceived Deficits Questionnaire. Health related quality of life: the Short-Form Health Survey Questionnaire (SF36).

6.1.2. Description of comparison groups in included studies

In Block 2010, the intervention, 'Project Shake-it-up' was a program which promoted self-efficacy through the process of personalised goal setting, alongside peer support, education and access to community resources. The programme involved 10 sessions

over 5 months. Each session was made up of two parts: firstly, interactive workshops which included health seminars, group discussions, role play activities, peer mentoring and goal setting; secondly, physical recreational activities (including sailing, kayaking, stunt kite flying, fishing, hand cycling, sled hockey). There were two comparison groups, a wait-list control and control with no intervention. No further information was given about the control groups.

In Eyssen 2013, participants in the intervention group consulted occupational therapists (OTs) who had taken part in two-day course followed by booster session during data collection period and had access to the client-centred framework and tools. The course trained OTs in client-centred practice based upon the 'Canadian practice process framework' (Townsend and Polatajko, 2007). This framework incorporates stages A, B, C, E, F and G and possibly D of the review's PCP model (detailed in Box 4.1. chapter 4). Participants in the control group consulted OTs that carried out occupational therapy as usual. This included goal setting and providing therapy. OTs in the control group did not receive extra training and did not have access to the client-centred framework and tools. However, authors highlighted some concern that OTs are already trained in client-centred approaches, thus it might not be a useful comparison to assess effectiveness of a client-centred intervention.

6.1.3. Excluded studies

Six studies (7 references) were excluded for various reasons including interventions being education only with absence of collaborative goal setting or action planning; goals being predetermined and studies which did not test an intervention at all (see table 6.2. below).

Table 6.2. Studies excluded from the review during the full-text screen

Study	Intervention	Reason for exclusion
(Barlow et al., 2009)	Chronic Disease Self-Management Program	Group education. Goals are not created as a shared process between patient and coach.
(Barnard et al., 2010, Holliday et al., 2007)	Patients receive a workbook about goal setting followed by an interview with key worker about how to advocate goals	Goals are selected from a predetermined list
(Embrey, 2005)	Self-management programme	This is a literature review and questionnaire which audits MS patients' experiences of self-management programmes. The self-management programmes do not incorporate goal setting.
(Miller et al., 2011)	Web-based self-management	Participants are sent electronic prompts. There is no shared goal setting.
(Oeseburg and Abma, 2006)	NA	A case study of an individual's experiences of interactions with clinicians.
(Thorne et al., 2004b)	NA	Interview study exploring experiences of people with MS, not attached to an effectiveness study

6.2. Quality appraisal

As justified in chapter 5, Cochrane's risk of bias tool (Sterne et al., 2014) was used to assess the risk of bias in both studies included in the review and the CASP tool checklist (CASP, 2013) was used to guide quality assessment of accompanying qualitative studies.

6.2.1. Summary of risk of bias assessments

Overall, Block 2010 was assessed as having a high risk of bias according to criteria outlined in the Cochrane risk of bias tool (see table 6.3. below):

Table 6.3. Risk of Bias assessments for Block 2010

Bias	**Authors' judgements**	**Support for judgements**
Selection bias	High	Participants chose whether to receive the intervention or not. Some willing participants were excluded from the intervention group due to access difficulties and were moved to the comparison group (the number of which was not reported).
Performance bias	High	Blinding was not carried out as participants chose which group to take part in.
Attrition bias	High	There is mention of participants' withdrawal from study, but they do not report on the number of missing data.
Selective reporting bias	Low	All outcomes are reported as intended
Other risks of bias	High	Authors described difficulties in organising accessible venues for intervention activities and times when they could not arrange this, which suggests fidelity to the intervention protocol, may not have always been possible.

Overall, Eyssen et al. (2013) was assessed as having an unclear to low risk of bias according to criteria outlined in the Cochrane risk of bias tool. (See table 6.4. below):

Table 6.4: Risk of Bias assessments for Eyssen 2013

Bias	**Authors' judgements**	**Support for judgements**
Selection bias	Low to unclear	Participating institutions were first stratified for type of institution and then allocated to control or experimental groups via random number tables, which were performed using computer-generated sequence, before patient recruitment. When it came to recruitment of participants, authors do not explicitly report on the way in which this occurred. However, they do state that they could not be certain consecutive patients were recruited.
Performance bias	Low	Occupational therapists were instructed not to inform participants of the intervention they were allocated to. Test assistants who collected the data were blinded
Attrition bias	Low	Intention to treat (ITT) and per protocol (PP) analyses were carried out and presented in the paper.
Selective reporting bias	High	Data at each data collect point since baseline was reported. However, the data they presented was the difference in change between the comparison groups, rather than the change itself. Thus, the difference from baseline is unclear. Additionally some outcomes, such as cognitive functioning (the Perceived Deficits Questionnaire); upper extremity function (nine hole peg test); pain and disturbing sensations effect on everyday life; (pain effects scale); and cognitive fatigue and health related quality of life (Short-Form Health Survey Questionnaire (SF36)) in ITT analyses were not reported.
Other risks of bias	Unclear to high	There were significant differences between intervention and control groups in at baseline in institution (hospitals: rehabilitation centres = I: 73%:27% vs C: 36%:64%, p=0.00); academic centre (I: 73% vs C: 17% academic centre, p=0.00).

6.2.2. Selection bias

In the Block 2010 study, participants chose whether to receive the intervention or not. Some willing participants were excluded from the intervention group due to accessibility difficulties and were moved to the comparison group (the number of which

was not reported). This method of allocation is likely to result in more engaged and physically able patients being members of the experimental group and evidence of any positive effects may be explained by these baseline characteristics. The Eyssen 2013 study was a cluster RCT. Participating institutions were first stratified for type of institution and then allocated to control or experimental groups via random number tables, which were performed using computer-generated sequence, before patient recruitment. Authors do not explicitly report on the way in which recruitment occurred, but they do state that they could not be certain consecutive patients were recruited.

6.2.3. Performance bias

In Block 2010, blinding was not carried out as participants chose which group to take part in. In Eyssen 2013, occupational therapists (OTs) who delivered the intervention were not blinded. This was due to the nature of the intervention that involved training the OTs. However, they were instructed not to inform participants of the intervention they were allocated to. Test assistants who collected the data were blinded. Effectiveness of this blinding was assessed by asking test assistants if they knew which arm they were allocated to at the end of each assessment. The majority of responses 38/52 said that they did not know.

6.2.4. Attrition bias

Block 2010 does mention participants' withdrawal from study, but they do not report on the number of missing data. Missing data was accounted for in Eyssen 2013 as intention to treat (ITT) and per protocol (PP) analyses were carried out and presented.

6.2.5. Selective reporting bias

In Block 2010, all outcomes are reported as intended. In Eyssen 2013, the data they presented was the difference in change between the comparison groups, rather than the change itself. Thus, the difference from baseline is unclear. Additionally some outcomes, such as cognitive functioning (the Perceived Deficits Questionnaire); upper extremity function (nine hole peg test); pain and disturbing sensations effect on everyday life; (pain effects scale); and cognitive fatigue and health related quality of life (Short-Form Health Survey Questionnaire (SF36)) in ITT analyses were not reported. In order to gain further information, efforts were made to contact authors, but requests were not responded to.

6.2.6. Other potential sources of bias

In an accompanying ethnographic report, Block et al. (2006) note that the consent process may have discouraged people at high risk of alcohol and substance use from participating in the study. This discouragement of participation based on social or behavioural characteristics, means the study's external validity may be compromised. Authors also described difficulties in organising accessible venues for intervention activities and reported occasions when they could not be arranged. This suggests fidelity to the intervention protocol may not have always been possible.

In Eyssen 2013 there were significant differences between intervention and control groups at baseline in institution (hospitals: rehabilitation centres = I: 73%:27% vs C: 36%:64%, p=0.00); academic centre (I: 73% vs C: 17% academic centre, p=0.00).

Additionally, in terms of the content of the intervention in Eyssen 2013, it may be too similar to usual care carried out by OTs in the control group. Authors described usual

care as being client-centred and incorporating goal setting. The authors made efforts to demonstrate differences between groups by the intervention groups completing questions about their level of adherence to the Canadian Practice Process Framework (CPPF), the theoretical basis of the intervention. Both groups also carried out an evaluation of client-centred process "ECGP" (*sic*) questionnaire before and after data collection to assess client-centredness. The ECGP questionnaire is derived from CPPF which the intervention is based on. Therefore, this only assesses whether the specific intervention worked in the intended way, and may have overlooked other ways that the interactions may have been client-centred and incorporated stages of PCP.

6.3. Quality appraisal of qualitative findings

Block 2010 also used qualitative methods to examine how participants experienced the intervention. Qualitative elements were appraised according to the Critical Appraisal Skills Programme (CASP) Qualitative Research Checklist (2013).

6.3.1. Appropriateness of qualitative methods

Qualitative methods were used in order to record which goals had been selected, identify perceived barriers and enablers to goal attainment, and assess the ways in which the intervention helped. As the aim was to understand and interpret actions and experiences of participants, qualitative methods were appropriate.

6.3.2. Methods used

The methods used to collect qualitative data were; 1) an independent living assessment designed by the investigators to provide overall picture of a participant's life, this was a form with a predetermined list; 2) a personal activity log: to document goals, progress towards goal attainment, supports and obstacles encountered; 3) semi-structured and

open-ended interviews in order to assess participants' chosen goals, barriers and support to achieve goals. The appropriateness and quality of these tools is unclear, as there is little description provided.

6.3.3. Appropriateness of the recruitment strategy

Authors claim that interviews were carried out on a randomly selected subset of the intervention participants. It is standard practice in qualitative research not to use randomisation, but to recruit purposefully (Morse, 2010) which means selecting participants who are likely provide pertinent insights for research aims. Therefore, rather than a randomly selected subset, it may have been more appropriate to recruit interview participants based on specific characteristics to test and confirm emerging themes from ongoing analysis.

6.3.4. Data collection

It is unclear whether the setting of data collection and method of collection was appropriate due to lack of description and justification from authors. There is little detail provided to describe the qualitative data collection tools. It is stated that interviews were recorded, but we do not know the format used. Authors described coding interview text, but they do not explain if this was verbatim or subtext. It is unclear if sample size was appropriate as there is no explanation about how sample size was decided.

6.3.5. Relationship between researcher and participants

Authors did not report on the relationship between researchers and participants and its potential influence on data, therefore it is unclear whether this was considered.

6.3.6. Data analysis rigour

Rigorous data analysis in qualitative research can be ensured if more than one investigator analyses data and findings are compared and clarified. Examples from data should be presented to support and contradict arguments made and investigators should reflect upon potential influence they have on the analysis (CASP, 2013). Block 2010 report that four team members coded interview data independently and then compared identified themes. Quotes from interview data were used to support themes presented and some contradictory data was taken into account, therefore the data analysis process appears to be rigorous.

6.4. Effects of the interventions on outcomes

6.4.1. Improvement in health and wellbeing

Only Eyssen 2013 assessed changes in health and wellbeing. The study recorded measurements at baseline, 4 months and 8 months since introduction of the intervention. They used the disability and impact profile (DIP) a 39-item self-administered questionnaire to assess symptoms, mobility, self-care, social activities, communication and psychological status, with an additional two questions about fatigue. Impact on participation and autonomy (IPA) assessed impact of condition on perceived participation and autonomy of outdoor and indoor autonomy, family role, work and social relations.

Eyssen et al.'s (2013) presentation of data was unusual because it did not provide direct data from outcome measurements, but presented the difference in change between the control and intervention groups. Therefore, we are unaware of the direction and amount of change from baseline to data collection points in both arms. There were also

discrepancies in the outcome measures which were reported in the ITT analysis compared with the PP analysis. For example, the ITT analysis reported physical fatigue, whereas PP analysis reported both physical and cognitive fatigue. Quality of life, bodily pain and vitality were all reported in PP analysis but not in ITT analysis; and the none-hole peg test was missing from both analyses. In the ITT analysis, the study presented no significant differences in change between the control and intervention groups, apart from fatigue. At 4 months, the difference between changes in fatigue scores was presented as significant and in favour of the control group, however at 8 months the difference in change was in favour of the intervention group, although not significant. Thus, any advantageous effect of control over intervention is unclear and potentially irrelevant, because the amount that scores changed by is not recorded and any effects appeared to be short-lived.

6.4.2. Improvement in capabilities to self-manage

Only Block 2010, assessed the effects of the intervention on self-efficacy. This was measured through the General Perceived Self-efficacy Scale, which assesses participants' perceived sense of resourcefulness, ability to deal with unexpected events, and capacity to find solutions to problems using a 10 items Likert scale. However, Block 2010 presented the effects of the intervention in an overly favourable light. They begin their results section (p.747) by reporting a significant difference in favour of the intervention group compared with the control groups. However, authors omit that this difference is due to a decline in control group self-efficacy scores, rather than an increase in the intervention group's self-efficacy scores and also it is unclear to which data-point post intervention they are referring. At this point, it is worth noting an ethical concern that differences in self-efficacy scores between groups may be due to

participation in the study having a detrimental effect on control group participants. Self-efficacy scores worsened for the control group, so it is possible that the exclusion of willing participants from the intervention, due to access difficulties (as noted above), lessened perceptions of self-efficacy for the control group participants.

The authors then go on to describe improvement for self-efficacy scores for intervention and decline in the control group at 6 months. They report that at 6 months self-efficacy scores improved for those in the intervention group on average by 1.83 points (n=24) and declined on average by 2.86 points for the control group (n=16). However, this claim is inconsistent with sample size and self-efficacy scores presented in their table of summarising self-efficacy scores. The final paragraph reporting self-efficacy results notes briefly that there is no significant difference for participants in the intervention group at any of the assessment points. As the aim of the study is to assess the intervention effects on self-efficacy, this finding would ideally appear earlier and be given more prominence in this section.

The paper concludes that:-

"There was a statistically significant difference in the change in self-efficacy scores for intervention participants compared to non-participants. This indicated that participation in Project Shake-It-Up may have improved and/or prevented a decline in participants' perception of their ability to manage challenging situations and accomplish goals." (p.752)

However, any differences between the intervention and control group are arguably more likely to be due to the high risk of allocation bias, lack of blinding and participants being excluded from intervention group due to access difficulties or groups being self-selected. Thus, it is unsurprising that the participants who volunteered to take part in the intervention had higher self-efficacy scores, than those who chose not to or were

excluded due to difficulties accessing the intervention. Additionally, there is no evidence to support the suggestion that the intervention may have improved self-efficacy, as claimed in the statement above. The only outcome the evidence does suggest, is that taking part in the control group may have caused decline in self-efficacy.

6.4.3. Improvement in formal healthcare service use

Neither Block 2010 nor Eyssen 2013 reported data on use of formal healthcare services.

6.4.4. Attainment of personal goals

Neither Block 2010 nor Eyssen 2013 reported data on attainment of personal goals.

6.5. Why and how personalised care planning works

Participants in Block 2010 monitored their goals, and facilitators and obstacles encountered using Personal Activity Logs (PALs). PALs were developed by investigators and loosely structured, open-ended and qualitative. These were used to explore the experience of setting and aiming to achieve goals during PCP. Participants identified factors such as lack of transport; lack of access and social discrimination; physical and cognitive impairment; their personal perspective of ability/impairment; financial restrictions; and lack of support from family and friends as barriers to completion of goals. Factors that aided completion of goals included support groups; support from professionals and family members; technical adaptations to houses and vehicles; and perseverance. Some participants said that the intervention helped with enabling access to community services and the role-play element aided in self-advocacy. These findings suggest that PCP can be complex and its success seems to depend upon contextual factors that may limit or support processes.

6.6. Conclusions

6.6.1. Is personalised care planning effective for people with MS?

Based on the studies included in this systematic review, it is unclear whether personalised care planning is effective for people with MS. Problems identified in the quality appraisal of Block 2010, meant no useful conclusions based on this study could be made. The quality of Eyssen 2013 was better, however they reported differences in change rather than direct results and some outcome data was not reported, which made it difficult to make any useful conclusions. Also, the approach to occupational therapy consultations in the intervention and usual care groups appeared to be very similar in Eyssen 2013. This may explain why there appeared to be little difference in results between groups.

6.6.2. Why does personalised care planning for people with MS work or not work, and how does it work?

As neither study provided evidence of PCP interventions that were effective, it is not possible to examine processes that aid or inhibit PCP planning based upon these studies.

6.7. Discussion

There were few studies that reached the inclusion criteria, confirming that PCP for people with MS is an unexplored area. To reiterate, of the studies included in the Cochrane Review (see chapter 4) the majority were for people with diabetes (12/19) and none were for people with MS or any other neurodegenerative conditions.

Comprehensive interventions integrated into routine care were particularly successful in the Cochrane review (see chapter 4). The intervention in Block 2010 was

comprehensive and covered many stages of the PCP model. However, reports from the accompanying ethnographic study (Block et al., 2006) noted problems in organising and accessing locations suggesting it may not have been performed as intended, and its implementation was limited due to high level of resources required. Although not strong evidence, it does support the Cochrane review finding that interventions that were incorporated into routine practice were most successful (Coulter et al., 2015) and also reinforces the argument that organisational features and resource availability play an important role in supporting PCP (Wagner, 1998, Batalden et al., 2015). Ethical concern that self-efficacy scores worsened during Block 2010 were highlighted in this systematic review. If interventions are inaccessible and impractical for people with MS (as Block 2010 appeared to be) then it is possible this may have detrimental effects of feelings of self-efficacy. Therefore, aspects such as how interventions are embedded into routines, the organisational support and practicalities are likely to be vital when designing PCP interventions which are safe and effective.

In an accompanying paper, Eyssen et al.'s (2014) secondary analysis demonstrated how the intervention group took on average 11.4 days longer to formulate their first goal, compared with the control group. It is possible that the process of PCP takes longer, therefore longitudinal studies are needed to assess the effectiveness of such approaches. Eyssen 2013 used an occupational therapy setting and usual care seemed congruent with PCP ideals and Block 2010 was not set within a healthcare setting. It would be useful to consider PCP for people with MS within a broader general healthcare setting. MS is a complex condition and healthcare services are broad and vary between individuals. Consultations with GPs or MS nurses may provide a central point of contact to consider varied factors and coordination of care.

Section three
Findings from interviews with people with Multiple Sclerosis

Chapter 7

How do Participants Discuss Uncertainties Related to Multiple Sclerosis?

7.1. Chapter summary

As explored in chapter 2, MS is a highly heterogeneous and unpredictable condition. Illness trajectories, severity, potential symptoms and complications vary widely. Its exact aetiology and pathogenesis are unknown, and although widely understood as an autoimmune response, theories exist which contradict this. Currently, there is no cure, and evidence suggests that treatments available have limited effectiveness, do not slow long-term progression and may have potentially severe side effects. Such factors create a context of uncertainty for people with MS.

This context of uncertainty was an area that participants frequently discussed during interviews. During analysis three key themes emerged from the interview data. These themes were 'existential uncertainties', (uncertainties about the way in which one will exist); 'embodied uncertainties' (uncertainties in defining or categorising illness experiences); and 'intervention uncertainties' (uncertainties about intervention effects). Each of these themes and their implications are examined throughout this chapter.

7.2. Existential uncertainties

When people develop a chronic condition, their identity and previous assumptions about their body, life and future are reconsidered due to the potential limitations their chronic condition may bring (Bury, 1982, Charmaz, 1983). Here, I have referred to the

uncertainty surrounding this phenomenon as 'existential uncertainty'. Adamson (1997, p.134) described it as follows:

"[E]xistential uncertainty is that form of uncertainty which is experienced privately by the individual patient upon the realisation that the future of his or her mind, body, and self is in jeopardy."

Broadly speaking MS follows a pattern where there are periods of relapse (exacerbation of symptoms) and remission (absence of symptoms), alongside long-term impairment (Rae-Grant et al., 2011). However, the heterogeneous nature of MS means that rate, pattern, severity and type of symptoms vary, and thus an individual's illness trajectory is unpredictable. Many participants found the existential uncertainty they experienced undesirable and difficult to deal with. Adam, Hannah and Kamila discussed the difficulties they experienced when dealing with an unpredictable illness trajectory:

> *Adam:*
>
> *"I remember thinking at the time of diagnosis I'd like to, you know I'd like to know how many good years I'm gonna have so I can make some decisions. Is it gonna be three good years and then downhill? Five? Ten? I'd like to get away with it. No, cos no-one can give you that sort of picture. No-one will. [Um] So you're sort of left then to think about the rest of your life amidst that kind of uncertainty […] it does always lurk in the back of your mind quite what the future's going to be. And I think just the uncertainty around a condition like this [um] is one of the hardest things"*
>
> *[Adam (11), male, 36 years old]*

Hannah:

"It was, somebody at work, there's this Irish guy who talking about this Damocles, the Sword of Damocles and I thought, yeah that's how I feel, 'cos you don't know what's going to happen next. Because soon after diagnosis all these different things, I lost my sight in one eye for about two weeks, and then the other one for another two weeks. And then various symptoms started popping up for two weeks and going away.

Right.

But if I'd expected those I would have felt more in control."

[Hannah (20), female, 49 years old]

Kamila: "You know that's the whole thing about MS you don't know what's going to happen and that's why it's so frustrating. […] But I had no idea what it meant and [um] to be honest I think the hardest thing for me was to accept the fact that I have something that I can't do anything about. […] it doesn't matter what I do it's always going to be there, and [um] I can't change it."

[Kamila (21), female, 29 years old]

"The Sword of Damocles" referred to by Hannah is a fable in which a sword hanging by a horsehair is suspended over a king's throne. It depicts constant fear, precariousness and anticipation of danger. Phrases such as "lurk in the back of your mind" and "always going to be there" reflect the intrusiveness and consuming nature of existential uncertainty. Another participant James, described this as a "consistent digging torture". Toombs (2002) described an 'inescapable embodiment' experienced by people with MS due to its potential to affect one's whole body. She argued this 'inescapable embodiment' is exacerbated further by an unpredictable illness trajectory.

7.2.1. Envisaging worst case scenarios

When considering their future with MS, participants described how they imagined the potential symptoms they may experience. They reflected upon how initially they feared

that their MS would be severe and imagined worst-case scenarios. James and Ulrike remembered how they envisaged severe symptoms around the time of diagnosis:

James:

"And I've been told I've got something that's never going to go away, that's just, it is what it is and I, I was fearful of the, the worst case scenarios [um] at the time I was fearful that I could wake up one day and I'd be blind. I was fearful that I would wake up one day and I wouldn't be able to walk, completely, I would be in a wheelchair […] There's, it's almost a consistent sort of digging torture. And it took me a long time to get over that fear of how am I going to wake up tomorrow?"

[James (03), male, 33 years old]

Ulrike:

"You think you're going to wake up one morning paralysed and all this is sort of going on in your head, or in a wheelchair. […] And I still think it's very very serious. I don't, I haven't sort of [um] come to regard it as, as a lesser condition. But I, I think I no longer feel that I'm going to wake up one morning with no legs available to me. I don't think that any more, which is, I did think.

[Ulrike(14), female, 62 years old]

Both James and Ulrike recalled how they used to imagine that they would "wake up one day" to find they had developed severe symptoms. This anticipation of severe symptoms suddenly appearing whilst they are sleeping not only reflects their understanding of MS as unpredictable, but could also suggest a lack of control in that they anticipate their appearance when they are not conscious.

7.2.2. Biographical abruption

In Bury's widely cited concept of 'biographical disruption', he described chronic illness as "a major kind of disruptive experience" in people's lives. Its presence is unexpected and draws attention to bodily states and actions to be taken (Bury, 1982). Everything

understood about oneself comes into question and the experience of chronic illness may change one's perception of their identity or sense of self (Charmaz, 1983). Developing Bury's concept of 'biographical disruption' further, Locock et al. (2009) introduced the concept of 'biographical abruption' to describe experiences of people with motor neurone disease (MND). They distinguished these concepts as follows:

> "Whereas 'disruption' implies disturbance and unwelcome change, 'abruption' is intended to convey a sudden ending, literally a 'breaking off'. It encapsulated a commonly reported feeling that the diagnosis was a 'death sentence', that life was in effect already over and that individuals had been denied a future." (Locock et al., 2009, p.1047)

Locock et al. described MND as a condition that is both terminal and chronic. Due to this status, they argued that 'abruption' differed to 'disruption'. However, although MS is not considered a terminal illness, some participants described perceptions which were similar to 'abruption'. In the examples below, Anna and Ulrike recalled their thoughts around the time of diagnosis about how MS may affect their future:

> *Anna:*
>
> *"When something comes along as devastating as MS completely obliterates life as you knew it, […] Everything you knew and took for granted is, is gone. […] But I had, oh I can't stress enough without sounding like a tragic single parent but it, I was up against a time limit, I knew I was ill. I knew I was going to get worse, and I knew I was that close to ending up in a wheelchair very quickly or doing myself a serious injury."*
>
> *[Anna (07), female, 39 years old]*

Ulrike:

"I just thought, it was like a sort of [um] like a whirlwind happening over you. You know you've been taken over by this, this thing, this that had invaded your body and, and I really thought was going to just sort of strip me of everything.

***Mm**.*

I, I couldn't see a way; I couldn't see how I could work. I could, I thought, "How can I stay in this house? Do I want to move? Do I? If I move, go somewhere where nobody knows me," I was really at that stage."

[Ulrike (14), female, 62 years old]

Although participants did not discuss literal death as Locock et al.'s participants did, phrases such as "obliterates life as you knew it"; "a whirlwind happening over you"; and "strip me of everything," suggest people with MS can envisage a 'sudden ending' or 'breaking off', rather than a 'disturbance' or 'unwelcome change' to their lives (as referred to in Locock et al.'s distinction). To further this point, Bury (1982) described chronic illness as something more gradual than how participants in this study seemed to envisage their futures:

"One of the most important features of chronic illness is its insidious onset. Non-communicable diseases do not "break-out" they "creep-up"" (Bury, 1982, p.170)

It is important to note that this 'abruption' reflects the *perceived* (rather than *experienced*) gravity of MS for some at the time of diagnosis. This could be argued for all reports of imagined biographical 'disruption' and/or 'abruption', but the gulf between perception and reality seemed to be particularly marked for some participants. This is discussed further in the following section.

7.2.3. Reconceptualising MS effects and 'biographical restoration'

The presence of uncertainty may be more desirable in certain contexts. Some have argued that, the longer people with chronic conditions live with 'continual uncertainty', the more favourable this uncertainty may be perceived. This is because uncertainty can provide hope that one will not be severely affected, which is preferable to more certain severe prognoses (Mishel, 1990). Strickland et al.'s (2017) study exploring perceptions of those newly diagnosed with MS described how participants felt their sense of 'self' was irreversibly changed. However, all their participants were diagnosed within the previous 6 months, thus their perception that change was *irreversible* may alter with time and reflect the impact of receiving a diagnosis.

On the contrary to Strickland et al.'s findings, many participants found their expectations changed as they believed their MS trajectory would be more favourable and effects on their life would be less damaging than previously imagined. This reframing was shaped by their own personal experiences of (a milder) MS and accessing different types of advice and information (further explored in chapter 8). Although previously fearful of potential symptoms, Katy's experiences of MS so far have helped her to think more positively:

> *"I'm not in any pain; it doesn't stop me doing anything it's just not nice having it. And I'd say I'm, you know I'm a best case scenario as it were, knocking on wood as I speak but, and maybe sometimes I think well there's a whole gamut of what could happen but the likelihood is that the worst won't happen, you know, you're more likely to be run over by a bus which is probably true.*
>
> *[Katy (03), female, 41 years old]*

Although Katy is aware of the possibility of various symptoms, her so far mild experience of MS and lack of disruption to her life has enabled her to reimagine her MS

as remaining relatively mild. Ulrike described how she found positive personal examples helpful:

> *"[An event] was called "Living Successfully with MS," and they had [er] somebody who'd had it for 30 years came and told her life story.*
>
> ***Oh really?***
>
> *And of course everybody thought they were going to see somebody appear in a wheelchair, and she just bounced up. She'd come on her motorbike in her leathers, you know. Brilliant.*
>
> *[…] And you can read the statistics and I think the last one I read was it's now under 50 percent of people will need a wheelchair."*
>
> *[Ulrike (14), female, 62 years old]*

These examples contrast with the initial perception of 'biographical abruption', as participants who described previously feeling distressed whilst imagining worst-case scenarios and an ending to their imagined futures, reconceptualised their MS and considered the potential for milder trajectories. Building on Bury's core concept of 'biographical disruption', various theorists (including Bury) have explored concepts of 'biographical repair' for people with chronic conditions who reconstruct biographies or narratives and incorporate their condition to create a new normality, or 'biographical flow' where chronic illness is expected and considered a normal aspect in one's life course (Williams, 2000, Williams, 1984, Radley and Green, 1987, Bury, 1987, Charmaz, 1985). Cases of 'repair' or 'flow' do still involve existential change (whether anticipated or not) to pre-diagnosis biographies.

However, in the case of MS, following initial perceptions of 'abruption' there appears to be some 'restoration' of pre-diagnosis biographies. The interview data suggests that existential uncertainty experienced by people with MS, provides a setting where

perceptions of a severe trajectory appears to lead to perceived 'biographical abruption' typically around the time of diagnosis. However with experience and information, initial perceptions of severe trajectories are later reconceptualised as much milder and, rather than incorporating the effects of MS into a 'repaired' biography, or considering MS effects as an expected part of biographical 'flow', some people with MS seemed to 'restore' elements of their previous biography. Put differently, rather than *biographies being reconceptualised* to incorporate MS symptoms, their *perceived illness trajectories are reconceptualised*, which in turn enables 'restoration' of previous imagined futures.

In the extract above, Katy's perception seemed to lack 'biographical disruption' when she said "I'm not in any pain; it doesn't stop me doing anything" and she uses the analogy that she is "more likely to be run over by a bus" than be severely affected by MS. People often use this analogy to demonstrate a low everyday risk, Katy's use suggests she perceived the risk of harm caused by MS as being low and her experience of MS as not being far removed from 'normality'. In Ulrike's extract, she was impressed to see someone with MS who appeared well and to be continuing with everyday activities (such as riding a motorbike). She contrasted this to her expectation, which was to see someone more severely affected, reflecting imagined MS illness trajectories and other types of information available (further explored in chapter 8). She also referred to statistics about the likelihood of using a wheelchair as being lower and contrary to her previous understandings. Ulrike also was empowered by her neurologist who helped her to rethink MS and its effects on her life:

" "Look," he said, "I am not telling you, you have a life threatening illness," he said, "You have a chronic condition and it's up to you to manage it." And he sat back and do you know there was, those words were I think the beginning of the recovery.

Oh really?

Yeah.

Yeah.

Because you know it, the thing is all encompassing. Your limbs don't work. You think you're going to wake up one morning paralysed and all this is sort of going on in your head, or in a wheelchair.

Yeah.

And then somebody who is obviously completely together and knows what he's talking about says, "You've got a chronic condition. You know, manage it. Do it, you know, sort it out." He says, "We'll help you, you know." […] And that was, you know, that was the beginning of the, of the change really from panic to you know to managing.

Yeah, yeah. Yeah so

That was absolutely life changing."

[Ulrike (14), female, 62 years old]

It appears that when Ulrike refers to her recovery she is not referring to recovery from physical effects of MS, but recovery as gaining control over its potential threat to her life; recovery is from the perceived 'biographical abruption' as she reconceptualised her condition and she went from "panic to managing". She positions the notion of a severe trajectory as "going on in your head", which reflects it being her perception rather than her experience. On the one hand, references to managing her MS, suggests Ulrike may be carrying out something more similar to biographical 'repair' rather than 'restoration' to her previous biographical status. The key factor is that, although she may be incorporating *some existential change* due to MS, she is not incorporating her previous perception of a severe illness trajectory (or responding to the 'biographical abruption')

but perceiving a different, more manageable version of MS. The extent to which participants 'repaired' or 'restored' biographies varied between individuals. The fluctuating nature of MS means its trajectory may be asymptomatic for long periods, there may be episodes of symptoms (relapses) or people can live with symptoms that are more permanent on a daily basis. Such factors are likely to relate to the extent to which biographies are 'restored' or 'repaired'.

Although narratives of perceived 'abruption' followed by 'restoration' was a common pattern identified during analysis, it was not present in all participants' accounts and appeared to be less common in those more severely affected by MS. This is perhaps unsurprising as the larger the impact MS has, the more likely it is to redefine or disturb biographies. Mozo-Dutton et al.'s (2012) study described how, as people with MS experienced ill health, a "self-body dualism" (p.1212) emerged as they found their body could not be relied on in the same way as it had previously. The possibility of "self-body dualism" for people with MS may appear to contradict the finding that many people with MS experience 'restoration' of previous biographies. However, I would argue that in reality such contradictions reflect the variability of experiences of MS. At this point, I would also like to note the potential bias within the sample, as those who present examples of 'restoration' or 'repair' in their biographies may be more willing and able to discuss their experiences and volunteer to be interviewed.

In summary, existential uncertainty provides a setting where people with MS may initially envisage a severe version MS illness trajectory and perceive 'biographical abruption', but this perception seems inappropriate (or exaggerated) for many people with MS. Again, existential uncertainty, alongside accessing information and experiencing (a milder than imagined) MS allows an opportunity for 'restoration' (or

'repair') of previous identities. In turn, their MS trajectory is reconceptualised as milder than initially expected and closer to what was imagined before they were diagnosed. This theme is directly related to quality of information accessed and the implications of this, which is examined further in chapter 8.

7.2.4. Life choices

Participants' accounts suggest that the presence of existential uncertainty drew their attention to life choices. They reported various considerations such as whether their ability to work would change; whether they should change jobs; how their social life might change; whether they can continue with hobbies; when and if they should have children; where they should live in the future; who would provide care for them if needed; and whether to make adaptations to their home. Certain life decisions such as where to live, career choices and children, appeared to be particularly salient for younger participants who were in their 20s and 30s. Most people with MS are diagnosed between ages of 20 and 40 years old (Ontaneda et al., 2012, Dua et al., 2008) which may be considered a particularly transient time. Those closer to 20 may be less likely to have established life choices, compared to those nearer to 40. Anna said she was glad she had already made many life decisions before being diagnosed:

"MS is so cruel. You know it comes when you're deciding about kids and you're deciding about your career, deciding whether to travel the world. So in hindsight I'm so glad that I got that out of my system before everything, [developing MS]

***Yeah**.*

You know. But I've got friends that haven't and are stuck, you know, not being able to decide either way what they want to do.

***Yeah**.*

So I feel very fortunate,

It's all these life choices isn't it? At that time of life you're making…

Yeah. I've been married, I've been divorced. I've had the kid.

Right yeah.

You know I've lived abroad for four years. You know I've done everything I set out to do. But you know some people haven't quite got that, hadn't done all those things yet. It's a shame. It's a horrible disease."

[Anna (07), female, 39 years old]

Anna said she felt "fortunate" that she had faced many life choices before developing MS, so she did not have the worry of considering MS during such decisions. This reflects the perceived weight of existential uncertainty when making life decisions. Some participants said the uncertain future encouraged them to do things sooner than they might have without an MS diagnosis. For example, Adam said he is more likely to compete in running events since being diagnosed:

"I got back into it [running], did quite a lot of it and would enter races and do things that previously I'd have been, "Oh do you know what, I'm not quite ready for this race. I'll probably do quite badly in it or injure myself or [um] I'm better off waiting for you know the next one, or next year, or one at the end of the season." Or whatever it was. So it's things that I might have put off in the past I didn't. […] 'cos I was just pleased to still, pleased to be able to do it so it didn't matter whether I [um] did times that I thought were, were respectable or not. It was just good to do it.

[Adam (11), male, 36 years old]

Adam felt encouraged to take part in running events, as he envisioned a future where his MS would mean that he would be unable to. However, he appeared more cautious about his career:

"I mean I'm sort of someone who is not dreadfully unhappy in his job, but has never quite fitted in, never quite settled into it. That's, even before diagnosis I was thinking do you know what, is this really is this really you? Should you be doing something else? Should you be trying something else? Going back studying again, whatever it is

Yeah

that you want to do [um] and, which is, which is true. I'd love to do that but there's a part of me that thinks well you know, you might not be able to work for that much longer, you're kind of safe where you are. It's, you know, it's paying okay, [um] it means you can stay in [city], you know, you're sort of, you're well supported there. There's, they'll look after you and if, if I move I might not be sort of as well looked after. [Um] All these sorts of things [um] have kind of stopped me from acting in that sort of sphere. […] I mean I've been much more cautious and conservative and, [um] you know I'll sit tight in a job where okay it may not be my dream job but I'm quite, you know they're really around this sort of thing. I've been treated quite well, they're very good for flexible working and that kind of stuff, it just feels secure. [Er] Security suddenly matters"

[Adam (11), male, 36 years old]

According to Adam's account, existential uncertainty altered his priorities. In contrast to his perception before diagnosis, he now considers his *capability* to take part in running events more important than performing to a certain standard, which encourages him to

take part whilst he still can. In regards to his career choices, flexibility and security have become more important considerations than previous ideas about finding a career that is more enjoyable or one that suits him better.

Several participants discussed how their MS diagnosis affected their decisions about having children. Joanne and Laura both discussed choosing to have children sooner than they would have if they did not have MS. At the time of the interview (one year after her diagnosis) Kamila was considering whether and when to have children. Her concern was that her child could potentially become a carer at a young age. Within a year of receiving her diagnosis, Joanne had got married and had a child:

> *"All that I thought about then [soon after diagnosis] was wheelchairs and stuff.*
>
> ***Right**.*
>
> *I thought oh my God, we have to get married now quickly 'cos I don't want to be wheeled up the aisle, I want to walk up it.*
>
> ***Right you, yeah, yeah.***
>
> *Everything crosses your mind and I didn't really dwell on it but it was something that did cross my mind and I just thought I know I definitely want to just get married […]*
>
> *Aye. I just I would just love to tell people that like you know if you get diagnosed with it young like it's not the end of the world. […] But like [um] I would just say don't rush into like marriage, children all that there like, you know it's not that I rushed into, we were engaged anyway before I was diagnosed but like [um] we got married, then we had our other baby, but that, not that it felt a bit rushed 'cos we would have always have done it, but oh it, just made us do it a lot sooner than we probably would have."*
>
> *[Joanne (18), female, 28 years old]*

In hindsight, Joanne believed that she did not need to marry or have children as soon as she initially thought. Linking to the previous section, this extract demonstrates how

initial assumptions that one's progression will be severe can shape life decisions. Such implications may be reviewed as unwarranted in cases where people with MS reconceptualise their prognosis as milder than initially assumed or as they experience biographical 'restoration' or 'repair'.

7.2.5. Daily uncertainty

For some participants their condition fluctuated on a daily basis. This meant that their condition was at the forefront of their mind every day and their plans often changed in response to them. This is demonstrated in Oliver's and Ian's accounts below:

> *Wife: "We might be going out for our lunch a bit later, [um] but it, it'll depend how*
>
> *Oliver: On how I feel.*
>
> *Wife: On how [Oliver] feels. If he feels, because although we could take the wheelchair, we're not going to, 'cos, but it's a little walk where we going to, just not far. Say from here to the bungalow over there. And it's if you feel; feel up to it, walking there. And then when we've sat down for a bit, if you can come back. So that'll sort of, we'll decide nearer the time whether we're going to go or not."*
>
> *[Oliver (12), male, 58 years old]*

Ian:

"The only thing I can say is like you know ever since I relapsed it's just been pain, chronic pain but random, where it hits is random. Every day is different.

***Yeah**.*

And some days there's no pain and some days there's lots of it. [Um] Some days I can walk round the town quite easily, other days I can walk around the town and I have to use a stick. [Um] I've got a wheelchair and I've used it twice in two years […] With MS it's an, every day is different. Yeah we know that but I think I'm the [er] example of how bad 'every day is different' can be.

***Right**.*

Because in my little slice of the cake of what my MS is like, it is literally every day is different but overwhelmingly it's the pain that's different every day."

[Ian (16), male, 42 years old]

The daily fluctuations mean that, for some, their condition is constantly considered. Some participants reported feeling concerned about plans they had made, and some avoided making plans altogether. After being unwell at her niece's confirmation Joanne had felt very concerned that she might be unwell at her sister's wedding:

"I swear I was dreading my sister's wedding after it because I had such a bad experience. […] And you just, it was like nearly like a mental fatigue then just came over me and I was just like "Oh my God," and I knew we had to go out for dinner and get the pictures taken, and I was like, "Oh my God," and then I had, I was worrying about the wedding the next week and I was kind of like "Oh my God," and I just thought, "Oh." But I pushed myself and I went, didn't make it, ended up having to leave half way through the starter, it was so embarrassing.

[…] I want them [my family] to think I'm coping. I am coping, but I don't want to, I don't want to worry them either. But when I had to leave the first communion dinner, came back here and I thought, "Oh my God, my sister gets married now in five or six days. This is going to be terrible" 'cos I was so worried that the same thing would happen then at the wedding."

[Joanne (18), female, 28 years old]

This extract exemplifies a point made by Bury that "maintaining normal appearances in a social gathering […] have become deliberately conscious activities, and thus frustrating" (Bury, 1982, p.175-6). He elaborated on this and described how such frustrations mean for some "the effort simply does not seem worth it. The erstwhile taken-for-granted world of everyday life becomes a burden of conscious and deliberate action" (Bury, 1982, p.176). Bury's observation is evident in Mary's account, as she no longer made plans, due to concerns that she may not be able to take part:

> *"So you can't have plans and actions. It's the same with you know with going out. [Um] I used to try and make plans to see people or to go out or but I have to cancel them so often that I get embarrassed so in the end I just don't do it. […]*
>
> *My son found that he was doing one [a talk] in [town], not too far away. And he said, "You know, do you think you could manage it?" Oh and I so wanted to do it. And [er] I spent about two weeks every night thinking, "now could I have gone tonight?" And sometimes I'd think well it's not necessarily the getting there, it's the getting back and feeling like, you know on the way home that you're just going to die in the car. It's just, and I just thought no, I don't want to waste his money by ending up not going and [um], but sometimes you know you get things like that and you think "oh I wish I could do it". And my husband takes my mother-in-law shopping every week, and she's always saying "Oh I wish [Mary] could still come." And I think "Oh I'd just love to go."*
>
> ***Yeah.***
>
> *But I just, know I you know I'm going to ruin it for everybody […] it just gets to the point that it's just not worth bothering."*
>
> *[Mary (05), female, 61 years old]*

In addition to the MS-caused physical difficulties of taking part in social events, the extracts above also demonstrate the implications of daily uncertainties on one's sense of self. Potential for embarrassment felt by people with unpredictable chronic conditions can affect their perceptions of 'self' as they no longer take part in events (either voluntarily or involuntarily) in ways they used to, or others expect them to. People's worlds and identities become shaped and restricted by their illness (Charmaz, 1983).

Although Mary wanted to take part in social events, she disengaged from them due to the embarrassment of cancelling plans and concerns she will "ruin it for everybody". The prospect of being unwell at a family event caused Joanne anxiety due to concern about how other family members would perceive her.

7.3. Embodied uncertainties

MS causes disruption and damage to the central nervous system (CNS), thus symptoms caused by MS are as varied as the functions which the CNS may affect (Mohr and Cox, 2001). Most commonly reported symptoms include motor dysfunction, sensory problems, fatigue, visual disturbances, balance problems, incontinence, pain, cognitive dysfunction, behavioural problems and sexual dysfunction (Dua et al., 2008, Mohr and Cox, 2001). During diagnosis, MRI scans (alongside clinical presentations) provide evidence of disease activity. However, the way in which data obtained from MRI scans directly relates to symptoms experienced is unknown (ABN, 2009). Therefore symptoms caused by MS are varied and mostly detected and assessed through personal experiences.

7.3.1. Causes of problems

The variety of potential symptoms people with MS may experience – alongside the absence of instrumental objective measures that directly indicate disease activity – means there is often confusion over whether the problem experienced is caused by MS, or something else. There were many examples of this confusion throughout participants' accounts, two of which are presented below:

Paul:

"I don't know whether it's got anything to do with bloody MS but particular, sometimes I can really lose my temper with inanimate objects.

Right.

It really, really weird [um] in that just you know if, if something falls off the shelf or something like that it will really annoy me. Or you know when you're trying to get something off the shelf, that you're trying to really make sure that you don't knock an item off the shelf, but you knock it off and it's Aargh, I get really annoyed with it. [..] you ask a question, "Well could it be the MS? Could it be something else?"

Yeah.

And that's, and that's, that is a question you have with MS every time.

Right.

Is, is it MS? Is it something else? So you can't, unfortunately very rarely you can answer that. The tingling down my arm I can pretty much guess it's the MS because you know I haven't, I don't feel like I've trapped a nerve in my shoulder or anything like that. But you know it, it could still be something else"

[Paul (04), male, 27 years old]

Sadie:

"But I'm sure my eyes have got worse. My eyes have got really bad. I don't know if that's due, I know that was the first sign, was my eyes but,

Right yeah.

Again see you don't know is it just old age that they're deteriorating or is it,

Yeah.

you just never know."

[Sadie (17), female, 50 years old]

Even in the two examples above, the heterogeneity of problems (mood change, sensory problems and visual disturbances) that participants believe may be due to MS is clear. These examples demonstrate the confusion that people with MS can face when

assessing whether problems are caused by MS or something else. Even symptoms that appear to be more certainly attributable to MS, such as Paul's "tingling down my arm" are questioned to some extent. The act of attributing (or misattributing) problems to MS may cause anxiety because the presence of (what are believed to be) MS-caused symptoms act as signs of 'an MS' which may be progressing (Jopson and Moss-Morris, 2003). This embodied uncertainty may also mean that other causes go undetected and untreated, or people may worry about other potential causes being missed.

'Embodied uncertainty'– sometimes coupled with lack of confidence in healthcare professionals' opinions – caused some participants to question the validity of their diagnosis:

> *Kamila:*
>
> *"I am still thinking what if they made a mistake? You know, what if I haven't got MS? Which gets dismissed straightaway [um] I did question it actually and [um] I went to see a GP because I thought 'okay well I think I would like someone else's opinion' because of my whole experience with this neurologist and this MS nurse I didn't feel really confident, and I thought Okay, I think I would like a second opinion and I went to a GP"*
>
> *[Kamila (21), female, 29 years old]*

> *Donna:*
>
> *"It's only funnily enough in more recent times where I've just very occasionally I've thought, "I wonder if that [MS diagnosis] is true? I wonder if that is really what is going on?"*
>
> ***Really? Yeah?***
>
> *I, just occasionally you do that thing, maybe they're wrong"*
>
> *[Donna (06), female, 49 years old]*

Mary: "I've heard an awful lot of people with MS say the same thing, that it's [um] you know you begin to wonder if it's all you know in your head."

[Mary (05), female, 61 years old]

Participants questioning their very diagnosis indicate the strength of influence that embodied uncertainties can have. It may also suggest that the MS they are experiencing is different to preconceived ideas of what the experience of having MS would be like.

Some participants expressed a desire to have tests or scans to confirm the cause of their symptoms or to record any asymptomatic disease activity. Ulrike discussed her desire to have regular MRI scans, as she believed the symptoms she experienced were only "the tip of the iceberg", suggesting she was conscious and wanted to see evidence of potential asymptomatic disease activity. Donna also wanted to see her MRI scans:

"I want to see those MRIs. I want to see my brain.

Mm. Yeah.

Because I want it to be concrete. I want to be able to go, "I know I've got a lesion there, there, there, there, there. I don't know what it means, but I, you know I want to be able to visualise it properly"

[Donna (06), female, 49 years old]

Donna's belief that MRI scan readings would be more "concrete" and allow her to "visualise it properly", reflects her perception that understandings based on her experience are unclear and less valid. Sadie said she would like to have a scan that she hoped would confirm whether her back pain is caused by MS:

"I just wish they would do, I don't know maybe a scan on my back or

Yeah

Maybe, just do something different just so I know that it's not anything else"

[Sadie (17), female, 50 years old]

When discussing conceptualisations of illness, Nettleton (2006b) described how the biomedical model (which emerged in 18th century) locates disease within the anatomy, and technologies such as MRI scans reinforce this rhetoric. She argued that such perceptions:-

"fabricate "genuine disease" as opposed to the "experience of illness". This also means those whose symptoms are unexplained may become more 'at sea'" (Nettleton, 2006b, p.84).

Both Donna and Sadie appeared to believe that MRI scans provide more valid or certain information about their MS. Ryan and Raisanen's (2012) research into experiences of young people with epilepsy found that fears can be exacerbated when faced with evidence of their seizures, such as scans, filmed recordings, or eyewitness accounts, in addition to their embodied experience. Similar to people with MS, the presence of additional evidence of disease activity seems to make seizures more 'real' for people with epilepsy. Although common practice in the USA and Europe, regular MRI scans post diagnosis are generally not performed on MS patients in the UK (ABN, 2009). Some MS patients may prefer regular MRI scans but, if they were introduced in the UK, it would be important to consider the potential emotional impacts of seeing such information alongside the practical purposes, if any, this would serve.

7.3.2. Embodied uncertainty in consultation settings

Participants also described concerns about the potential that, and recounted scenarios when they believed, healthcare professionals had misattributed the causes of the problem presented. Healthcare professionals hold a powerful position when interpreting patients' presentations, i.e. "doctors not only decide who has what disease, but also what counts as the disease" (Scambler and Scambler, 2010, p.2). Thus their judgements have direct implications of course of action followed. Some participants felt that healthcare professionals inappropriately dismissed their symptoms as either caused or not caused by MS. According to participants' accounts, this sometimes caused worry or inhibited their access to treatment. At the time of interview, Sadie was concerned that her GP assumed that her back pain was caused by MS, when it could be something else:

> *""Is it, have I done my back in?" When I spoke to my doctor the other day she signed me off and she's put 'MS related', she says it's because the muscles are weakening. But you just don't know. Is it that? Is it, could it be something different? And I said to [my partner] yesterday, you know they're saying that you know it's my back and it's happened again because the muscles are weakening, but what if it's something completely different? 'Cos nobody does any tests to find out if it's something completely different […] So I tried to like say maybe it's something else, 'cos it's happened again so soon, but she was sort of straight back with the MS."*
>
> *[Sadie (17), female, 50 years old]*

This extract demonstrates the worry people may feel when their concerns (in this case potential for other causes) appear to be overlooked by healthcare professionals (further explored in chapter 9). Chris disagreed with his previous neurologist's assessment that his migraines were not a symptom of MS. However, his current neurologist agreed that they were:

"For the sort of first [er] eight months or so of [er] after sort of first symptoms I had some blinding headaches every day and I went back to the neurologist a couple of times about that. He said that there was no relationship between [er] migraines and headaches and MS.

[…]I saw him [new neurologist] and he said, "Well actually from what you've described about your first symptoms and then the, all the headaches and then the sort of double vision, I would have classed those as relapses and would have put you on medication. But because you've now had a gap of nearly two years from that,

Right.

[Er] You therefore still don't qualify for any medication."

[Chris (08), male, 38 years old]

Initiation of disease modifying drugs (DMDs) depends on the pattern of symptoms experienced. After Chris's symptoms, which would have justified commencement of DMDs, he happened to have a period without symptoms. This meant that by the time his current neurologist had assessed the previous symptoms as MS caused, his recent period of good health meant he was currently not eligible. Chris's account demonstrates the potential for healthcare professionals' assessments of causes to differ due to uncertainties in the way in which MS manifests. As healthcare professionals act as gatekeepers, such discrepancies may limit or enable access to treatment. Melissa had a similar experience; however her GP assumed her problem was *caused by MS*, thus limiting her access to correct diagnosis and treatment of a fractured femur:

"I've been having severe muscle spasms and nerve pain in my right side, which is my, that's the side that's affected [um] and I'd been having these for about a year, and it was getting worse and worse. And everybody kept putting it down to my MS, "It's your MS, it's your MS," and I was given drugs, Baclofen for the muscle spasms, and Amytriptiline 10mg for the nerve pain,. […] she referred me to another consultant who prodded my leg and said "Take some Baclofen." And, and [um] nobody, because I've got MS nobody thought oh it could be something else. And the spasms got really really bad. […]

I ended up going to casualty and had a, I'm laughing 'cos I can't believe this, I had an emergency hip replacement 'cos my, I had a fractured femur, and that was why I had all the muscle spasm. [...] And they think it started off with a hair line fracture and just got worse and worse and it got to the point where the ball of the femur was completely gone. That's when I went into hospital. Couldn't, I'd been walking around on it for a whole year and it had gone like this. I can't believe it still really"

[Melissa (13), female, 46 years old]

A report commissioned by the World Health Organisation (Dua et al., 2008, p.28) identified the difficulty in identifying the presence of MS as "a matter of concern" because its symptoms are easily confused with that of other conditions. The extracts from Melissa's and Sadie's interviews above exemplify the same concern but in the reverse; a concern that once diagnosed with MS, problems are put down to MS and other causes are not considered. Not only is there the potential that (non-MS) problems go untreated and exacerbate, but people with MS may also experience anxiety if their concerns about potential other causes are not alleviated or investigated further.

7.4. Intervention uncertainties

There are various interventions and strategies that people with MS use, in order to prevent, manage and treat their MS. However, those which are licensed for MS are limited and the effectiveness of these varies between individuals and even at different stages of one's disease course (Dua et al., 2008). From a clinical perspective the best course of action is often unclear, this along with difficulties in assessing disease activity

(discussed above) and the unpredictability of disease trajectory means that assessing the effects of interventions on an individual level can be challenging.

7.4.1. Uncertain treatment effects

An implication of unpredictable illness patterns is that assessing the effectiveness of interventions is problematic. There is often confusion over whether recovery or the absence of symptoms results from the intervention effects or whether it happened to coincide with a period of remission. Oliver had an operation to treat chronic cerebrospinal venous insufficiency (CCSVI); he and his wife discussed the difficulty they experienced when assessing whether it was effective:

> *"Oliver: No that's it, you don't, that's it, you don't know.*
>
> *Wife: What we don't know as well is has it halted the progression?*
>
> *Oliver: Yeah would have I been worse if I hadn't had the operation?*
>
> ***Mm. Yeah. Yeah.***
>
> *Oliver: So that's hard to know.*
>
> *Wife: You can't measure that can you?*
>
> *Oliver: Yeah would I be worse now*
>
> ***That's the thing isn't it?***
>
> *Oliver: without the operation? […] It was a lifeline, it was something that might work, might be working. We don't know."*
>
> *[Oliver (12), male, 57 years old]*

Although initially they may have hoped for more pronounced effects, in the extract above Oliver and his wife explain how they could not confidently assess whether the operation was successful. As a result of unpredictable trajectories, many participants

chose to continue with an intervention whilst being unsure of its effectiveness, and sometimes suffering with side effects in the hope that it was working:

Ava:

"I do take supplements, [er] Starflower oil, which is sort of like a potent Evening Primrose Oil, that is one thing that the MS Society will actually recommend. [Um] And Fish Oil, that's another one. And then just vitamin and a think called MSM, one of those I've been taking for donkeys' years and it's one of those things, I think well I don't know if they're doing me any good or not, but I don't want to stop them in case they are. You know and it's hard"

[Ava (15), female, 63 years old]

Laura:

"I would like perhaps that, it's hard, if you give yourself five years off a drug to see how I will progress on my own but then if I'm not taking the drug for those five years I don't know what good or what bad it's doing. It's really, it's really tough. It's really hard."

[Laura (19), female, 30 years old]

Kamila:

"The side effects [of DMDs] were so bad, I thought actually I, I don't know, I don't think I can do this. 'Cos it's literally you know shaking so badly that you can't do anything. You can't stop and you've got such a bad fever and it's just you know do I want to have this every other day you know? […] I'm aware of the fact that I don't need to take them but as I say so far it seems to be working."

[Kamila (21), female, 29 years old]

Such uncertainties around intervention effects means people might assess ineffective strategies as effective, or be reluctant to dismiss strategies that may appear ineffective (further explored in chapter 8). The unpredictability of MS trajectories appears to reinforce reluctance to cease treatments where effectiveness is unclear as people with MS cannot predict how they would fare without it.

7.5. Discussion

The findings suggest people with MS may face a range of uncertainties. At the point of diagnosis, unpredictable prognoses and heterogeneity in the type and severity of symptoms leads to existential uncertainty. Such existential uncertainty results in situations where participants often imagined severe prognoses and perceived 'biographical abruption'. This can result in feelings of fear, anxiety and powerlessness for people with MS. Not only are these feelings undesirable in themselves, but low mood has also been shown to be a predictor of impeded functional ability for people with MS (Smith and Young, 2000). In addition to potential upset, the way in which people with MS envisaged their potential future with MS also appears to shape life decisions made. With time, space provided by existential uncertainty allowed opportunities for many participants to carry out 'biographical restoration' (or 'repair') as they reconceptualise their MS as milder, and futures as more favourable than previously imagined. This suggests initial perceptions of 'biographical abruption' can often be unrealistic and its consequences unnecessary. In Sharpe and Curran's (2006) model of adjustment to illness, they identified the importance of the match between the perceived illness representations that people develop (based on previous beliefs) and the reality of their illness experience. They described adjustment to illness as follows:

> "Adjustment to illness can be viewed as the process to maintain a positive view of the self and the world in face of a health problem. […] Assuming these positive views are accurate, then positive psychological functioning will result" (Sharpe and Curran, 2006, p.1161)

Thus, a tendency for people with MS to imagine worst case scenarios may have detrimental effects on their adjustment to MS. This may be avoided, especially when such illness representations are inaccurate. Sharpe and Curran also argue that:

"[b]ecause in most instances, illness representations are relatively new, less stable beliefs, it is likely that individuals will find it easier to incorporate new helpful information into their illness representations in the first instance" (Sharpe and Curran, 2006, p.1611)

This highlights an opportunity to support people with MS to develop more positive (and often realistic) views of their potential future in order to aid adjustment and avoid emotional impact and decisions based on unrealistic futures, during perceived 'biographical abruption'. Although, at this stage, it is important to note that this may be more appropriate for those with milder trajectories of MS than those severely affected.

The fluctuating nature of MS means that some find their symptoms differ on a daily basis which affects their capabilities to take part in everyday life and planned events. Participants' accounts suggested that this daily uncertainty meant their condition consumed their thoughts as its implications were often at the forefront of their mind. There were clear examples in the data of participants not only changing daily plans due to being physically restricted by the effects of MS, but there was also evidence that frustrations or burdens accompanying the planned activities, being more deliberate and conscious, meant some avoided making plans.

The lack of instrumental biological measures of disease activity and the variety of potential MS-caused symptoms means that people with MS can experience embodied uncertainty when assessing causes of problems. Similar to the daily uncertainty of a fluctuating illness, considering whether causes of problems are MS may also be consuming as, again, it frequently brings their condition to the forefront of their mind. Additionally, the act of (mis)attributing problems to MS, may cause additional distress in that it depicts a more active MS, which may not be accurate. Embodied uncertainties also have potential consequences in the clinical settings. Some participants felt concerned that access to appropriate help may be limited if healthcare professionals

overlook other causes or dismiss symptoms as being unrelated to MS. This could result in either problems not being addressed appropriately, or a worry that they are. Attending to such concerns through discussion during consultations or further investigations may alleviate worries felt.

The unpredictable course of MS means that it is particularly difficult to assess the effectiveness of interventions because absence of symptoms could be due either to intervention effects or to periods of remission. The lack of instrumental measures means that people with MS must rely on their own experiences to assess the presence of MS, but this also appears to be problematic as participants said they lacked confidence in such interpretations. This results in situations where people with MS may continue with interventions that have unclear effects (and sometimes severe side effects).

Chapter 8

How do people with MS discuss their perceptions and use of information in relation to managing their condition?

8.1. Chapter summary

People with MS use an array of information in different forms when managing their condition similar to others with long term conditions. This chapter focuses on areas in interview data that emerged as particularly pertinent in terms of how participants used information when managing their condition. It begins with an exploration of how people with MS perceive information provision during diagnostic stages and the consequences of seeking information independently during this time. It then examines the value and limitations of own experience as an information source when managing MS, and how participants used experiential information and peer support from others with MS.

8.2. Information around the time of diagnosis

The period before, during and after diagnosis was identified as particularly important for participants, in terms of their support and information needs and emotional distress experienced.

8.2.1. Pre-diagnosis period

Before diagnosis of MS, symptoms potentially caused by MS are defined as 'clinically isolated syndrome':

"Clinically isolated syndrome (CIS) is a term that describes the first clinical episode with features suggestive of multiple sclerosis (MS). It usually occurs in young adults and affects optic nerves, the brainstem, or the spinal cord. Although patients normally recover from their presenting episode, CIS is often the first manifestation of MS." (Miller et al., 2012, p.157)

The presence of CIS alone is not enough to confirm diagnosis; subsequent evidence in the form of tests to exclude other conditions, MRI scans and subsequent symptoms to confirm MS are required (McDonald et al., 2001, Polman et al., 2011). So for most people with suspected MS there is often a liminal stage; a period of waiting for a diagnosis after experiencing first symptoms, or "limbo-land" as one participant put it.

Symptoms without a disease label can cause distress; those experiencing them may feel their illness is illegitimate, feel under scrutiny and are not granted "permission to be ill" (Nettleton, 2006a, p.1167). A recent review of information provision for people with MS highlighted the time around diagnosis as being "emotionally intense and unforgettable" and that increased distress was linked to lengthy periods before diagnosis confirmation (Solari, 2014, p.399).

The analysis in this study suggests that participants also felt distress during the pre-diagnosis stage. However, it was common in participants' accounts that the frustrations felt were aimed at healthcare professionals, rather than the situation (of MS being difficult to diagnose). Participants interpreted healthcare professionals' communication as unhelpful, evasive and sometimes felt they were not taking them seriously:

Robert:

"So after that second attack I saw a consultant neurologist for the first time who, who said after the second attack that it may be MS, [um] but the way that he said that [um] was to this day rather infuriating because he, basically he said, "We don't know what causes MS. We don't know how to treat MS and my recommendation to you as you're a relatively young man is to go away and do your best to forget about it."

[Robert (10), male, 53 years old]

Laura:

"He just basically said, [um] "Your MRI would suggest that you have MS." And I said, "Okay." And he just said, "But [um] we would like, we would like you to have some more, [um] something before we formally diagnose you." Because I think he'd only seen the fact that I'd collapsed a couple of times as not being like a clinical relapse as such and [um] he was quite flippant, he said [er] he said, "We'll see you if it flares up again," or something like that. And I was like, "Thanks,"

Right.

That's not very like, you know, it's quite, he was, he was quite flippant. […] So I think [um] the first guy could have been a bit straight. [Crying] Sorry.

That's okay.

I have talked about it before but it's still horrible."

[Laura (19), female, 30 years old]

Ulrike:

"So they did an MRI on the spot and [er] and then he just said, "Oh it was very inconclusive, it could be this," he said, he said, "There's a, about a 50% chance it's a virus." He said, "Or it could be MS." He said [er] and of course it's worse, like I, 'cos I'd thought it but I hadn't really vocalised it.

Right.

And he said, "I'd go home and wait for things to happen to you." And that was it. […] Within twelve hours of seeing him I'd developed, if you could imagine what it feels like if somebody does that in your face

Mm.

punching it.

Right.

It's that feeling. My face all went sort of, and you think if you looked in the mirror it would have caved in, in fact it wasn't because it was just the nerves. That was within twelve hours of seeing him. And I then started on the big relapse that caused my leg to, and I put it all down to him.

Yeah, 'cos of the,

Yes, 'cos of the stress. Yeah."

[Ulrike (14), female, 62 years old]

As illustrated in the extracts above, it appeared to be common that participants were given little information and told they may have MS, but additional symptoms were needed to confirm diagnosis. Frustrations with healthcare professionals during this may reflect discourses around biomedical knowledge providing answers to health uncertainties experienced. Adamson (1997) described expectations of meetings between clinicians and patients as follows:

"The ideal medical encounter is one in which the physician is certain of the correctness of his or her clinical diagnosis, and the patient obtains a clinical response which alleviates his or her feelings of existential uncertainty" (Adamson, 1997, p.136)

Participants may feel frustrated with healthcare professionals who could not confirm diagnosis, if they are expecting them to do this. Not only that, there appears to be a lack of information provision during this time; being told that you *may* have a life-changing condition and to wait for further symptoms is unlikely to alleviate distress or concerns. Other participants remembered they were told they did not, or were unlikely to, have MS during this time. This also appeared to be unhelpful as it led to feelings of shock for one participant when MS she was eventually diagnosed (Kamila) and frustration for another who believed she had MS but her concerns seemed to be overlooked (Anna).

Based on these findings there appears to be an opportunity to improve information provision for people suspected MS undergoing diagnostic processes. NICE guidelines (2003) at the time of the interviews recommended that patients' information needs are assessed and reviewed during diagnostic investigations. However this recommendation has been omitted from most recent NICE guidelines (2014); although they recommend people with MS should be well informed and supported *at the point of* diagnosis, they do not provide guidance on information provision *during* the pre-diagnosis period, as previous guidelines did.

In recognition of the importance of information during this stage, a German study developed an information leaflet aimed at people with suspected MS (Heesen et al., 2009). They assessed its impact on people diagnosed within 2 years (retrospective cohort) and people with suspected MS (prospective cohort). The leaflet was comprehensive as it covered areas such as; the potential for benign MS prognoses: decisions about whether to have diagnostic tests; the potential negative and positive implications of receiving a diagnosis; the unpredictability of prognoses; the limitations of diagnostic tests i.e. they cannot definitely rule out MS; benefits and risks of early or delayed treatment initiation; and information about the diagnostic tests and processes.

They found that the leaflet was received favourably and most people preferred to be involved in diagnostic decisions. Openly addressing uncertainties inherent in MS was evaluated as particularly helpful. The leaflet did not appear to cause anxiety or make decisions about diagnostic tests more difficult. The majority of participants thought the period before receiving a diagnosis was an appropriate time to be informed about the potential implications of having MS. Despite favourable study outcomes, this type of information has not become part of routine care in Germany where the study was set and the reasons why are unclear (Heesen, 2016).

8.2.2. Post-diagnosis period

In additional to a lack of useful information during diagnostic investigations, participants also reported that the provision of information and support after confirming diagnosis was lacking.

Hannah:

"I left the hospital ward [after receiving a diagnosis] feeling as though you know you're on your own. There was no sort of support out there.

[…] Even at my first neurology appointment I thought I could have been given some kind of leaflet, document, explaining more about MS. I basically found out about MS [um] with my own research."

[Hannah (20), female, 49 years old]

Laura:

"But I don't, I don't find they give you enough information, that's the honest truth. […] I don't think they have, they have no leaflets available over here, they don't give you any point of contact, it's pretty much you, you're just, it's just you have to go and do that yourself,

***Yeah**.*

and I find that quite [um] frustrating. And I don't think it's [um] a good reflection on the NHS to be honest.

No.

I think that they should be better prepared. Definitely."

[Laura (19), female, 30 years old]

Kamila recounted her diagnosis as particularly distressing, firstly because she thought MS had been previously ruled out (a problem that could have been avoided with information about limitations of tests during pre-diagnosis stage) and secondly because she was informed by a letter with no follow-up information:

"I've been told by a letter coming through the post, [um]

Right.

that I have MS […] Which was obviously quite shocking. It was quite [um] disturbing especially because I didn't really expect it and [um] as I say I'd been told [um] that I have MS in a letter, and the letter didn't mention anything about a follow up appointment or make an appointment, nothing like that.

[Um] They left me hanging for about a month when I had to ring the hospital neurology for about, about three times actually forcing them to give me an appointment with a neurologist. [Um] The first time when I called the secretary said [um] "Oh I'll have a look if you've, if you are scheduled to have an appointment." And I didn't hear anything in two weeks so I rang again and she said that again. And I said, "Well you need to check because you know I've been told I have MS and I don't know what to do, you know you left me in the dark. [Um] I don't know what to do, I don't know what it means, I don't know if I have to take or have any treatment. I know nothing.""

[Kamila (21), female, 29 years old]

As exemplified in the extracts above, participants commonly reported feeling frustrated by the lack of information at the time of diagnosis. This information gap was also highlighted in a recent systematic narrative review into MS healthcare services in UK that identified a serious lack of information and support for those diagnosed with MS (Methley et al., 2015). Several participants (Chris, Adam and Sadie) even recalled being advised by clinicians not to search the internet for information about MS, which they did anyway. Such advice suggests that healthcare professionals may also be aware that information provision is lacking. Participants often felt they had to search for information independently. Analysis suggests that this was problematic as the information gap was often filled with negative representations of MS, which often had detrimental consequences.

8.2.3. Information accessed and worst-case scenarios

Participants recalled how around the time of diagnosis they accessed information about MS. This was typically by carrying out their own research and sometimes through personal contact and communication with others. The findings presented in chapter 7 demonstrated how people with MS seem to imagine severe prognoses around the time of diagnosis. Such perceptions seem to develop and be reinforced by representations of severe MS trajectories which are often accessed around this time. Mozo-Dutton et al. (2012) also identified similar experiences in their study and described how people with MS were "overwhelmed and frightened by the information they uncovered about MS, fearing that they would shortly be 'fit for nothing'" (p.1212).

Participants described how preconceived ideas about MS and external factors, such as other people's reactions, contact with others who appeared severely affected by MS and information that they accessed, further compounded their fear of potential symptoms. One participant recalled how, around the time of diagnosis, she was upset when her GP appeared to assume that she would stop working, and her auntie suggested she should move in with her mum. A common experience identified during analysis was that during the initial search for information about MS, participants said they found examples of people severely affected. Katy and Miles both found the information they initially accessed online particularly harrowing:

Katy:

"And I then did [look up MS online], and promptly dissolved into tears 'cos obviously you always get the worst-case scenarios come up first. [Um] It was fairly horrendous […] And, as I've said, you know, whenever you read about it in the papers or Google it or anything, you literally come up with the, you know, the people who are in wheelchairs and can't, incontinent, can't talk and do anything for themselves […] I woke up every single night having had exactly the same dream, of waking up, trying to get out of bed and not being able to walk. And I'd wake up so scared that I couldn't get out of bed […] it's so scary when you read that, and you see somebody going, you know, you think, "Oh my God, I'm going to be incontinent in a wheelchair, unable to feed myself."

[Katy (03), female, 41 years old]

Miles:

"What I found out shocked me to my very core. [Er] Which I couldn't walk afterwards because [um] I know now that that [er] shock and stress and pressure all play huge parts in MS and my ability to deal with those things then wasn't good".

[Miles (09), male, 44 years old]

Miles described how he struggled to deal with the information he accessed and related the "shock and stress" he experienced to exacerbation of physical symptoms. A similar relationship was evident in Ulrike's extract (in section 8.2.1 above) as she also linked a stressful experience with exacerbation of sensory symptoms. The analysis showed that other participants also believed that stress exacerbated their symptoms, and there is some recognition of this association within the literature (Mohr et al., 2004), although causal mechanisms remain unknown.

As well as online encounters, some participants described having face-to-face contact with people who appeared to be severely affected by MS and the effect this had on them when imaging their own potential futures. Whilst waiting to see a counsellor at an MS therapy centre, Robert saw other people who appeared to be severely affected by MS:

"And just because it's my character I went off and found out [er] a lot more about it. But probably found out a lot more about it in the wrong way, in that I went to a lot of MS clinics, support clinics and [er] was sort of around people who were ill 'cos MS is a real scale of how good or bad it could get. So, it was around a lot of people who [um] had significantly worse MS that me. So, I think I, I don't think that was a great decision actually. That, that kind of search for information, but that search for information was because I didn't get any information when I was, when I was first told that it could be MS. […] I was sitting waiting for her while there was all these people in wheelchairs and on crutches and Christ knows what, and [er] I just thought, "Oh this is," you know you just, and this guy [the neurologist] who had just told you that they didn't know how to treat this disease, and to go away and you know forget about it. And you just saw, you just saw your life completely changing".

[Robert (10), male, 53 years old]

The extracts above (from Robert, Miles and Katy) show how distressing seeing examples of severe MS prognoses can be. Having contact with, or accessing information about, others severely affected by a condition can be particularly distressing for people who share the same diagnosis. This is not just because of the upsetting nature inherent in this information, but also because they may compare themselves and see their own future reflected (Locock and Brown, 2010, Mazanderani et al., 2012).

Some representations of MS are accessed via chance, and such unsought sources may present particularly negative images of MS for various reasons. Others who are seeking help (and seen in places such as waiting rooms) may be those who are severely affected, as more positive examples of people with MS who are coping or not too severely affected may not require help from such settings or noticeably have MS, as their problems may be less 'visible'. Similarly, media stories about those with MS may be extreme examples, as people with milder versions make less sensational stories. These factors may skew perceptions of MS towards representations that are particularly negative and feed into conceptualisation of potential futures with MS.

As explored in chapter 7, analysis showed how initial perceptions of severe MS prognoses were often later countered with perceptions of a milder or more manageable version of MS. Initial access to negative illness representations can cause distress and impede adjustment to illness (Sharpe and Curran, 2006). Based upon the findings presented here and in chapter 7 there is a need for more information about MS around the time of diagnosis. This could include information presenting a broad range of trajectories, including more empowering and positive representations. In order to have a positive impact, such information may be more useful early on before illness perceptions become more established as Sharpe and Curran argue that:

> "[b]ecause in most instances, illness representations are relatively new, less stable beliefs, it is likely that individuals will find it easier to incorporate new helpful information into their illness representations in the first instance" (Sharpe and Curran, 2006, p.1611)

By providing more helpful information around the time of diagnosis to people with MS, they may develop more positive (and often realistic) views of their potential future and avoid distress and factor a more realistic perception of the future with MS when making life decisions (see chapter 7).

Borreani et al. (2014) trialled an information aid for people newly diagnosed with MS in Italy. The information aid included an interview with clinicians whilst using an IT resource and a take-home booklet. The exact details of the intervention's information content were not described, but they found that participants had improved attitudes towards, and understandings of, MS and some reported that it alleviated fears and helped them realise their MS might not be severe. However, they also found there were practical issues; there were difficulties in incorporating the process into busy practices and some participants felt that the timing of the interview did not suit them. Participants

suggested it could be timed according to individual patients' preferences and some thought it would have been useful earlier during the pre-diagnosis stage. This supports an argument made earlier, that people with *suspected* MS would benefit from more information. Some clinicians facilitating the information aid voiced concerns that it might not be suitable for people with more progressive forms of MS; interestingly this concern was not voiced by participating patients.

8.2.4. Psychosocial support

The findings presented in the previous sections demonstrate the potential worth of information presenting a broader range of MS trajectories that include more positive examples. In addition to learning about the breadth of potential trajectories, the analysis also showed that some participants did not value provision of biomedical information from healthcare professionals and they would have instead preferred more information about psychosocial aspects of MS.

> *Adam:*
>
> *"I suppose if there's something [um] I think is missing [in the information and support provided], I think it is stuff around [um] oh it's something naff, sounding crazed, but the kind of art of living with something like this. I wonder if there is something missing around [um] [pause 10 secs] what this means for, for kind of practical and emotional life I suppose. […]*
>
> *Yeah. I mean there's, everything's out there. I mean there's all sorts of, if you want to read about [um] bowel dysfunction in MS you can. [Um] But in terms of how to live with it, how to not let it, I suppose how to sort of reappraise your life and your ambitions and [um] [pause 10 secs] I suppose how to still design and manage a life with it.*
>
> *[Adam (11), male, 36 years old]*

Kamila:

"Yeah I, I definitely did [have questions about life implications] but I just didn't feel comfortable asking them […] And maybe I felt too stupid to ask, or I felt that the questions were too stupid.

Right.

I don't know. I just, I just know that I do remember coming out [of consultation] and thinking well that was a waste of time. I do remember that

Right

because it didn't really, you know, I had had months to do my research, I knew what MS is, I know how it you know why do you get it, and I literally when I found all the different factors that contribute to MS"

[Kamila (21), female, 29 years old]

Patients increasingly have access to biomedical information typically accessed via the internet. Both Adam and Kamila refer to the ease of accessing information about biomedical understandings of MS which made this less useful when presented by healthcare professionals during consultations. In light of this they both would have found information about "the art of living" with MS more helpful. It is increasingly recognised, especially by those who advocate person-centred care, that healthcare should expand further than biomedicine, to include emotional needs, life issues and difficulties of every day (Entwistle and Watt, 2013). Although they would have liked to discuss these broader issues, descriptions such as, "something naff, sounding crazed" and "I felt that the questions were too stupid", suggests they felt that it was unusual or inappropriate to discuss these types of information.

Expanding this point further, some participants seemed to believe that the type of information and support their healthcare professionals provided did not help them to manage their MS. Ava and Oliver felt they gained nothing from healthcare consultations:

Ava:

"And then they'd test all the various things they test. And they'd mark it all on the little man and [um] like I say tell you to go away and see you next year and, and that would be the end of that. So it sort of seemed a bit of a pointless exercise to, come on okay you've marked me at like, do you know what I mean? It wasn't getting any, it wasn't getting me anywhere. It might have helped their records but it wasn't getting me anywhere.

Mm. Yeah. So you weren't given any advice or

No.

information? It was just monitoring

Yeah. That's what it seemed to be

your condition.

Just monitoring. […] Yes I suppose I've developed that attitude over the years 'cos it, I haven't, I haven't gained anything much from the medical profession because they've got nothing really to offer me really I suppose. Nothing that's gonna work and therefore we've worked out our own way of managing things and [um] so that's, that's what I do, just carry on with it, and just hope I'm not really totally and utterly incapacitated in the end."

[Ava (15), female, 63 years old]

Oliver:

"Cos you have to, in fact you feel worse when you're actually seeing the neurologist you don't feel uplifted or anything like that, because you, you learn nothing from him. He's telling you nothing. […]'Cos what he's, what he recommends is nothing really"

[Oliver (12), male, 57 years old]

In the extracts above Ava and Oliver are referring to the yearly appointment with neurologists. These regular reviews are recommended by NICE guidelines (NICE, 2014). Ava and Oliver assessed these as being of no use, so much so that Ava has now decided to stop attending check-ups and Oliver only continues to attend because he hopes that there will be a treatment breakthrough one day and he will be in the system to gain access to it. In the context of a condition like MS, for many there are often no

medical treatments available, thus the value of consultations focused on medical treatment and monitoring only is questionable

8.3. Experiential information

Experiential information is gained from people's own experiences of MS or through experiences of others with MS. Participants discussed experiential information as a particularly useful information source for managing and understanding their condition. The heterogeneity of MS, and difficulty in objectively measuring disease activity, mean that people with MS rely on personal experiences of *their* MS when monitoring symptoms, progression and remission. For example MRI scans show indications of disease activity; however, the way in which these directly translate to symptoms is unknown (Wilkins and Scolding, 2008) and after receiving a diagnosis people with MS (in the UK) generally do not have regular MRI scans (ABN, 2009). Existing evidence and participants' accounts suggest that the way in which MS activity is assessed is generally through personal experiences.[8]

8.3.1. Embodied experiential information

The heterogeneity of MS means that people with MS often have to rely on their experiences to understand their MS. Participants described how they drew on their experiences to decide whether to take action; assess effectiveness of interventions; evaluate side effects; and recognise relapse triggers. Personal experience appeared to be a vital information source when learning about their personal relapse triggers. Both

[8] The most commonly used standardised scale to measure MS symptoms in clinical trials is the Expanded Disability Status Scale, which records illness experience, as opposed to more instrumental measurements which monitor illness activity in other conditions, e.g. blood glucose, BMI, or blood pressure.

Chris and Oliver identified patterns in their previous experiences that seemed to act as relapse triggers:

Chris:

"How do you learn to recognise symptoms and triggers?

Experience I suppose really is that when things, when things keep happening and [er] I just know that sort of if I overdo it then I, too much then I get overly tired then sort of I, that, this does trigger [er] sort of [er] sort of slurring and sort of stumbling around a bit and [er] I also know that if I get under the weather with a cold or something I get the same things so I try and avoid having a cold or, or being ill. And also just people in the office are ill then I tell them to go home rather than sort of stick around the office and make me ill. […] Okay yes. [Er] I think the first biggest way that I've [er] managed it is I [er] with the sort of tiredness and things I've changed my working hours very early on and so I generally sort of, whereas I used to go, get into work for about half eight now it's sort of half nine, ten o'clock in the morning.

Okay.

And [er] and just sort of changing my working hours and and so that I have more time to wake up in the morning has been extremely helpful. [Er] And [er] one of the other ways was really early on I told everyone in the office what [er] that I had MS and that if I'm erratic or tired or have to disappear off [er] that [er] I will do. And so that removed a lot of stress out of the situation. And so those were the, the two things that I did very early on.

[Chris (08), male, 38 years old]

Oliver:

"Well you, you know as soon as you're in the sun, and when you've got the fatigue with the heat, you know you're not going to do it again next time. 'Cos you are absolutely exhausted, you know you can't, I mean I was out there listening to, I think last year, it was a lovely day like this, sat outside in the sun listening to the cricket, only on about 10 minutes and I couldn't, I could hardly get back into the house."

[Oliver (12), male, 57 years old]

Both Chris and Oliver were motivated to change their behaviour due to their own previous experiences. Participants also gained knowledge about possible relapse triggers from sources other than personal experience. However, the heterogeneity of MS

means that triggers differ between individuals, and so they can only learn which relapse triggers are personally relevant, by experiencing them. For example, an MS nurse told Sadie that heat may cause relapses, but she personally has not found this to be the case:

> *"When I first was diagnosed they said stay out of the sun, but the sun makes me feel so much, my sister lives in Cyprus and every year since she's moved out there we've been going for holidays and I love it. That's when I'm at my best. […] I hardly take any painkillers*
>
> ***Yeah.***
>
> *I love it. I absolutely love it. I mean me and [my partner] was at one time thinking of selling up and moving out there."*
>
> *[Sadie (17), female, 50 years old]*

This extract demonstrates how generic information about MS may be countered by own personal experience. This reflects how generic information about MS is not always applicable on an individual level and the importance of considering personal experiences when managing illnesses.

8.3.2. Limitations of embodied experiential information

Although embodied experiential information is a vital information source, participants also understood its limitations. As explored in chapter 7, unpredictable illness trajectories make it difficult to assess the effectiveness of interventions. It would be impossible to judge whether intervention effects are 'real', based on participants' accounts alone. In addition to this, some participants commented upon the possibility that the 'placebo effect' may play a part when an intervention appears to be effective. In the extracts below, Oliver and Ava both discuss the possibility of this:

Oliver:

"Yes, if you're trying something yourself you, and you do feel better for it. You know it's definitely a, psychologically [er] stronger isn't it? That [er] after I had the [CCSVI] operation when I went into the restaurant I walked a lot better didn't I? […] 'cos not everything is going to work for the same person. There's, a lot of, it's a mental thing definitely.

[Oliver (12), male, 58 years old]

Ava:

"And I don't need to [try anything else] because I've found the thing [manual lymph drainage] that I believe in. I think that's what it is, I believe in it. […] So it's just, you find things that suit you for whatever reason.

Yeah.

Whether it's just psychological which I really doubt, but [um] doctors as a, as a sweeping generalisation, can be a bit cynical about these things and sort of, "Well you, you do that if you think it does you any good dear." Sort of thing."

[Ava (15), female, 63 years old]

Oliver's reference to "psychologically", "a mental thing" and feeling better for "trying something" suggests he questions interventions' 'real' effects. Ava seemed less cynical of their 'real' effects, but did explore the possibility that effects may be "psychological". She also displayed some ambivalence to this possibility, as she said, "you find things that suit you for whatever reason" suggesting it did not matter why an intervention seemed effective, the important thing was that it appeared to work.

As explored in chapter 7, unpredictable prognosis means people with MS may attribute (or misattribute) absence of symptoms to effects of an intervention, rather than a period of remission. Additionally, uncertainty around symptom causes may add to confusion in assessing intervention effects (see section 7.4). The limitations of the clinically proven strategies available appears to result in a situation where people with MS seek alternative approaches to treat, or manage, their MS. The variability in apparent

‘effective’ interventions may reflect the heterogeneity of symptoms and treatments effects i.e. ‘real’ effects differ between individuals.

8.3.3. Experiential information from others with MS

People’s use of health-related experiential information whilst managing their condition is not a new phenomenon, as people have always shared experiences about common health issues. The subject has gained increasing interest in more recent years, as the internet has provided platforms allowing experiential information to become more accessible and interactive (Ziebland and Wyke, 2012). Analysis showed how participants gained experiential information via face-to-face contact (MS centres, support groups, waiting rooms and courses), internet (social networking, forums, blogs), charities, and magazines. Locock and Brown’s (2010) research into support group experiences of people with MND and carers demonstrated how contact with others can provide positive outcomes, as support groups provide space for practical and social comparisons, allowing attenders to gain information about practicalities of living with MND and feelings of reassurance and camaraderie. Entwistle et al. (2011) also demonstrated the beneficial ways that people use experiential information to manage health such as clarifying their own values and reasoning, providing ideas or signposting to options and services, exploring practicalities and support with coping.

There were numerous examples in the interview data for this study of the various ways people use experiential information that support these previous studies. These include learning about the real-life practicalities, signposting to services, sharing ideas, gaining reassurance and empathy, and reinforcing perception of the heterogeneous nature of MS. Some participants discussed the reasons why they found experiential information

particularly useful. Anna compared the information she gained from a peer support session to the information she obtained from healthcare professionals:

> *"It allows six random strangers who are only, the only thing they've got in common is MS to exchange information and, and we know our illnesses better than any doctor can ever know. My neurologist is great but he hasn't got MS. You know so it's only ever going to be flat words on a page to him, no matter how great he is at what he does he is never gonna experience it. So by getting six people into a room and discussing one particular facet of MS [um] is in a way much more empowering than a GP or a neurologist just writing out another prescription, without going under the surface and saying, "Well why do you want this?" And "Why do you feel you're still on that medication?" You know and we all talked about the fatigue I was like, "Oh yeah that's a good idea," and you know.*
>
> *[Anna (07), female, 39 years old]*

Anna refers to the empathy gained from sharing experiences. She refers to knowledge gained only through experience, and not medical training. For Anna, sharing experiences with others not only provided a valued source of information, but it was also empowering as she felt involved in self-managing her own condition. Robert and Katy also found experiential information particularly valuable:

> *Robert:*
>
> ***"So what's, [um] useful about looking at people's, other people's experiences? Why is that helpful?***
>
> *Well I think it's always, I've always found it helpful to find out what the individual views are of anything really. [Um] But I think in this case it was particularly useful because there was such crap information elsewhere.*
>
> ***Mm.***
>
> *So, so actually in some ways, in some ways the only source"*
>
> *[Robert (10), male, 53 years old]*

Katy:

"I think the difference, why I like it rather than say the MS Society is, they're, they're giving you all the information, etc. but it's very impersonal whereas on Shift [social networking website] it's everyone on there either has it or most people on there have it to a greater or lesser extent. Or they are you know with someone who has it or someone in their family does. So it's personal experience. Which is why you know a doctor who's telling you about MS will say, "Oh you know, stress might not be blah, blah. You should probably try and avoid stress." But they don't actually have the experience themselves.

[…] I'm thinking it's stress and it's backed up by everyone else saying the same thing and if you've got a question you can put it on there and you get personal answers rather than just "Research shows that…" which it might well do but if that doesn't necessarily apply to me, […] So they might not have the medical knowledge about it, but they do have the anecdotal personal knowledge about it so I'd probably listen to them on how to deal with stress rather than a medical person,

Yeah.

You know like techniques that have worked for someone who is actually in that situation rather than someone who's, knows the theory of it but not, not the [um] the reality."

[Katy (03), female, 41 years old]

The analysis showed how participants sought out information relevant to their own experiences of MS and the practicalities of managing a problem. This supports evidence from a recent focus group study that showed, although there appears to be lots of MS information available, people with MS can find that the majority of it does not apply to them personally (Synnot et al., 2016). Specific experiential information may be perceived as particularly useful in a context where the heterogeneity of MS means that it is difficult to find generic information which is applicable on an individual level.

A limitation of learning from others' experiences is that typically it is not suitable to assess effectiveness of interventions. Previous studies have shown that rather than using it in this way, people tend to use experiential information as a tool to understand and

reflect upon problems and situations and explore different ideas, and combine it with other relevant types of knowledge (Entwistle et al., 2011, Armstrong and Powell, 2009). In support of this literature, the analysis showed that participants appeared to use experiential information in a measured and critical way. Although Laura often found experiential information useful, she made it clear that its usefulness is context-dependent and sometimes she avoided it:

> *"And try and steer away from too much you know personal anecdotes. Well they're helpful in their own way, don't get me wrong. I do read them and stuff and I use the MS website, society website and blogs and everything. I write a blog myself*
>
> ***Okay.***
>
> *[Um] so I would, I would use personal anecdotes to make a decision but not as much as I would rely on proper medical research.*
>
> *[…] I think they both have their place, I think they just they have their place in different times for you, for instance like for me you know if I'm in a, in a relapse I probably won't read a lot of anecdotal things, personal things because it can be quite tough to read that but I'll be more inclined to read sort of more straight lined fact. But then when I'm well, out of a relapse I'm more inclined to dip into blogs, people's experiences [um] because it's easier to read them whenever you're not in the throes of it yourself.*
>
> *[…] And like Twitter is a fantastic way of getting in touch with other people who have MS, without having to you know be too graphic or in depth, we just are, it's almost like a support network."*
>
> *[Laura(19), female, 30 years old]*

Laura implied that experiential information is less reliable in some contexts, by referring to medical information as "proper" and "straight lined fact". However, she also believed that experiential information was very useful, but for different purposes. Laura's claim that social media provides access to others with MS, "without having to […] be too graphic or in depth" exemplifies how online social networking can allow users to control their contact, thus enabling them to gain information whilst maintaining some distance at a level suitable to them personally (Mazanderani et al., 2012).

Not all participants displayed understandings of the limitations of experiential information; others considered experiential information less critically. Mary orders Low Dose Naltrexone (LDN), a treatment not licensed for MS, from a chemist who posts it to her:

> *"It hasn't been clinically proven, but it does help a lot of people having read on forums and so on, you know but, but yes it is a strange way to get it. [Um] There's a [er] a chemist in [town] that dispense it and we just ring up and they sort it out and post it."*
>
> *[Mary (05), female, 61 years old]*

The presence of others claiming LDN worked for them appeared to be sufficient evidence of effectiveness to encourage Mary to try it. Interestingly, it has been shown elsewhere in the literature that some people with MS regard experiential information as particularly reliable or trustworthy because it is seen as an independent source (with no other factors biasing its representation) (Synnot et al., 2016). Being persuaded by others' experiences that an intervention is effective may be more concerning in certain contexts. For example, LDN (referred to in the extract above) is relatively cheap, does not promise dramatic effects and has few side effects. It may be more concerning when people try expensive and potentially harmful interventions that promise dramatic outcomes, based on anecdotal accounts alone. One such intervention is surgery to treat chronic cerebrospinal insufficiency (CCSVI); a controversial unproven approach (see section 2.6.3). Several participants had researched CCSVI and subsequently decided against it. However, two participants decided to have the surgery, both paying £10,000+ for the procedure. At the time of the interviews, Melissa was scheduled to have surgery soon and Oliver had already had it. They both drew on experiential information as evidence for its effectiveness:

Oliver:

"Yeah because it was an Italian doctor, it was his wife who had MS. And he did the operation on her, then there was a, it might, when we went there [to the clinic], there was a young lad he was waiting to go in. […] He had, he had trouble, he had trouble walking

Wife: Yeah he walked, yes. As [my husband] said he did the zombie walk didn't he? [Um] And then he was about 35. And we went back the next day didn't we for this consultation, and he just, well we were just amazed.

He were just walking normal.

Wife: He just stood up and just walked straight in.

You wouldn't think there were anything wrong with him."

[Oliver (12), male, 57 years old]

Melissa:

"There were some. It was all about this Professor Zamboni from Italy, that was, has supposedly cured his wife by doing it, he's a vascular surgeon. [Um] There were some testimonials from people that had been abroad.

Okay

[Um] To Poland I think, and there was quite a lot of You-Tube clips of people that had gone in

Oh okay

In a wheelchair and walked out"

[Melissa (13), female, 46 years old]

Experiential information appeared to convince Oliver and Melissa of its effectiveness; they both introduced the topic of CCSVI with an anecdote of the Italian Professor who "cured" his wife with the surgery, and then referred to others who showed improvements after the procedure. Recent research into YouTube testimonies of people with MS who claimed to have had surgery to treat CCSVI found the procedure was overwhelmingly presented as being successful. In order to counter this, the researchers

conclude that healthcare professionals should engage with patients who wish to discuss it (Mazanderani et al., 2013) (explored further in chapter 9). At various stages during the interview, Oliver seemed uncertain as to whether the treatment was successful:

> *"I suppose I've made the mistake of going up and having that operation, but I don't know if it's worked or not. I might be worse, I might not be.*
>
> ***Yeah. Yeah.***
>
> *But it was worth taking that gamble for that"*
>
> *[Oliver (12), male, 57 years old]*

His claim that "it was worth taking that gamble" suggests he felt some desperation about his future and hope about the prospect of curative procedure. Such a context may compel people to try an intervention out of desperation, even with little evidence.

8.3.4. Peer support

In order to gain from experiential information people must identify with it, but this may be difficult if the process causes distress. Mazanderani et al. (2012) described this as a "tension of identity" (p.550) in their analysis of information use by people with MND. They found that this tension was overcome by "the cultivation of a sense of *difference in similarity*" (p.550); as they became aware of the variability of MND allowing them to identify with such information, without as much distress. Analysis showed that participants in this study also developed "a sense of *difference in similarity*" as their awareness of the variability of MS seemed to ease feelings of distress about their future. They also appeared to overcome this 'tension' by seeking out peer support from people similar to themselves. Chris, Donna and Adam all attended support groups that comprised members similar to themselves in terms of lifestyle:

Chris:

"The best bit of it is it's not really people sitting there talk… complaining about their symptoms all the time, it's just a general chat about whatever and [er] and the odd bit about MS. So, it's not like there's a lot of people on their death beds and wheelchairs […]

When you go to the hospital or you go to quite a lot of these [er] [er] talks, particularly the ones in, in [town names] you get so many people who are on their last legs in wheelchairs but to actually see people who are still working and carrying on with a, their life,

Yeah

[Er] And about, about sort of 70% of the people who go to it are still working. It actually gives you quite a bit of hope.[…] You're looking, meeting people carrying on with their normal lives,

Yeah.

That's a lot more hopeful. That's the one thing that I think is missing locally.

Mm. Yeah.

Because you then get a chance to meet other people fairly early on and I, and I think that was one of my big problems was that I only met people in wheelchairs in hospital for the first couple of years.

Yeah.

And [er]

Yes so how did it feel when you met that sort of people?

Well it basically it makes you really, well sort of seeing lots of people in wheelchairs or in hospital makes you think I really don't want to engage with this. [Um] Because you're looking at worst case scenario in the future."

[Chris (08), male, 38 years old]

Donna:

"*And we meet up in the pub, have a drink, go bowling, go to the dogs before they closed the dog track down. Yeah just we do anything, simple things. Go for a meal. I think next time we're meeting up we're going for a curry. [Um] And we just chat,*

Yeah*.*

And occasionally someone will say, "I've been having this, anyone else?"

Right*.*

And so occasionally something to do with MS will come up, but a lot of the time it won't.

Yeah*.*

It's just pure coincidence. But if you're having a bad time there's somebody who will, who'll know they don't know exactly what it's like being you and your MS, but can relate to it. That's the thing. It's every one of us knows that our MS is ours. And it won't be exactly the same as anyone else's but we share an awful lot of symptoms, you know it's a lot in common. And that's really helpful at times. And information, share information as well.

We tend to meet once a month, I think that's the idea [um] and they're just some really lovely people which is good so I've found it helpful"

[Donna (06), female, 49 years old]

Adam:

"Yeah and [um] and the beauty of it was [um] and remains I think, I haven't been to one for a while but, was they're people [um] a lot like you, [um] you know broadly the same age, broadly [um] at the same point in their careers, the extent to which MS affected them varied a bit but not massively, [um] you know most of us were still mobile for example and [um] walking around. And just sort of dealing with the frustrations of it and swapping advice so I think they were the people that you know you could maybe do with meeting fairly promptly after diagnosis. The people I was talking about who are, who are sort of [um] worrying about the same stuff you are worrying about but, but getting on with it. But nor are they sort of [um] you know they're not the people you see when you, when you type in MS onto the internet or whatever and you see people in wheelchairs and what not. […] So I think it was helpful to sort of have that group of people generally in the same place in terms of life and how their disease was affecting them."

[Adam (11), male, 36 years old]

Elsewhere, support groups have been shown to sometimes cause distress and "reinforce feelings of difference and exclusion from normality" (Locock and Brown, 2010, p.1503). Support groups that focus on social activities or lifestyle, rather than being focused on managing MS may ameliorate such feelings.

Hannah and Joanne also discussed how they sought contact with people from similar backgrounds after finding contact with general MS support groups unhelpful:

> *Hannah:*
>
> *"I've been to an MS society meeting where it's just, you know an MS Society meeting and I've felt a bit isolated, just sort of ignored and I didn't feel right being there. I don't know, so I left half way through. I just thought no, and [um] I've not seen other people who look like me, made me think oh maybe this isn't for me. I think I saw one or two but I just thought, I didn't feel included somehow in the whole thing so I just left. [..]*
>
> *Well you can see like, right I, like some people for instance, like the last lady I spoke to she uses a wheelchair, she's had MS for a lot longer than most people I know. But I think she's been isolated with her condition. She's got a big family, she's got four children so I don't think she's you know she hasn't acknowledged the fact that there are other people who use a wheelchair, who are black, who are women, who have families and, 'cos I could see when she did turn up she, you know there were other people that she was looking, "Oh right, it's not just me." I could see it in her face. She didn't have to tell me I could just see it."*
>
> *[Hannah (20), female, 49 years old]*

Joanne:

"And like me being pregnant too, when we were going to have our second child, I didn't know anyone else who sort of had a child […] oh I'd love to have chatted to anyone pregnant person with MS or someone who'd just given birth or something like that 'cos it was, looking back I don't know how I coped sort of being alone almost, do you know what I mean?

***Yeah definitely**.*

Aye. But there is a good support network now, there really, really is like I'm so thankful for it. […] [My friend] at the MS Society has actually set up like a, a young woman's group.

[…] I know what it's like going in there as a young person all them years ago when I was diagnosed and just seeing older people. It was a bloody nightmare. Puts you right off"

[Joanne (18), female, 28 years old]

Participants' accounts show how people with MS may negotiate the 'identity tension' and distress caused by examples of people severely affected by MS by seeking out people similar to themselves, almost rejecting identification with examples of 'worst-case scenarios'. The unpredictably of MS prognosis means that people may live with relatively few symptoms or may deteriorate very quickly. This differs from conditions such as MND, which can be more clearly defined as "a progressive and almost inevitably terminal condition" (Locock and Brown, 2010, p.1499). Thus, it may be more realistic for people with MS to detach their identity from people who appear severely affected by MS.

8.4. Discussion

Analysis of the interview data showed that information provision around the time of diagnosis is lacking and people with MS (or suspected MS) can find this period very stressful. These findings corroborated a recent systematic narrative review into MS healthcare services which concluded that:

"A lack of timely information on diagnosis, living with MS and treatment was found to directly link to negative emotional states such as anxiety and fear. Providing timely and credible information on these topics should therefore be a priority in improving the patient emotional experience." (Methley et al., 2015, p.1852)

The findings presented in this chapter identified some potential factors that may exacerbate this emotional impact and suggestions were made to ameliorate it. The findings demonstrate how more attention could be given to information needs during diagnostic investigations, as participants often recalled feeling confused and frustrated during this liminal stage. People with suspected MS may benefit more from information about diagnostic processes and acknowledgement they may have MS. Some neurologists have reported withholding information about the potential for an MS diagnosis to protect patients (Heesen et al., 2003) but these findings suggest this risks causing frustration if patients feel they are not taken seriously or shock when they are eventually diagnosed.

The way in which participants identified with others with MS appeared to be a crucial aspect aiding or inhibiting adjustment. The lack of information at diagnosis was often filled with information representing particularly active MS trajectories. Linked to the findings in chapter 7, accessing such information can initiate and reinforce a perception that they will be severely affected by MS, and in turn may cause distress, exacerbate symptoms and factor into life decisions. Participants seemed to overcome this 'identity tension' by accessing information that presented a broader range of trajectories, positive examples of managing MS and peer support more relevant to themselves. Some also found that generic information did not apply to them and so they tended to tailor the information they accessed, often seeking out experiences of others relevant to them personally.

With a lack of useful instrumental measures and variability in MS, participants found that their own experience was a vital information source to understand and manage their MS. But conversely, it was also often considered unreliable due to unpredictable trajectories and confusion over causes of symptoms (explored in chapter 7). Experiential information from others also appeared to be valuable for learning about real life practicalities, signposting to services, providing reassurance, sharing ideas, gaining empathy, and understanding the heterogeneity of MS. Most participants appeared to understand the limitations of experiential information and used it in measured and critical ways, but others seemed to consider it as evidence of effectiveness of interventions, which are sometimes expensive and promise dramatic effects. In a context such as MS where prognosis is unpredictable and potentially severe, treatment options are limited and biomedical understandings are incomplete, it is easy to see how alternative approaches may be particularly appealing and how seeing others who appear to benefit from interventions might compel people to try them.

Chapter 9

How do People with MS describe their Interactions with Healthcare Professionals and their Impact on their Relationships?

9.1. Chapter summary

People with MS may consult a broad range of healthcare professionals, including GPs, MS nurses, neurologists, physiotherapists and occupational therapists, speech and language therapists, psychologists, dietitians, social care and continence specialists (NICE, 2014). NICE (2014) guidelines recommend that people with MS should have a "comprehensive review of all aspects of their care at least once a year". This recommendation was evident in most participants' accounts who said they typically had yearly reviews with neurologists. In addition to yearly reviews, the frequency with which participants consulted healthcare professionals varied (some maintained regular contact and others rarely consulted healthcare professionals). MS nurses can act as a central point of contact for people with MS and coordinate care; however, their availability varies widely across the UK (Morse, 2011). Analysis of the interview data reflected how access to MS nurses differs widely; some participants did not have access to one, others had regular meetings with them and many contacted them via a telephone service as and when they needed support.

Although people with MS may consult a broad range of health professionals the data relevant to this chapter was primarily participants' accounts of their interactions with GPs, neurologists and MS nurses. The background literature presented in chapters 1 and

2 showed how despite support for personalised care planning (PCP), it does not appear to be routinely carried out for people with long term conditions (LTCs) or MS. Therefore it was unsurprising that participants' accounts suggest that much of the support people with MS receive from healthcare services appears to be reactive, as they typically consult MS nurses or GPs in response to symptoms and for support during relapses. The unpredictable nature of MS and breadth of symptoms might mean a reactive approach is inevitable to some extent, as it may be impossible to plan for the unpredictable.

This chapter explores the ways in which participants recounted their experiences of interactions with healthcare professionals and identifies behaviours perceived as helpful or unhelpful by people with MS. It also considers whether elements of shared decision making (SDM) and PCP are present or absent within participants' accounts. The sections are ordered in terms of the levels of attentiveness to their individual concerns and needs as described by participants. The interview data are not direct evidence of interactions, but are participants' presentations of interactions and beliefs about how these shaped subsequent actions and perceptions.

9.2. Attentive healthcare professionals

Although there were relatively few examples in the interview data when participants described interactions where healthcare professionals appeared particularly attentive, it is important to note that this is possibly due to the method of data collection. During interviews, participants may relay experiences that were particularly challenging and thus memorable. However, there were some clear examples where participants described encounters with healthcare professionals who appeared to be particularly attentive to their individualised needs and value their input during consultations. Much of the literature focuses on patient input being value-based and healthcare professionals'

being biomedical-based i.e. patients are considered experts in their experience of the illness, social circumstances, their values, attitudes and preferences, whereas healthcare professionals are considered experts in diagnosis, aetiology, prognosis, treatment opinions available and outcome probabilities (Coulter and Collins, 2011, Charles et al., 1997, Morgan, 2008). However increasing access to information and educational resources means that patients, especially those with LTCs, may be knowledgeable about biomedical information regarding their condition, practicalities and treatments (Horton, 2003, Snow et al., 2013). Some of the participants identified particularly helpful instances when their healthcare professionals acknowledged that patients were potentially well informed and recognised their own limitations. Janet considered herself very knowledgeable about MS and felt healthcare professionals varied widely in their acknowledgement of this. She appreciated it when her neurologist appeared to recognise her expertise:

"She [my neurologist] knows I read a lot, so quite often what she does is she fixes it for me to get the last appointment in the clinic and she'll do my appointment and then she'll kind of push my notes aside and say, "Right what research has there been that I need to know about?" And, so she treats me as an expert,

***Yeah**.*

in many ways. Which is great, it's just wonderful that she does that because it really makes you feel respected. […]

So I went in to see this other Neuro, and she had a medical student in with her, [um] we chatted away for a few minutes, and I was saying you know, "Yeah, but what about this?" and "What about that?" And "But isn't that [um] aren't there risks of seizures with that? And I've already got epilepsy, and blah blah, blah." And after about five minutes she turned to the medical student and said, "You see I told them, I told you some of them know more about this than we do."

***Mm**.*

So again that's just fantastic recognition of the knowledge that a lot of patients bring.

[Janet (01), female, 50 years old]

Janet seemed to be forthcoming in displaying her level of knowledge, which may have provided a cue to her neurologist, allowing her to feel comfortable in explicitly recognising Janet's knowledge. But this may not suit all patients, and healthcare professionals may face difficult situations when assessing patients' level of knowledge. Mary also was impressed when her (temporary) GP recognised that Mary might know more about MS than she did:

"She did say, "You probably know more about it than me." […]

Okay, so how did it feel when you're, when that GP said to you, "You probably actually know more than I do about this?"

Because of the way she said it I was quite happy about it. I mean I do, I read extensively and [um] you know I don't think there's an MS publication that I haven't read. [Um] I can't necessarily remember them when I've finished them, but [um] I do know a lot about it. […]

But most GPs don't want to be told things about [er] health, whether it's your health or, you know like you can't go into a GP and say, "Oh I think I've got so and so." They seem to want to establish that themselves. They don't seem to like it if you do that. [Um] And I think that's, this other lady that I saw, I can't even remember her name [um] she just seemed more caring, more you know, more understanding and when she said that it certainly wasn't said in a, [um] a way to sort of belittle me or anything, which is often how I feel when I come away from the doctors, which is why I just don't go anymore, you know."

[Mary (05), female, 61 years old]

Throughout Mary's interview, she reported being largely dissatisfied with her encounters with GPs, to the point where she became reluctant to consult (explored further in section 9.3.1). However, the instance quoted above stood out in Mary's interview as being particularly positive. She identified this simple act of recognising her knowledge as being unusual and perceived it as being more "caring" and "understanding" than other GPs she had encountered. Although she did not explicitly refer to feelings of empowerment, she did compare this approach to her expectation of typical interactions with GPs where she feels "belittled". In Mary's case the act of recognising her knowledge worked to overcome unequal power divides.

Chris described a particularly positive experience when his GP acknowledged the limitations of medical knowledge. After his neurologist appeared unable to treat his recurring migraines and double vision and assessed them as unrelated to MS, Chris sought advice from his GP. As explored in chapter 7, the breadth and variation of MS symptoms leads to uncertainty as it is often unclear whether symptoms experienced are

caused by MS. In Chris's case it was unclear if his migraines and double vision were related to MS. Initially his GP did not appear to know the best course of action or cause, but spent time during consultation looking up information. Chris did not mind that his GP was unsure and was impressed with this approach, not only because his GP's recommendation seemed to be effective, but also because he was "being listened to and being taken seriously". Some healthcare professionals have reported a 'professional reticence' whereby they withhold information because they believe it is unprofessional to burden patients, arguing that uncertainties inherent in medicine can cause unnecessary anxiety for patients and reduce trust in healthcare professionals' abilities (Elwyn et al., 1999). However when Chris's GP appeared to work through the uncertainty whilst looking for a solution in a transparent manner, it was a positive experience.

A few participants described interactions where healthcare professionals seemed to be particularly attentive to issues that were important to them. Ian said that his GP checked in with him regularly and was considerate about his experience of administering medications. Ian felt his GP acknowledged that he does not like taking so much medication and so researched if any adaptions could be made to his regime. He said that she tested his vitamin D levels in order to reduce the quantity of tablets he took and prescribed different sized needles to those the drug company provided to enable him to carry out painful injections more quickly. Ian felt very well supported by this particular GP (whom he compared to others in the practice) and thought they had a good relationship.

These are examples of instances where participants felt their healthcare professionals were particularly attentive to their individual needs and perspectives. In terms of SDM, they are not archetypal examples, because in some instances healthcare professionals

seem to act as ‘perfect agents’ rather than in partnership. When healthcare professionals act as ‘perfect agents’ they make the decision on behalf of the patient, whilst being particularly attentive to the individual’s perspective and consider the best course of action for them personally, rather than a solely biomedical view (Gafni et al., 1998). In some instances decisions did not seem to be made. However, healthcare professionals seemed to acknowledge their own limitations and recognised patients’ expertise; this led to feelings of empowerment. This in turn may be conducive to enacting more equal partnerships, a central facet of SDM.

Considering the data in terms of PCP, there was almost no evidence of its presence, except in Miles’ account. He was unusual in appearing to have experienced an approach that resembled PCP. It was also one of the few examples in participants’ accounts of interaction with a physiotherapist:

"We'd [Miles and his physiotherapist] sat down and had a chat, and it was what did I want to achieve […] I can remember one of my answers was, "I want to be able to walk round my house without using crutches." Which I can do now. […] Yeah I, I have had some pretty horrific falls. And I don't particularly like falling 'cos it hurts [um]. […] I've been attending physio for I'd say six months and yeah I'm, I'm an awful lot better than I was. I think I've had one fall since Christmas. […]

With your, the action plan you put together with the physiotherapist [um] did you, you know, the goals you came up with,

Yeah.

was that you who thought of the goals or was it the physiotherapist, or was it more of a joint...?

It, it was, I'd say 60:40 for me joint. I came up with the majority,

Okay

of what I wanted to achieve.

Okay. And do you think it helps having that plan?

I think it helped at the time."

[Miles (09), male, 44 years old]

Miles's experience appeared to adhere to PCP ideals as he said he collaborated with his physiotherapist in a proactive process that enabled him to set a goal important to him. None of the other participants recalled interactions that resembled this approach and when asked directly about their experiences of PCP, participants were often unaware of the concept and requested further explanation. This reflects wider literature that shows that patients are typically unfamiliar with this approach (Burt et al., 2012, Newbould et al., 2012, Fitzpatrick et al., 2010).

9.3. Feeling "dismissed" and overlooked

There were various examples within participants' accounts where they said information they had brought to consultations was dismissed and times where participants' perspectives appeared to have been overlooked or undervalued. Perspectives being

overlooked were distinguished from feelings of dismissal, as participants felt dismissed when a healthcare professionals seemed to assess information they presented as unworthy of serious consideration, e.g. a healthcare professional might assess (or dismiss) diet change as ineffective and decide not to discuss further. When participants' perspectives appeared to be overlooked, it was not that the relevance of their perspective seemed to be assessed as unworthy of consideration, but more they went unnoticed or seemed to be undervalued. Boundaries between perceived dismissal and overlooking of patients' perspectives are sometimes blurred, but their impacts were distinct in participants' accounts.

9.3.1. Feeling "dismissed"

One of the most illuminating findings was the negative consequences of instances when participants perceived their healthcare professionals as being dismissive about topics they wished to discuss. The term 'dismiss' is contentious as it implies judgements about the intentions of those who dismiss and suggests a use of power which one must have in order to dismiss. However, there were clear examples of this in participants' descriptions of interactions and many participants used this term themselves. In order to clarify the perceived behaviour I am referring to, 'dismiss' is defined as follows:

> "2. Treat as unworthy of serious consideration" (OUP, 2016)

Therefore, when discussing participants' perceived dismissal, I am referring to their perception that a healthcare professional treated information they presented as unworthy of serious consideration. The consequences of feeling "dismissed" varied, but accounts suggest they often had damaging effects on patient-healthcare professional relationships, participants' perceptions and consequent actions to some degree. When participants felt their healthcare professionals were dismissive, many chose to opt out of

further consultations and if healthcare professionals appeared to dismiss approaches outside clinical recommendations, participants were rarely dissuaded and often went ahead without consultation.

Many participants said they decided to stop consulting specific healthcare professionals who they had felt had been dismissive. For example, Chris said that when his he felt his neurologist dismissed symptoms as unrelated to MS, he sought a second opinion from his GP and transferred to a different neurologist. Mary recalled how results from previous tests and the prospect of alternative therapy appeared to be dismissed by her neurologist:

> *"He [the neurologist] wanted to start all the tests all over again, everything, lumbar punctures, everything. And, and was really nasty when I sort of said "Well, you know I've had all these, I don't need to do it again." […]He wanted his own facts and figures and things like that. But he was just so nasty about it and [um] I'd met somebody with MS who had [er], through my sister who'd recommended [um] oh, she wasn't a dietician, [um] but she, oh gosh what was she called? I mean she put me onto quite a strict regime of, of diet and [um] various supplements and so on because she reckoned that I'd got candida very badly[…] And I mentioned this to him and he was just so evil with me about it, that because I'd spoken to somebody I suppose that wasn't a normal doctor, [um]. But I, I, you know a lot of people have supposedly had this candida problem, [um] but he wouldn't even discuss it. Her words were "I was riddled with it." And I used those words and he just laughed at me and said that I was ridiculous or something.*
>
> *[…] And [um] so I just sort of dropped the subject, but I did say to my husband when we came away, he wanted me to have [um] various tests, [er] visual evoked potentials, and all these various things again and I just said to my husband I, I'm not going to go back, I'd rather cope with it on my own than deal with somebody like that."*
>
> *[Mary (05), female, 61 years old]*

When putting forward an alternative theory that a fungal infection (canidida) exacerbated her MS symptoms, Mary felt she was unfairly treated. Her statements that "he wouldn't even discuss it" and "he was just so evil with me about it" suggests that it

was the lack of discussion and manner of communication which upset her, rather than the stance the neurologist took on it. If we consider this situation from the neurologist's perspective, effective communication with a new patient who has proposed alternative theory may be challenging and there may have been good reasons why he requested that tests be re-run. Healthcare professionals may be unaware how their behaviour is interpreted, in this case to the point where Mary felt she would rather "cope with it on her own" than consult this particular healthcare professional again.

Some participants decided not to consult healthcare professionals about specific topics due to previous interactions where they experienced what they perceived as dismissal. Paul described an occasion when his wife asked his neurologist about dietary changes:

> *"And so she [my wife] asked him [about diet] and he basically kind of blew it off as, as not really anything of anything. […] [My wife] mentioned the dairy thing to him and he was like, "No, you know, there's nothing to it." So I just think to myself well if I've got in a proper conversation with him about it, I wouldn't get far. Or he might go so far as to say you shouldn't really be doing that. So you know it's,*
>
> ***Yeah**.*
>
> *the negative aspect unfortunately of speaking to people about it far outweighs the positive aspect. So you just get to the point where you think well I'm not going to bother.*
>
> *[…]I would like to try it [Low Dose Naltrexone, which is unlicensed for MS], but you know, it's one of them.*
>
> ***Have you ever [um] discussed that with a neurologist or doctor or suggested it?***
>
> *I don't think I did. I did contemplate [um] mentioning it to Dr [Name], but I think it was on the basis of, of his reaction to the [um] the dairy question when we asked him that, I think he, I think he's the kind of doctor that will only entertain ideas that are properly licensed or you know have, have massive conclusive evidence that you know all the other neurologists would listen to."*
>
> *[Paul (04), male, 27 years old]*

Paul now chooses not to consult his neurologist over certain topics due to previous interactions where suggestions seemed to be assessed as unworthy of serious consideration. He even stated a concern that if he discussed his diet changes, his healthcare professional may recommend he stopped. The presence of this concern is telling, as it would present a dilemma for Paul as he may find it difficult to go against, or challenge, his neurologist's advice, reflecting neurologists' authority regarding what is judged to be the best course of action. Previous interactions seemed to shape Paul's opinion of his neurologist, as he now understood his role as confined to conclusive evidence based information only.

In addition to people with MS opting not to consult specific healthcare professionals or avoiding certain topics, some participants such as Melissa stopped consulting healthcare professionals more generally due to previous interactions where they felt healthcare professionals dismissed their concerns:

> *"Because of my bad experiences I think I've, I'm now, I'm now frightened to say anything to them really 'cos I don't think I'll get anywhere. I think if I go to the doctor I'll just get brushed off.*
>
> ***Right.***
>
> *So what's the point?*
>
> ***Yeah.***
>
> *That's how it feels.*
>
> ***Yeah.***
>
> *You know. [Um] Which is sad really.*
>
> *[…] I mean even now I'm frightened to phone up about things because I get, I'm frightened of being dismissed. [Um] You know I feel as if they're not really interested in what, what happens and what I say."*
>
> *[Melissa (13), female, 46 years old]*

The extract shows how consultations where patients feel dismissed can have negative implications for how people perceive and use healthcare services more broadly. In Melissa's case, previous interactions seemed to affect her confidence to consult; she felt the need to protect herself from potential situations where she may feel dismissed again and avoided consulting healthcare professionals more generally.

As explored in chapter 2, the theory of chronic cerebrospinal venous insufficiency (CCSVI) is particularly controversial because it challenges the widely understood aetiology of MS, proposing that MS is a vascular condition rather than neurological. Private companies offer vascular surgery in order to treat CCSVI, which may be very appealing for people looking to treat their MS, especially as it claims to have dramatic effects. When people with MS present the prospect of having this procedure during consultations, neurologists are likely to be faced with a difficult situation as it goes against their disciplinary position on the aetiology of MS.

When telling their neurologists about their plans to have vascular surgery to treat CCSVI, Oliver and Melissa appeared to have similar experiences. They both said their neurologists not only dismissed the prospect, but also warned them of its dangers:

Oliver:

"I told the doctor about that [plan to have CCSVI procedure] he just totally dismissed it.

[…] Yeah, no he just didn't want to know. Just, just said, he just warned me about the [er] the risks of dying. Says so many people have died having this operation, which, which they hadn't,

Right.

It was other complications, but he just totally dismissed it. Unless it's their treatment that they're offering they just don't want to know. […]

You, you brought it up during that consultation?

Yeah, yeah I was [er] yeah I told him all about it, and he just well looked at me like I'd got two heads. Just thought, just wasting money, waste of time."

[Oliver (12), male, 57 years old]

Melissa:

"Oh I remember I did go over and see [my] doctor for a routine appointment and I printed all this stuff off from the internet and he just said, "I don't advise that," and that was it.

Oh okay.

He didn't even look at my information so,

Really? So was, you printed the stuff about the procedure

I printed it all off

and the

Yeah. And I said, "I really would like to try this, what do you think?"

Right.

"Oh it hasn't been proven, and [er] it has been quite dangerous for some people 'cos a couple of people have died apparently […]

Mm. But would it have been useful to talk about it?

It would have been, yeah, it would have been useful to, rather than it just be dismissed as being completely pointless I would have liked to have, see that's one thing that I feel as if I wasn't given, by the MS nurse and the neurologist, right from the start, even though you may have to pay for things why can't they just give you the options that, or tell you about things that are available."

[Melissa (13), female, 46 years old]

In the extracts above, both Melissa and Oliver recalled how neurologists warned them of dangers and did not engage in discussion about it. Broom and Adams' (2009) research into attitudes of oncology healthcare professionals towards complementary and alternative medicine (CAM) found that when patients wanted to discuss CAM, many oncologists felt they had to dismiss it and warn patients of potential dangers (of interacting with adjuvant therapies and adverse effects). This was in order to protect and dissuade patients, and show that they do not endorse it. Interestingly, the oncologists acknowledged that their knowledge about CAM was limited, thus warnings about risks

were speculative and based on "the personal belief of the individual consultant" (Broom and Adams, 2009, p.326), rather than research evidence.

We do not know the intentions of Oliver's and Melissa's neurologists. However, their attempt to dissuade them did not succeed; Oliver went ahead with the procedure and Melissa was planning to at the time of the interview. In these cases, perceived dismissal meant participants made the decision without discussion with healthcare professionals, despite wanting to discuss it (as they introduced the topic during consultations). Oliver and Melissa both referred to experiential information they had accessed which showed the procedure to be effective (explored in section 8.3.3). Research into testimonies on You Tube found that the procedure to treat CCSVI was overwhelmingly presented as having favourable outcomes. For this reason, Mazanderani et al. (2013) argued it is important that healthcare professionals do engage in discussion about these types of experiential information.

The examples discussed above demonstrate the damaging effects perceived dismissal can have on patient-healthcare professional relationships and future interactions. Healthcare professionals may be in difficult positions when patients want to discuss controversial or alternative approaches. They may not feel equipped to discuss these approaches, there may be time restraints, or they may feel it is unethical if discussion is interpreted as endorsement to some extent. Here lies a tension between epistemological positioning of evidence based medicine and promotion of patient preferences. Commonly the experience of feeling dismissed by healthcare professionals did not function to dissuade participants, but they often seemed to disregard healthcare professionals' advice and opt out of future interactions. They appeared to opt out of future interactions about specific topics, with individual healthcare professionals or healthcare in general.

9.3.2. Overlooked patients' perspectives

Participants recalled some scenarios where healthcare professionals seemed to overlook knowledge they had gained through their previous experiences. Ian remembered how, whilst in hospital, he told healthcare professionals he was having a relapse (identified due to a specific pain he gets during relapses). But they seemed to ignore this and continued to carry out investigative tests. It was not until a few days later when his regular neurologist got in touch with the hospital that they appeared to acknowledge it was a relapse and provided appropriate treatment. In a condition such as MS where the breadth and variability of symptoms make judgements challenging, it may be particularly important to prioritise information gained from patients' experience of the condition when making assessments. Overlooking such information may result in wasted resources on investigative tests and delays in access to treatment.

James and his neurologist in Italy had worked out optimum dosage of Tysabri (a disease modifying drug) for him specifically. Rather than travelling to Italy every 6 weeks, he wanted to receive infusions locally in UK, but he said that his neurologist seemed to undervalue his previous experience of taking the medication and would not accommodate his tailored regime:

"They refused to entertain the six weekly mark. […] And at this point now I'm 36 infusions [um] into Tysabri. So the chances of me developing PML [Progressive multifocal leukoencephalopathy; a potentially deadly side effect] after 36 infusions, in Italy I'd be known as a "Nonno", I'm a grandfather of Tysabri I've been on it for such a long time. The chances, I could win it back to back weekends the European lottery. I've got more chance of that than really ultimately falling foul of PML. But they refused me

[…] But they're all equally allowed to work as doctors, not just as pencil pushing people in white jackets. As doctors and use their good sense and help evolve the practice to get the very best from the medicine, with the very least amount of risk. That practice isn't, they don't take that practice here in the UK because when I asked them about it they told me, "No, no we're not happy to go beyond the licence. We're not willing to take that responsibility."

So my argument was so you're not prepared to be a doctor. You're not actually a doctor, you're just a person that sticks a needle in my arm every four weeks and [um] if anything goes wrong, well you know please see the licence, the terms and conditions. That's not healthcare. Because inside healthcare, there's the word "care." Care means due diligence as far as I understand it."

[James (02), male, 33 years old]

James's tailored regime of six weekly intervals was a lower dosage than advised by the license guidelines of every four weeks (Biogen, 2016), which meant side effect risks would be lowered; James wanted to avoid overtreatment which (in his case) adhering to the guidelines would result in. There are likely to be many differences within any patient group and so research evidence about treatment effectiveness does not necessarily apply directly on an individual level. For this reason it has been argued that decisions ought to be tailored and responsive to individuals, their illness experiences and reactions to treatment (Gray, 2004). James believed that his neurologist was undervaluing his personal experience in favour of protocol guidelines, which meant he could not receive optimum treatment in UK and this led to negative opinions of his neurologist. At this point it is worth highlighting that a request to go against guidelines is likely to be contentious. Based on James's account, we do not know the way in which

his request was presented, the reasoning behind his neurologist's decision or additional restrictions that may have applied.

Participants described some interactions where it appeared that their personal preferences were not taken into account during interactions. Throughout her interview Sadie commented upon the good relationship she had with her GP, who had supported her through a difficult past with an abusive partner. However, Sadie had concerns that her GP appeared to overlook. She disliked the amount of painkillers she regularly took and paramedics attended to her after she accidently overdosed. They suggested she visit a pain clinic at the local hospital to learn ways to manage pain without medication. Sadie requested a referral from her GP, but believed she has now forgotten (and is reluctant to ask again). Throughout Melissa's interview she described various interactions where her preferences seemed to be overlooked or unexplored. She had a strong anti-drug preference. Her neurologist alongside her husband and friend thought she ought to start disease modifying drugs (DMDs) and so she complied and started treatment. At no point was her preferred option to avoid DMDs explored. After four months of treatment, she independently made the decision to stop DMDs. After making the decision to stop, she felt "abandoned" by the medical profession, because she said there was no follow up and nothing else was offered. She now uses alternative therapies instead.

9.3.3. Reluctance to express views

During the interviews, when participants said that they felt that their perspectives were overlooked or dismissed, they were probed further and asked how they felt about putting forward their perspective. Many participants referred to the reluctance they felt at the prospect of expressing their view during consultations with GPs and neurologists.

Even those who assessed their healthcare professionals to be attentive and their relationship with them as good described feeling uncomfortable, as Ulrike put it: "In one way it's easy because he's very receptive, but in another way he sort of, you know, you do feel a bit like you're talking to God". Oliver's neurologist's warnings about the procedure to treat CCSVI contradicted the research he had carried out, but he did not feel comfortable challenging this advice:

> ***"Okay and was it easy to bring that [prospect of CCSVI procedure] up during the consultation?***
>
> *No, no. Not really. No because [um] no, no he just a matter of talking it, he was just writing something down, I'd just told him what was, what we were planning to do and he [er] said really it's a waste of time. […]*
>
> ***And did you [um] did you challenge your neurologist at all? Or…***
>
> *No 'cos he's not very forthcoming with any, any constructive comments. He don't really tell you anything.*
>
> ***So it wasn't very easy for you to feel comfortable enough to say, "Actually I've read …"***
>
> *Yeah probably I should have challenged him on that*
>
> ***Well I don't think, I mean it's creating an environment isn't it, where you feel you can.***
>
> *Feel you can yeah, because obviously you know he knows more than I do, so I can't really tell him his job."*
>
> *[Oliver (12), male, 57 years old]*

Sadie would have like her GP to investigate the possibility that her back pain was caused by something other than MS, but did not feel comfortable suggesting this:

"I'd like a scan on my back, just, for just for peace of mind maybe but then someone said "well tell her [GP], just tell her".

Yeah.

Except I don't want to be seen to be telling her her job.

Right yeah.

She's the expert not me so."

[Sadie (17), female, 50 years old]

There is some conflict within Oliver's and Sadie's accounts; although they questioned their healthcare professionals' assessments, they also made reference to them being better placed to make assessments than they were when they said "he knows more than I do" and "she's the expert not me". This contradiction suggests that although they may not have full confidence in their healthcare professionals' assessments, they seemed to adhere to traditional understandings of patient and healthcare professional roles, where healthcare professionals are the purveyors of knowledge and patients passive recipients. When explaining their reasoning not to voice their opinions, they both stated that they did not want to tell healthcare professionals how to do their job. This reflects how cultural understandings of patient and healthcare professionals' roles can shape behaviours, and in this case restrict opportunities to put forward opinions. Barry et al.'s (2000) research which compared patients' agendas before primary care consultations with what they discussed during consultations, demonstrated how much of patients' agendas are often "unvoiced" during consultations:

"In consultations, patients can seem only partially present, with only limited autonomy – that it, to make requests but not to suggest solutions. Outside consultations patients are more fully present: as socially and contextually situated, thinking, feeling people, with their own ideas on their medical condition and opinions and possible criticisms of medical treatments" (Barry et al., 2000, p.1249-50)

It appears that Oliver and Sadie are "only partially present" during consultations they described in the extracts above, because outside the consultation they had "own ideas on their medical condition" and "criticisms of medical treatments", but said they did not voice these.

9.4. Imposed consumerism

The analysis of interview data suggests that when people with MS are eligible for DMDs, it is often assumed (by both neurologists and patients) that they will start them. Although many people with MS manage their condition without DMDs, no participants described instances of healthcare professionals raising the possibility of choosing not to use them. Some participants who chose not to start DMD said they made the decision against neurologists' recommendations. Currently there are eleven DMDs available for people with relapsing forms of MS and numerous therapies are in development (MS Society, 2016). There are two classes of DMDs categorised as having 'moderate efficacy' with less severe side effects and 'high efficacy' with more severe side effects. The class of DMD recommended is dependent on the level of disease activity (Scolding et al., 2015). Within these classes, there are different types that are comparable. The decision about which DMD to start is complex due to a lack of trials that directly compare the different types of DMDs (Scolding et al., 2015), DMD trials which are still in progress and increasing numbers of licensed DMDs (Heesen et al., 2011). Their effectiveness differs between individuals and at different stages of one person's MS disease progression and so there are no standardised markers when and which DMDs are appropriate (Kachuck, 2011). In addition, people may consider different methods of administration, storage requirements and potential side effects that vary in severity, when deciding which DMD to start.

9.4.1. Lack of advice from healthcare professionals

The majority of participants who started DMDs described interactions that were consumerist in style when making the decision about which type of DMD to start. Their accounts suggested that neurologists and MS nurses acted as information sources, and patients made decisions independently, without their guidance. Participants recalled how they were given leaflet-based or DVD information, signposted to an online decision aid 'MS decisions' (MS Trust, 2015) or open days run by pharmaceutical companies, but were given no direction and were requested to make a decision about the type of DMD independently.

> *Adam:*
>
> *"But there's lots of information, lots of setting out the options [um] that you have for the various different types of injection and things, and websites out there to help you through the decision making [er] but no-one gives you a view and sometimes you know you just want someone who [um] knows a thing or two about these things to kind of give a recommendation at least [um] or what they'd do in your position sort of view. But they won't, and they didn't."*
>
> *[Adam (11), Male, 36 years old]*

> *Laura:*
>
> *"They just said, "These are the options, here's the information. Here's what they do. Here's how they work." They have like sample you know injection pens and stuff, [um] they let you have a look at, feel at, [um] and they say, "Right away you go. Have a think about it. Get back to me whenever you think whichever one you want to take." And I, as I say I still find that completely mind boggling. Like my father's got a heart condition and his doctor tells him he has to take X, Y, Z at morning, noon and night or whatever, and he takes them and he is, he stays well. So I just can't understand how it, why there isn't [um] why they wouldn't recommend a drug."*
>
> *[Laura (19), female, 30 years old]*

Kamila:

"I actually asked my MS nurse "Can you please tell me which one is the best to go for?" And she said, "No I can't because [um] it has to be your decision. It's unprofessional and I can't make that decision for you." Which [um] you know, I do understand the decision is still mine but a little bit of help"

[Kamila (21), female, 29 years old]

In the archetypal consumerist interaction patients have high levels of decisional power and healthcare professionals have low (Stewart and Roter, 1989), as patients independently make decisions and use healthcare professionals as information sources. On a surface level, this style is present in participants' accounts about DMD decisions. However, it is more of an 'imposed consumerism', as accounts suggest that people with MS typically did not opt for this style of interaction[9], but felt it had been imposed it upon them. There are clear examples of this in the extracts above; Adam and Kamila said requests for advice were denied and Laura found the approach to be at odds to what she would normally expect. This 'imposed consumerism' reflects an underlying power dynamic which is the reverse of a consumerist approach, as healthcare professionals appear to have high levels of power when deciding on the style of interaction and people with MS have low; rather than being empowered to take control of the decision, they felt instructed to make the decision independently.

[9] None of the participants indicated that they opted for this style of interaction. Of the examples where a type of DMD was chosen, 9/12 experienced this 'imposed consumerism', two said they were not involved in the decision and one was unclear. But in all these examples, no participant said they actively opted to make the decision independently.

9.4.2. Dissatisfaction with decisional responsibility

Although some participants did not have a problem with this approach, most were dissatisfied and would have appreciated more guidance. Participants often interpreted perceived lack of advice as a lack of support:

> *Adam:*
>
> *"But the disease modifying drugs: I remember being I suppose infuriated [um] at the unwillingness of, of, of [um] medical teams to give a view I suppose. I know that there's probably reasons behind it. […] I suppose it feels like I have been on my own more than I'd have expected with that sort of choice. That sort of decision rather.*
>
> *[Adam (11), Male, 36 years old]*

> *Joanne:*
>
> *I remember she was kind of like, [um] "It's up to you what you do with them. You can pop these here notes in the bin, you can put them under the stairs, or you can read them. It's completely up to you." They're sort of really kind of patient led type of thing; do you know what I mean? They don't like force things on you. But like [um] I suppose it's good in a way but no at the time I just thought, "Oh they didn't really care what I go on," blah, blah, blah. Do you know what I mean?*
>
> *[Joanne (18), female, 28 years old]*

> *Kamila:*
>
> *"Regarding the decision which one to go for I had oh you could say, you could say I had no support from, from the healthcare professionals"*
>
> *[Kamila (21), female, 29 years old]*

Rather than feeling empowered to be involved in the decision, the extracts show how people with MS can feel unsupported and alone in making this decision without advice from healthcare professionals. Joanne referred to the discourse of engaging patients in healthcare as a positive, but noted how it did not suit her at the time and instead it felt

like her neurologist and MS nurse "didn't really care". These examples provide support for criticisms of consumerist interactions, that they can lead to loss of healthcare professionals' expertise and responsibilities during decision making processes (Coulter, 1999). Some participants' accounts suggested that they felt unprepared to make the decision. They said they had very little experience and knowledge about MS and DMDs compared to their healthcare professionals, especially if newly diagnosed:

> *Laura:*
>
> *"It's not something I'm trained in or I know anything, obviously I know what I know through reading but [um] I think it's quite a hard thing to leave in the hands of a patient. And something that I struggled with at the time I think […]*
>
> *Such a, a big decision for someone to make, especially when they've just gone, undergone diagnosis which can be quite hard to get your head round, to be asked then to make a decision on a drug which about a, an illness which you know absolutely nothing about, it can be quite, I think it's quite, I think it's quite a big ask."*
>
> *[Laura (19), female, 30 years old]*

> *Joanne:*
>
> *"He [neurologist] doesn't spoon feed you anything.*
>
> ***Right.***
>
> *Which is sort of what I wanted really because like I'd just been diagnosed and I was so young as well, but like you did want someone to go, "Aye, go on the once a week one. That is brilliant." Do you know what I mean?"*
>
> *[Joanne (18), female, 28 years old]*

Similar to other participants, Laura and Joanne made this decision soon after diagnosis. As explored in chapter 7, the time around diagnosis was found to be emotionally distressing for people with MS as they often envisioned their uncertain future. This emotional distress alongside inexperience of DMDs and MS seemed to make participants feel less able to choose which DMD to start.

It was not only those recently diagnosed who felt ill prepared to make this decision independently. For example, Adam decided to start DMDs after his condition worsened:

> ***"How did it feel having to go away and make the decision of which type, and make sense of the information? Did you feel prepared to do that?***
>
> *[Um] No I don't think I was. I was just, I think at the time I just felt frustrated that they weren't just telling me which one to go on.*
>
> ***Yeah***
>
> *[Um] Or offering a recommendation, the fact that it was sort of, you know these are the options, here's something to help you through your decision. It was all a bit [um]…, and we just sort of… a steer would have been good. I remember sort of thinking, even it was sort of you know, "Don't worry about the five of them, or however many there are, it's you know, in your position it would, I reckon this or this." [Um] But there's none of that, no. I don't know if that's a policy thing around DMDs or, or what but there was just no [um] yeah lots of information but no advice, if that kind of makes sense. I think I was kind of just hoping for a bit of, a bit of advice."*
>
> *[Adam (11), Male, 36 years old]*

Elsewhere in the interview, Adam said he was particularly upset about the prospect of starting DMDs because he considered it a sign that his condition was progressing. It may be common that decisions about DMDs are made during particularly distressing periods. Patients' feeling of vulnerability, dependency and emotion can work against adoption of a purely consumerist role (Lupton, 2004) as they may not feel empowered to make decisions independently. Conversely more choice does not always mean more control, particularly in contexts where options are numerous and patients feel overwhelmed and lack confidence in making the decision (Schwartz, 2005). In general, when healthcare professionals prescribe medicine, patients do not choose the type of medication. Thus, it is unlikely that patients would be familiar with making such decisions, and many of the participants seemed perplexed and surprised as to why they seemed to be expected to make this specific decision alone, especially when they would have preferred guidance. Many participants felt their neurologists and MS nurses could

have provided more guidance without making the decision. For example, Adam previously suffered with anxiety and depression, he felt his healthcare professional could have guided him away from DMDs which have this listed as a side effect.

9.5. Discussion

Unsurprisingly, participants said they were satisfied with consultations where they felt healthcare professionals tended particularly well to their perspective and addressed issues that were important to them personally. Much of the literature refers to patient input as value-based, but many of those interviewed were knowledgeable about the medical aspects of MS and when their healthcare professional seemed to acknowledge this, they often felt respected and empowered. In addition, in the few examples where participants said their healthcare professionals seemed open about their knowledge limitations, this was received well and did not seem to raise concerns about healthcare professionals' abilities.

Consultations where participants remembered healthcare professionals as being less attentive to their perspectives appeared to have detrimental effects. Instances where participants thought healthcare professionals were dismissive of information they introduced, often seemed to damage their relationships, to the point where some participants opted out of consultation about specific topics, with particular healthcare professionals, or healthcare services more generally. Stewart and Roter (1989) described four types of patient healthcare professionals relationships, three of which referred to widely cited concepts of paternalism, consumerism and mutuality (or SDM) (further explored in chapter 1). But they also described a fourth type 'default' where interactions result in patients opting out of care:

"In this case, a patient may drop out of care completely because of failed expectations, concluding that all doctors are incompetent, insensitive, or otherwise inadequate. Most often the physician will be unaware of the reasons for the loss of this patient; the patient will simply not come back." (Stewart and Roter, 1989, p.21-23)

There were echoes of this 'default' relationship in some of the interviews. Some participants opted out of future interactions when they felt dismissed by their healthcare professionals and found it difficult to challenge them. Thus, in such instances healthcare professionals may not be aware of the existence of problems in their relationship (in line with the description above). Participants' reactions were more measured in comparison to Stewart and Roter's definition, as few decided to "drop out of care completely" and no participants said that "all doctors are incompetent"; however, they did decide to opt out of certain consultations after experiencing what they perceived as dismissal.

Situations where patients want to discuss untested approaches are likely to be difficult for healthcare professionals. SDM ideals aim to integrate clinical expertise and patient values with the best available evidence (Stacey et al., 2009). However, tensions arise when considering the best way to balance these elements. The way in which these types of information are prioritised is a considerable biomedical ethical dilemma, especially when they come into conflict (Beauchamp and Childress, 2009). Some critics of EBM have warned that prioritisation of evidence based information may result in other types of information being devalued during healthcare interactions (Sandelowski, 2004). Understandably healthcare professionals may consider it unethical to discuss unproven strategies and theories that conflict with widely accepted aetiological understandings, and arguably the act of dismissing such topics is not only professional, but ethically sound too.

Yet participants' accounts suggest they would like to discuss these topics and perceived dismissal can be damaging to their relationship and future interactions, whatever the professional motivation may be. Participants' accounts suggest they did not tend to dissuade participants from following a course of action. Instead, they followed these courses, without useful discussion with healthcare professionals. Perhaps topics that are worth serious consideration during consultations should be guided by preferences of patients rather than presence of research evidence i.e. patients decide which topics are worth serious consideration. But, the question remains whether it is within a healthcare professional's remit to engage with and discuss approaches that conflict with their understandings of the evidence.

There is little literature on the practicalities of SDM examining the prospect of disagreement between patients and healthcare professionals and appropriate conduct in such scenarios (see chapter 1). Charles et al. (1999) addressed the prospect of disagreement briefly, suggesting that when agreement is not reached patients have to consult another healthcare professional if they wish to pursue it. But this is not the case when patients are deliberating whether to try interventions other than those healthcare professionals do not gate keep, those alternative or complementary to regulated approaches. In a context such as MS, where there are numerous unproven strategies patients can access, it may be useful for them to have detailed discussions with healthcare professionals.

Creating settings where patients feel comfortable putting forward their perspective is one of the challenges of SDM and it is argued that traditional paternalistic roles are a central barrier to SDM (Elwyn et al., 1999). These findings support this argument as they suggest that traditional roles appeared to inhibit participants in expressing their views. Barry et al.'s (2000) study identified the presence of patients' 'unvoiced

agendas'; patients' perspectives that are not discussed during consultations. In follow-up interviews they found that patients and healthcare professionals were still reluctant to address fuller agendas during consultations, even when there were subsequent problems related to 'unvoiced' agendas. In order to enact SDM, healthcare professionals may need to instigate and encourage patient involvement because the traditional unequal patient-healthcare professional power positions mean patients often assume a passive role by default, and thus have less power to effectively shape and negotiate the their roles and relationships with healthcare professionals and they may be unaware of any alternatives (Stewart and Roter, 1989, Stiggelbout et al., 2012). As Barry et al. suggest:

"Both doctors and patients need to change their behaviour to improve outcomes. We believe that by changing doctors' views and behaviours, patients can also be facilitated to change." (Barry et al., 2000, p.1250)

But the problem is not as simple as healthcare professionals eliciting patients' perspectives. Patients' preferred levels of involvement have been shown to vary according to their background and changing circumstances (Coulter and Ellins, 2009). In order for healthcare professionals to respond to patient's preferred level of involvement, they need to first assess it. How they assess this is an area of some contention; some argue that healthcare professionals should be explicit and ask patients to state their desired levels of information and involvement (Towle and Godolphin, 1999), whereas others argue that explicit reference to levels involvement is intrusive and that patients' desired levels of involvement may be accessed intuitively from non-verbal signals (Elwyn et al., 2000). Whether such assessments are realistically feasible and the way in which they are carried out is unclear.

Frequently, decisions about which type of DMD to start appeared to be carried out in a consumerist fashion, i.e. healthcare professionals provided information and people with MS made their decision independently without advice from healthcare professionals. Interestingly, participants' accounts suggest that this approach was not opted for, but imposed upon them. Many participants felt unsupported and unprepared to make this decision independently, and would have liked more advice from healthcare professionals. At the time of the interviews, the Association of British Neurologists DMD guidelines recommended people with MS are well informed and involved in decisions about DMDs:

"Decisions to start or stop treatment, or to perform MRI for diagnosis and management, should recognise the central importance of patient choice; patients should be fully informed of relevant facts and uncertainties before making a decision in discussion with their treating neurologist." (ABN, 2009, p.4)

The description reflects the 'imposed consumerism' that many participants described, because "the central importance of patient choice" was recognised and they were "fully informed" about DMDs. However, guidelines also recommend that decisions are made "in discussion with their treating neurologist" which suggests a collaborative approach, rather than making decisions independently, without neurologists' guidance. This suggests there may be other influences that make neurologists and MS nurses reluctant to provide advice or make recommendations. Ziebland et al.'s (2006) study explored treatment decision experiences of women with ovarian cancer. They found that, similar to this study, women who reported having to make treatment decisions without oncologists' advice were surprised and felt burdened. Ziebland et al. argued that healthcare professionals should engage in discussion about such decisions with patients and explain why they are being asked to make a decision independently:

"Being asked about their preferences can surprise or shock women. Healthcare professionals need to explain about the clinical uncertainty and how individual values and preferences may relate to treatment decisions […] If the rationale for the choice is not explained and communicated in a clear and timely fashion the choice can seem […] 'bizarre'" (Ziebland et al., 2006, p.361-4)

Similar to Ziebland et al.'s study, participants were unsure why neurologists and MS nurses appeared to take this approach. Some believed it was due to policy or the presence of pharmaceutical company competition that made healthcare professionals reticent to show bias for specific drugs. It could be that there are so many DMDs with comparable effectiveness that healthcare professionals really do not feel they can provide advice nor have the time to do so. Nevertheless, as these pressures could be apparent in any prescribing activity, it remains unclear why this approach is taken for DMDs in MS specifically. There may be valid reasons why neurologists and MS nurses are not involved in the decisions about which type of DMD to start, but accounts suggest these were not explained to patients.

Two key themes 'feeling "dismissed"' and 'imposed consumerism' represent particular problems for interactions between healthcare professionals and people with MS. These areas warrant further examination to understand contributing factors and ways in which they may be overcome. In the final stage of the research these problematic areas were presented to consultant neurologists during structured qualitative interviews and the findings from these are presented in chapters 10 and 11.

Section four
Findings from interviews with Consultant Neurologists

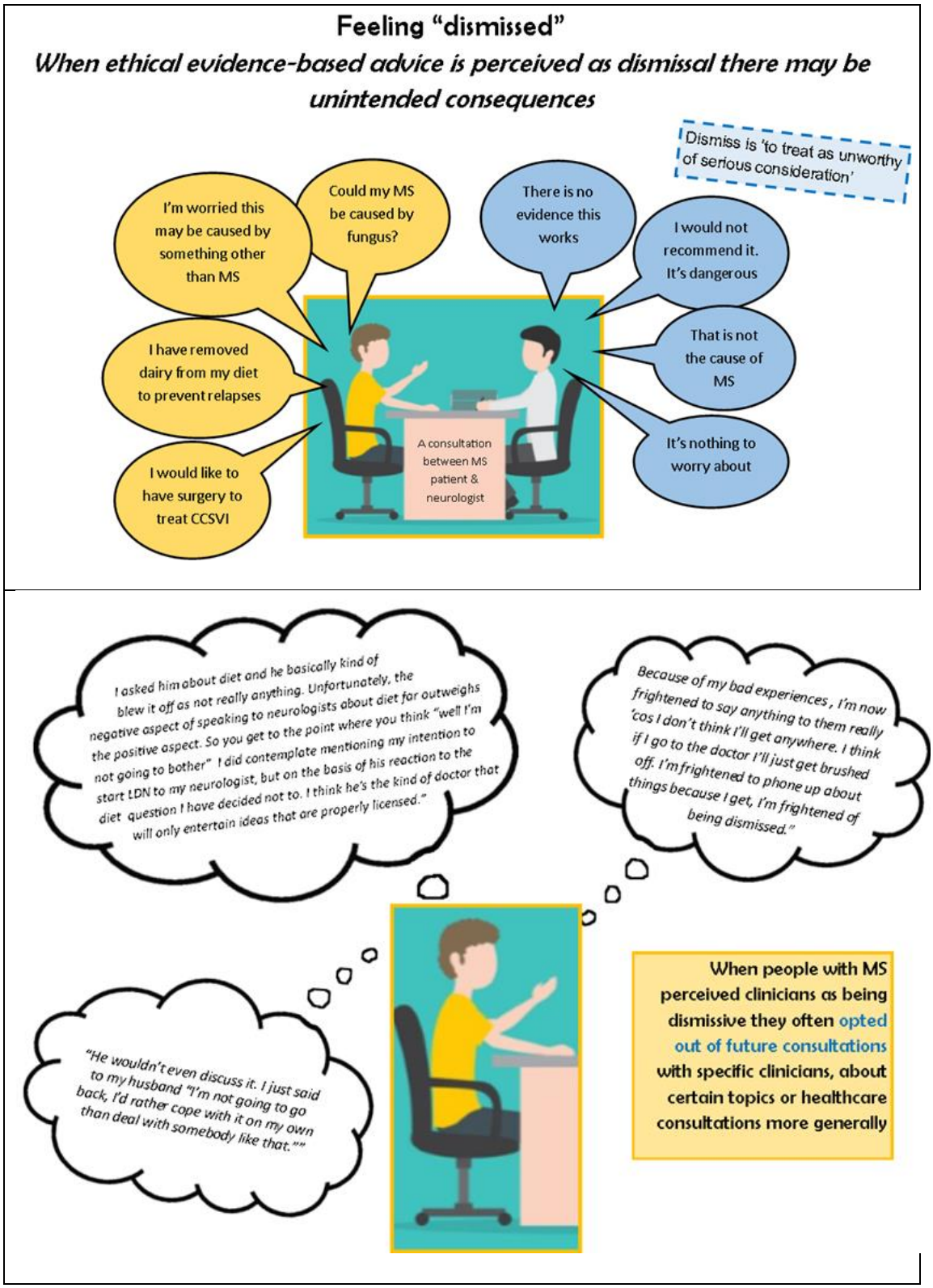
Chapter 10

Findings from Interviews with Consultant Neurologists on the Emergent Theme of Feeling "Dismissed"

As discussed in chapter 9, the findings from MS participants' interviews suggest that sometimes when participants felt healthcare professionals were dismissive of information they brought to the consultation, this led to them opting out of future consultations with specific healthcare professionals, about particular topics or with healthcare services more generally. We may assume that healthcare professionals do not aim to appear dismissive, but discussing complementary and alternative medicine (CAM) (interventions not licensed nor endorsed by the medical profession) can pose a particular dilemma of how to balance respect for the person's interest in the topic with regard for the evidence.

As described in chapter 3, six consultant neurologists took part in structured qualitative interviews across the UK between August and November 2016 in which they were presented with an infographic (fig.10.1) of the dilemma described above and asked to discuss their interpretations of, and the best ways to overcome, the dilemma.

Figure.10.1 Feeling Dismissed Infographic



Neurologists recognised this dilemma and gave a broad range of responses – with no clear solutions – on how to overcome it in current healthcare systems. Analysis of the interview data revealed potential reasons why neurologists may be perceived as dismissive during such discussions and identified barriers that inhibit accommodation of patients' preferences to discuss certain content.

10.1. Disdain towards those promoting unsubstantiated claims

Presentation of this dilemma evoked strong reactions in most of the neurologists during interviews. They had varied views depending on the type of CAM, however there was a clear consensus that some alternative treatments were particularly unethical because they were judged as ineffective, expensive and sometimes dangerous. There was a perception that people with MS were particularly vulnerable to their appeal, because MS is incurable and potentially progressive with limited mainstream treatment options available. Neurologists interviewed typically perceived those offering such approaches as "charlatans" who targeted people with MS due to their perceived vulnerabilities.

> *"Some of the things my patients endure are terrible, awful, scandalous and criminal" CN02*

Although neurologists thought their strong discouragement on ethical grounds could be interpreted in different ways by different patients, they recognised the potential that they may appear dismissive.

> *"Dismissive is a difficult word, isn't it?*
>
> ***Yeah, it's a strong word**,*
>
> *[Er] It, I can imagine consultations which [um] some of the parties would want to say were dismissive and other parties wouldn't, and [er] and a third-party observer might or might not describe as dismissive." CN04*

> *"Yes, I understand the dismissive thing I can see why people think they get dismissed […] I can see how they have unintended damaging consequences for patients, because the patients just get turned off." CN01*

Some of the neurologists interviewed did provide ideas about how to engage in discussions about alternative approaches. Most of these methods were based upon providing patients with contextual information. For example it was suggested that around the time of diagnosis, neurologists could talk to patients about the large amount of unsubstantiated theories and approaches that exist for MS and equip them to be conscious of this when encountering information independently. One neurologist said that he provides patients with a historical context of theories which have been popular in the past in order to give perspective on ones which are currently popular. The same neurologist suggested that patients could be educated to critically appraise information they access and referred to a guide developed by the charity 'Sense About Science' that provides information for people with incurable conditions about how to assess the reliability of information presenting dramatic claims (Sense about Science, 2013). How such guides are received is unknown and research would be required to examine their suitability. There is evidence that well designed written health information is useful in improving health literacy, particularly alongside guidance from healthcare professionals (Coulter and Ellins, 2007, Synnot et al., 2016). However, this refers to health information, rather than guides to assess reliability of information – as referred to above.

10.2. Prioritisation of content

There was consensus that in order to discuss alternative approaches without endorsing them, complex and lengthy discussions were required. Prioritisation of more clinically

relevant content was identified as a central motivation behind avoidance of in-depth discussion about alternatives:

> *"If you have time it is easier to take the patient seriously. But if you have half an hour and you have to get through a particular agenda because that's the purpose of the consultation and someone then brings something which, to a clinician feels a bit left of field,*
>
> ***Mm***.
>
> *[Um] then then the clinician feels challenged because they've got an agenda that they feel is the right and proper use of that consultation." CN01*

> *"There is not much time in a brief consultation to address not only the important questions, the issues about drugs, dealing with their symptoms, but also the concepts of the care system around them that are very relevant to help them [to] not feel as though they're being dismissed." CN05*

> *"Neurologists are specialised and they do not have time to discuss crackpot ideas" CN03*

Furthermore, comprehensive discussions about alternatives were seen as time-consuming, not only within the consultation but also outside. Most thought the majority of neurologists would not have the knowledge-base to discuss CAM confidently nor time to maintain knowledge about popular theories.

> *"It is very difficult to keep up to date with proper evidence, without having to keep up with unsubstantiated evidence as well". CN03*

Some said the level to which they engaged in such discussions depended on the level of harm the specific intervention may entail. If judged to be relatively benign, they were more willing to appear neutral. If judged to be harmful (physically and/or financially) or used instead of clinically effective approaches, then they thought it was more important to actively discourage it.

10.3. Neurologists' professional stance

Neurologists varied in their willingness to have comprehensive discussions about CAM. Some thought it was not part of their role to accommodate in-depth discussion on these topics, whereas others judged it as an important topic to engage with if the patient brought it up. Those who felt it was not part of their role said it was important to assert their professional stance on approaches that are not clinically proven. Their justification was that discussing such content may be seen as endorsement (to some extent). Illustrating the dilemma, one neurologist reasoned that appearing dismissive and accommodating such discussions were *both* dangerous:

> *"To collude with a patient for doing something that doesn't work, that is expensive, that has risks, because it's what the patient wants is also unethical.*
>
> ***Yeah***.
>
> *And there are situations where you see that quite a lot. [Um] Investigations are a really good example. You know that somebody [er] hasn't got a problem that they they they've got,*
>
> ***Right***,
>
> *but you say to them, [um] and you know they've had a scan three or four years ago, [um] and you say, "Okay well lets [um], let's re-scan you just in case something has changed." And you give them a dose of radiation as a consequence. Now that's,*
>
> ***Right okay***,
>
> *Now that's colluding with a patient, around something they want […] so colluding or dismissal are both dangerous.*
>
> ***Mm. Yeah***.
>
> *And so for the doctor this is a much better ethical place than the one that says, "Well if it's what you want, maybe you should go off and try it." CN01*

In the extract above, use of the term "collude" suggests an action which is secretive or a purposeful deviation from a professional standard. This reflects how some healthcare

professionals may feel uncomfortable in having such discussions. The suggestion that from a professional perspective it is better to err on the side of appearing dismissive to avoid any endorsement of unregulated approaches illustrates the perceived seriousness of endorsing untested approaches.

Some explained that, as neurologists, they are publically funded to provide specialist neurological advice, rather than to discuss CAM. Thus, such discussions may be considered inappropriate use of resources and must be justified on a professional level.

> *"I can't really discuss in detail whether something is good or not good if it's not gone through a proper process of showing whether it's good or not.*
>
> ***Yeah***.
>
> *So, it's quite difficult to discuss it because we're going outside professionalism. It's not our expertise.*
>
> ***Yeah***
>
> *Well it's not what we're expert for and we would be criticised if we discuss things that we aren't trained to discuss." CN05*

> *"It's difficult to justify resourcing discussions about alternatives which have no sensible rationale" CN01*

When patients asked about alternatives, one neurologist said she asserted her professional position by explaining she could only discuss treatments recommended by NICE guidelines and referred them to their online NICE guidelines for MS treatment. Although this approach may seem dismissive to some extent, it may work to clarify to patients what they could expect from their neurologist.

Several neurologists said they aimed to have comprehensive discussions about CAM with those who were interested in pursuing it. One thought neurologists could appear dismissive if they focused too much on sharing their expertise and, in doing so,

overlooked motivations behind seeking such approaches. Most agreed that ideally they would explore these motivations. This would enable neurologists to consider the information patients accessed and understand their rationale. Drawing on this exploration, neurologists could then explain any potential limitations of the information, gain understanding of the patient's perspective or potentially consider methods (such as mindfulness or counselling) to alleviate any distress acting as a motive. However, most neurologists interviewed said this was an ideal case and reiterated that time limitations act as a barrier in taking such an approach.

One neurologist interviewed did appear to devote time to ensuring he could have comprehensive discussions about CAM. He subscribed to an MS related magazine (that mainly features content about CAM) in order to keep up-to-date with the latest trends in this area. He also said that if a patient brought up something he was unaware of, he would let them know that he would go away and learn about it in order to discuss it properly. When asked how he managed this in terms of time limitations, he explained that he gets behind schedule and had no solution to overcome this.

> *"These conversations take a hell of a long time.*
>
> ***Yeah***.
>
> *And you get behind and that's very difficult.*
>
> ***Yeah***.
>
> *[Um], [er] and I don't have a solution for that, sometimes I get behind. [Um] And you know these aren't the majority of consultations, but they are a significant part of what we find ourself doing." CN04*

Although he considered comprehensive discussion about CAM was an important part of his role as a neurologist, it did not appear to be feasible to do this within his schedule.

This provides further support that accommodation of such discussions is potentially unsustainable.

When exploring solutions to this dilemma, some neurologists suggested that their specialism meant that other healthcare professionals such as MS nurses or GPs might be better placed to accommodate such discussions.

> *"Whether as neurologists we should be doing it or someone else should be doing it, I don't know.*
>
> ***Yeah***.
>
> *But certainly, there's no reason for us not to, ideally you don't need to be as specially qualified as we are to do this, and, and the MS nurses or other people with an interest in the illness could do it" CN02*

> *"I'm not sure that your neurologist is the person that should be talking through a lot of saggy crackpot ideas out there. And I, I don't think, I think when they're coming to you you're trying to first of all diagnose it, trying to find out if there are any treatments you can give to them.*
>
> ***Yeah***.
>
> *We're not really there as a counsellor." CN03*

However, the same issue of time limitation and prioritisation of clinically relevant content may apply. It has been shown that GPs are overworked and under-resourced (Hobbs et al., 2016) and access to MS nurses varies widely throughout the UK (Morse, 2011). One neurologist suggested that each healthcare profession prioritises particular content in consultations:

"I think we don't do well as an MS community is co-ordinate between nurses, physicians and allied health professionals, a message that can be ribboned appropriately.

Right.

And when you're in a clinic, for example that is [er] tiered into a specific mandate, like disease modifying therapy clinic,

Mm.

But then there's another clinic called the symptom management clinic, and another one that is you know a bladder clinic, they become compartmentalised in certain case, so if you start mentioning things outside of it right, it becomes difficult to balance because of the priority set aside because of the time and the mandate of the clinic.

Yeah.

So it's a very difficult thing with constrained resources be able to adjust appropriately to make everyone satisfied." CN05

He suggested that discussion about CAM is overlooked because it is unclear which healthcare profession ought to facilitate such discussions that potentially conflict with mainstream medicine (and thus their professional stances).

10.4. Patients' role

Some of the neurologists talked about the patients' role in interactions. They thought patients often feel emotional and hopeful about CAM interventions and have decided they would like to try them before they consult their neurologist. Some thought that this emotional investment meant that patients typically looked for approval and so were more likely to interpret neurologists as dismissive when they did not endorse the treatment, regardless of how this stance was presented.

"If fundamentally you [the MS patient], you've come in because you're very excited about a new technique and you want to have something positive back from the person,

Yeah

the clinician, and the clinician knows about the technique and what's been written about it, and thinks it's valueless. [Um] Then that is always going to be a sticky interaction" CN04

"It's an impossible situation. And you sometimes will receive anger, and there is nothing one can do because when people are frightened they will be angry.

Mm.

And so just because you (interviewer) link in your research bad comments against a doctor, that doesn't actually mean the doctor is to blame.

No.

It's just they're looking for a blame because there isn't a treatment. That's where all this comes from. There's no treatment. CN03

One neurologist also talked about how the way in which CAM is introduced by patients may cause differing responses from neurologists. She thought patients who asked a neurologist their opinion about CAM would elicit a more open response than those who said they intended to try it.

Some neurologists suggested that lack of agreement was inevitable, as those patients interested in CAM were guided by a different set of principles about the meaning of knowledge than those underpinning mainstream medicine.

"They [patients interested in CAM] would see this [professional opinion] as nerdy piffle and narrow. They want a world in which there's more drama and dramatic medical breakthroughs" CN04

The context of an MS patient is very different to that of the healthcare professional, as they are the ones experiencing the illness and facing an uncertain and potentially

debilitating future. Thus, the hope of "dramatic medical breakthroughs" could be particularly appealing. Some neurologists believed that reaching agreement in such cases could be unattainable. Whether patients interested in alternatives are guided by different principles in reality is unknown. But if neurologists perceive they are, this may act as a difficult starting point for useful discussion when faced with such topics during consultations.

Although a distinction between patients interested in CAM from patients who were not was apparent in neurologists' accounts, it is unclear whether this reflects reality of patients' reasoning. From a healthcare professional's perspective, their training and professional context means mainstream medicine and CAM are clearly distinct. However, it seems unlikely that patients would typically use the same distinctions and some may be unaware of the differences. The argument that some patients cannot be reasoned with because they are guided by differing principles that conflict with those of mainstream medicine may be somewhat presumptive and act as a barrier to engaging in useful discussion if reasoning is deemed futile.

10.5. Discussion

In order to overcome this dilemma, the neurologists interviewed agreed that *ideally* they would explore patients' motivations, work through their reasoning and identify whether they could use other means to help alleviate any issues. But there were differing opinions about the extent to which they ought to facilitate discussions about CAM, with some suggesting it is part of their role to engage in such discussions and educate patients, and others arguing that this was unprofessional and beyond their remit as specialists in neurology. In light of the findings presented previously in chapter 9, we might conclude that it would be most helpful if neurologists took the former approach,

yet such approaches appear unrealistic as various barriers make comprehensive and responsible discussions challenging.

Cribb and Gewirtz (2012) argued that discourse around user-involvement and user-professional-partnerships is one of oversimplified functionality that overlooks the complexity of social relationships and implications on moral identities. They described the tension between collaborative relationships and the potential for patients and healthcare professionals to disagree:

"[T]here are inherent tensions that arise from shared decision-making. For example, in some cases the patient's preferences might seem irrational or unreasonable to the professional […] In these circumstances, something has to give, and often, in practice, either the professional has to veer towards paternalism in how they frame things so as to steer the patient in what they see as the right direction, or they have to allow the patient to do things that the professional thinks are not in their interests and might even harm them. A further complicating factor is that, although the decision is supposed to be a joint one, the professional is frequently still accountable both legally and morally. So if a treatment exacerbates the patient's symptoms, the doctor cannot simply turn round and say 'that is what you wanted'. The doctor will still feel responsible for, and up to a point is legally responsible for, preventing patients from doing things that are bad for them and for getting them to do things that are good for them." (Cribb and Gewirtz, 2012, p.513)

Cribb and Gewirtz's interpretation alludes to the impossibility of partnerships when patients' opinions conflict with the professional view. This tension was apparent in neurologists' accounts as they felt strongly that certain alternative treatments were unethical and that it would be unprofessional to discuss them. How such conflicts are managed appears to be overlooked in much of the SDM literature (Labrie and Schulz, 2014). Charles et al. (1999) refer to this briefly and conclude that when agreements are not made, patients would need to consult another healthcare professional if they wished to pursue their preference.

But the implications of this tension differ somewhat in context of patients' interest in CAM, where healthcare professionals do not act as gatekeepers (i.e. patients do not need permission to access it). This brings us to another point made by Cribb and Gewirtz (2012): they argued that literature on user-involvement is one-dimensional and depicts power being transferred to patients; the patient becomes more activated and empowered. But they argue that in reality involvement comes with various burdens; one of which is normative expectations of *how* to be involved. If we consider this in terms of the findings here, patients may (at times) exert autonomy in choosing medicine, but this choice is limited to a list of those that are licensed or recommended. Exerting preferences for CAM seems to be less well received and in such contexts, healthcare professionals may feel less obliged to foster collaborative styles. Some may suggest (as some neurologists interviewed did) that CAM – by its definition – does not come under health service provision. Nevertheless, arguably it is an aspect of healthcare more generally, as it affects wellbeing and thus is relevant to healthcare discussions. The data from both interview stages (with people with MS and neurologists) suggest there is a lot of CAM and openness to trying it. Such interest has led some to argue that the content of healthcare consultations ought to facilitate discussions about CAM when required by MS patients (Heesen et al., 2013, Mazanderani et al., 2013).

Elsewhere in the broader literature, others have recommended ways in which healthcare professionals can engage in more open, encouraging and non-judgemental discussion about CAM (Tasaki et al., 2002, Schofield et al., 2010). One literature review of CAM discussions during oncology appointments developed these guidelines:-

"(1) Elicit the person's understanding of their situation; (2) Respect cultural and linguistic diversity and different epistemological frameworks; (3) Ask questions about CAM use at critical points in the illness trajectory; (4) Explore details and actively listen; (5) Respond to the person's emotional state; (6) Discuss relevant concerns while respecting the person's beliefs; (7) Provide balanced, evidence-based advice; (8) Summarize discussions; (9) Document the discussion; (10) Monitor and follow-up." (Schofield et al., 2010)

But when considering these guidelines in light of neurologists' views, they seem unrealistic. There are specific practical implications for professionals' roles during SDM that are not addressed in much of the literature (Cribb and Gewirtz, 2012). These were apparent in neurologists' accounts. Those interviewed all identified how time limitations, lack of communication training and prioritisation of other content act as barriers against such discussions. The guidance – detailed in the subheadings quoted above includes 44 pointers, which although comprehensive – is unlikely to be realistic in practice in neurologists' consultations with people with MS.

Putting time and resources aside, the recommendations above may be useful in fulfilling the needs of patients already interested in CAM. However, none of the studies advising best ways to engage in discussions about CAM, explore the potential that healthcare professionals' engagement might be interpreted as endorsement. The way in which healthcare professionals can engage in non-judgemental ways whilst still communicating their professional stance (which by definition may judge the approach as not worth pursuing) remains unclear.

Conclusions drawn previously in chapter 9 suggest that neurologists ought to accommodate discussions of what patients judged to be worthy of serious consideration, in order to allow useful discussion and avoid patients opting out of future consultations. But interviews with neurologists suggest that responsible accommodation of such

discussions in current healthcare is not realistic largely due to resourcing and lack of clarity about the best way to have such discussions responsibly.

There are strong arguments for and against considering CAM as legitimate content for medical consultations. Findings in chapter 9 demonstrated that some choose to opt out of future consultations because they have felt dismissed when posing prospects of CAM. There is confusion about whether it is appropriate content for healthcare consultations: on the one hand, by its definition CAM lies outside mainstream medicine; yet on the other hand, it has effects on a patients' health and wellbeing.

If engaging in such content is deemed appropriate, then there are ramifications which need addressing. Clarification about whose role (which healthcare profession) it is to facilitate these discussions; investment in training medical professionals in how to have these conversations and acquire knowledge base and how this will be justified is required. If engaging in such content is deemed inappropriate, then consideration needs to be given to other sources where patients gain advice on such approaches and how limiting specific content fits into patient centred care perspectives. Patients would also need clarification about the limitations of the content they can expect to discuss with healthcare professionals. The dichotomy between mainstream medicine and CAM may be obvious to those medically trained, but the distinction is likely to be less obvious to the average patient.

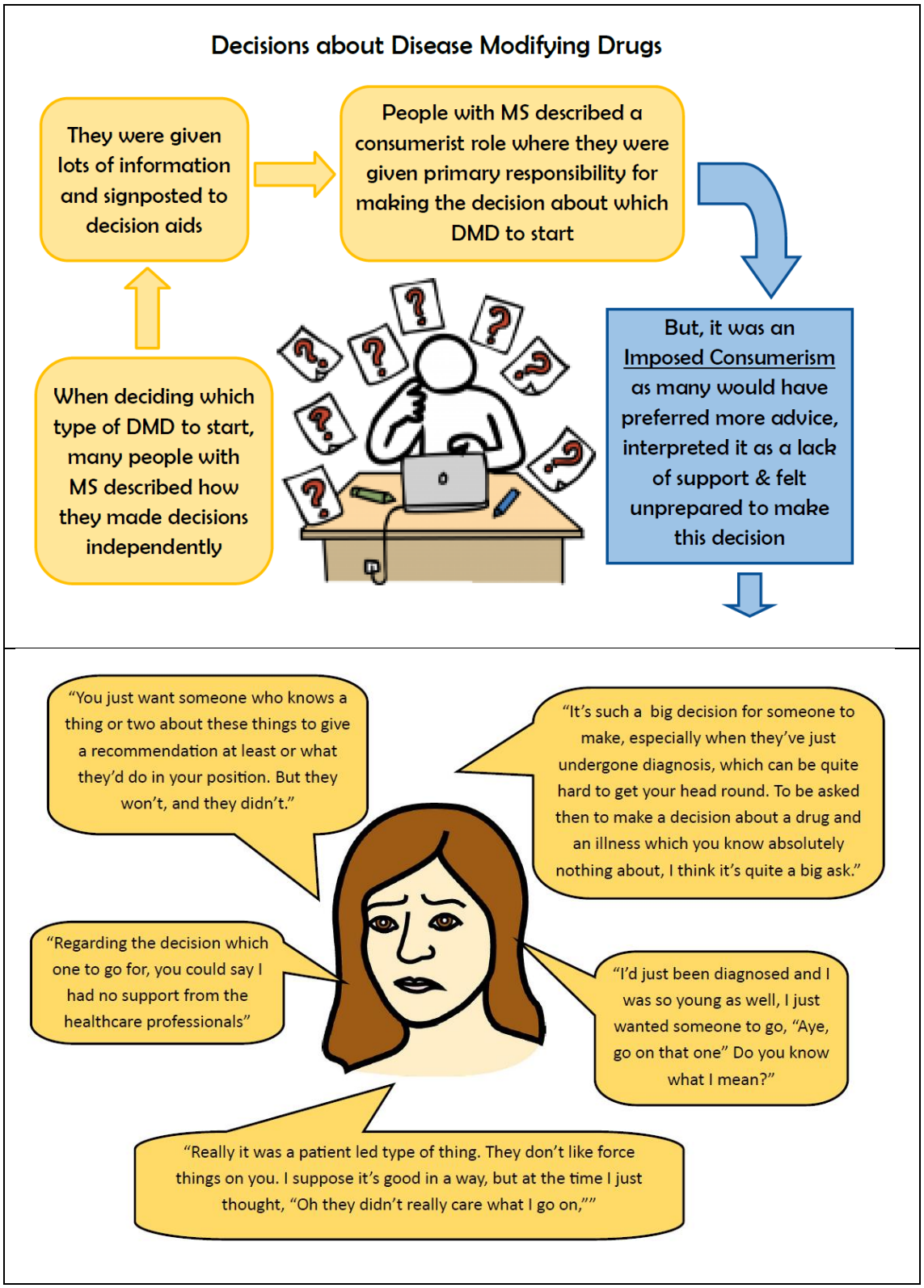
Chapter 11

Findings from Structured Interviews with Consultant Neurologists on the Emergent Theme of 'Imposed Consumerism'

11.1. Introduction

'Imposed Consumerism' is a concept developed from findings presented in chapter 9 to describe a common pattern in MS participants' experiences when deciding which disease modifying drug (DMD) to start. Most participants said that their neurologists and MS nurses expected them to make this decision independently. Participants typically said they were surprised to be given this decision, did not understand why healthcare professionals seemed unwilling to give advice and they subsequently felt unprepared, overwhelmed and burdened by the decision. The term 'imposed consumerism' was created, because although patients have the power to make this decision (akin to a consumerist model of patient-doctor interaction), they did not feel they could choose their decisional role, and decisional responsibility was described as unwelcome – i.e. imposed upon them. As described in chapter 3, six consultant neurologists took part in interviews between August and November 2016, in which they were presented with this finding in infographic form (fig11.1) and discussed their interpretations, potential solutions and barriers/facilitators for these.

Figure.11.1 Imposed consumerism infographic



11.2. Recognition of the problem

The neurologists recognised patients' preferences for involvement in decisions about DMDs as an important and sometimes contentious issue. Some referred to academic literature in this area and said they had had previous discussions about this topic with colleagues. Although many of the neurologists appreciated that preferences for involvement differ, some of the neurologists seemed to imply that most MS patients would be satisfied with making the decision independently.

> *[Describing how she poses the choice to MS patients]*
>
> *"You've got a choice out of these three. Off you go and read about it, and then you can come back to me,"*
>
> ***Mm***.
>
> *Okay. And the majority of people, that is thought to be the best way. Now there are some very opinionated people who will want to be more in charge and there is some much more passive people who will be uncomfortable with it" CN03*

In the extract above, patients described as 'opinionated' and 'passive' are positioned as adjuncts to the 'majority'. However, feedback from patient and public involvement members further supported the 'imposed consumerism' finding and although qualitative methods cannot be used to generalise, it is worth noting that the pattern was very common in interviews with people with MS as none of the participants indicated that they opted for this style of interaction. Of the examples where a type of DMD was chosen, nine out of twelve experienced this 'imposed consumerism'. Of the remaining three, two said they were not involved in the decision and one was unclear. But in all these examples, no participant said they actively opted to make the decision independently. Another neurologist suggested the finding was subject to interpretation. He thought that the situation might be considered 'imposed consumerism' when framed

negatively, but 'avoidance of paternalism' when framed positively. If focusing on healthcare professionals' actions and intentions alone, these may be described as 'avoiding paternalism'. However, 'imposed consumerism' is an interpretation of patients' reactions, emerging from analysis of MS participants' interview data. It refers to how participants felt about interactions in the context of this specific decision. Neurologists' responses to the infographic suggest that although many were aware of some of the contention surrounding patient involvement, they seemed less aware that patients often did not welcome this decisional responsibility and felt burdened by it.

11.3. Contextual elements leading to 'imposed consumerism'

11.3.1. Clinical equipoise

During the interviews, neurologists reflected upon the reasons why patients are often asked to choose a DMD, without guidance from neurologists or MS nurses. All neurologists agreed that the main reason was clinical equipoise. They explained that although there are many different types of DMDs available, research evidence shows that their levels of effectiveness within subgroups are comparable; therefore, in their clinical judgement, there often is not a best course of action. Consequently, the factors affecting the decision – such as administration or storage methods – were based on patient preference.

> *"So, drugs are identical, the side effects are identical,*
>
> ***Yeah***.
>
> *The efficacies are identical but the time, the day and the week in which you take them is a bit different." CN06*

"The decision-making tools just encourage it on how the different drugs are given, and what suit's you from a lifestyle point of view, nothing to do with how effective they are or the side effects. Because they're the same.

Yeah, yeah.

You know do you try a paracetamol or do you try and ibuprofen? Which drugs do you try? They, you know certain drugs will just work the same and there isn't an answer." CN03

The analysis suggests clinical equipoise was a key reason why neurologists do not provide guidance during this decision. Neurologists' views that they cannot offer guidance may reflect a perception that their input is focused on clinical advice, rather than facilitating decision making. When discussing decisions about DMDs, Heesen et al. (2013) suggest that:-

"[i]n cases difficult to decide on, PA [patient autonomy] might be misused by health professionals as a possible strategy to avoid responsibility" (Heesen et al., 2013).

None of the neurologists said this was a factor motivating their *own* actions; however, it was suggested that some neurologists may prefer patients to decide, due to lack of confidence in discussing the decision. This was regarded as unprofessional, but understandable, given the reality of potential inexperience and time pressures.

11.3.2. Aims for best practice

Neurologists also cited their aims for best practice as a key motivation for giving patients decisional responsibility in this context. Using terms such as 'patient centred care' and 'shared decision making' (SDM), neurologists described how they aimed to involve patients and some contrasted this with paternalism, which was judged to be outdated and bad practice.

"You don't want to be paternalistic. […] We don't belong to either a generation or part of the world where that is either acceptable or we actually like doing it. So we always like people to be in charge" CN06

"What's happened is that [er] neurologists have a body, as a body have decided to err on the side of non, not, what you might call positively non-paternalism" CN04

"You know we are criticised heavily if we're biased.

Yeah.

You know paternalism is the word that we're told is, is what in the old day's doctors would tell their patients what to do. Nowadays patients are much more in control." CN03

As discussed in chapter 1, a move away from paternalistic models of care has been driven by advocacy for increased patient involvement. Healthcare professionals aiming for good practice may aim to increase patient involvement wherever possible. Thus, in contexts of clinical equipoise, it may seem appropriate and reasonable to suggest patients make decisions.

Some neurologists thought patients ought to make the decision: firstly, because of the possibility of side effects; and secondly, due to the belief that patients are more likely to continue with medication if they choose it.

> *"If you [as the clinician] take ownership of it [chose the DMD] because they're in a distressed state at that time point they may not thank you two years down the stream when they look up the data and at that stage they realise that, actually they hadn't realised that this drug was, had this side effect and that drug, if they get the side effect from drug A that they weren't really realised was worse than in drug B[…] We're not just there to make them feel good at that moment in time when they're choosing their drug, we're making sure that they feel comfortable that they've made a decision over the time they're taking that drug, when they're out of that difficult phase. […] It may not have been the wrong thing to make it a bit of a difficult time, if downstream they don't regret their decision. I don't know that making everybody happy all of the time is good medicine" CN03*

> *"We always like people to be in charge and it goes a long way in compliance as well. So I find that more people are likely to comply with your treatment if they've been involved in that decision" CN06*

In the extracts above, the neurologists thought it was particularly important for patients to be involved in choosing a DMD. Interestingly, phrases such as "take ownership" and "be in charge" reflect how some people may consider decision making responsibilities as dichotomous, i.e. either the patient or the clinician make the decision.

One neurologist suggested that patients were required to understand information and make the decision in order to fulfil informed consent requirements. However, when this argument was posed during subsequent interviews with other neurologists, they disagreed and thought patients' preferences for involvement could be accommodated without compromising informed consent. There was agreement that patients' preferences for involvement differed and that some were more likely to welcome decisional responsibility than others. However, they all thought the ways in which involvement preferences could be assessed was unclear and so tended to initially expect patients to make the decision as the default.

11.3.3. Pharmaceutical influence

During interviews with people with MS, participants were asked about potential reasons why they thought healthcare professionals seemed reluctant to give advice. The MS participants did not suggest any of the reasons discussed above – and most said they did not know – but some thought that neurologists and MS nurses were being careful not to favour one pharmaceutical company over another (see chapter 9). There were mixed responses from neurologists on this topic. One suggested this did factor into motivations to appear impartial:

> *"Well I think this is, as I say this is, from an evidence based view thought to be the best way that the doctors aren't imposing biases, because of course you can imagine that, that people can be said, "Well Doctor so and so must be using, in this drug company's pocket, he's always telling them to give that drug and," so this is about unbiased [um] treatment, and this is about what healthcare systems feel is best for patients as I said. CN02*

None of the other neurologists interviewed initiated discussion about such concerns, but they were all asked whether an ethical stance not to favour one pharmaceutical company over another affected their reluctance to provide guidance. In direct response to this question, neurologists said they did not think such concerns influenced them much and that impartiality shown by neurologists was more likely to be motivated by the DMDs having comparable effectiveness.

"No. I mean no, not at all. I think, I'm not, if I thought something was better for somebody then I'm obliged to tell them" CN02

"I think there is a bit of that.

Yeah.

But another way of saying the same thing is that if there's no good evidence that drug A is better than drug B why pretend that there is?" CN04

Neurologists did not discuss this topic in depth, but interestingly those who elaborated further did appear to be conscious of potential concerns about pharmaceutical influences and the need to appear impartial because of this. One neurologist said he warned patients that information DVDs on DMDs could seem biased and have "*an element of salesmanship and propaganda about it"*. This indicates that he was conscious of perceived bias and felt the need to provide information with a caveat. In doing so, he introduced an element of distance between his own stance and that of the company producing the information. Other neurologists spoke more generally about how professionally they should not show bias; one noted the variability between centres in the type of DMDs prescribed that led to criticism of bias and another referred to the importance of not showing bias, despite receiving hospitality from pharmaceutical companies.

"And there might be one centre who's giving all drug A and another centre was giving all of drug B. Now clearly those patients aren't that different. They were equal. So clearly one neurologist was sending all their patients down one end, and, and this was not patient led.

No.

That, that was heavily criticised." CN03

"Well of course all have been fortunate in some way or other of accepting hospitality from these companies, but we wish to remain professional and completely uninfluenced by that

Yeah.

And we'll be rigid and stubborn about that." CN02

Although analysis suggests that their impartiality is primarily motivated by the comparability of DMDs and aims for best practice, this is in a context where they are also conscious of the danger of appearing biased towards pharmaceutical companies.

11.4. Solutions and barriers

During interviews, neurologists discussed ways in which 'imposed consumerism' could be avoided and the barriers inhibiting such solutions. Various solutions were proposed, including: working through the decisions with patients; assessing and responding to patients' preferred level of involvement; and trying to reduce the perceived burden of the decision. These are explored further below.

11.4.1. Working through the decision

Some of the neurologists discussed the prospect of them taking a more active role by helping patients to consider how values and preferences factor into the decision and potentially making the decision on the patient's behalf if appropriate. These approaches would emulate the mutual approach of SDM, or 'physician as a perfect agent' (Gafni et

al., 1998) where decisions are made by a healthcare professional who considers the patients' values and preferences and makes a decision based on these. However, similar to barriers identified when discussing the theme 'feeling "dismissed"' (see chapter 10) all the neurologists said that comprehensive and useful discussions to prevent 'imposed consumerism' were inhibited by lack of resources, in terms of their communication training and time limitations.

> *"Do you want a doctor who is good enough, sometimes makes mistakes, but is empathetic? Or do you want a doctor who gets things right, but is less empathetic? The reality is, with limited resources, this is a choice that has to be made. […] I mean I would not underestimate the skill level required in the conversation to do that.*
>
> ***Yeah***.
>
> *You know particularly for, I mean I've been a consultant for twenty years. [Um] I have never done a communication skills course. […] It takes time to get [er] high levels of competence with regard to the underpinning knowledge base. And then you have to develop your communication skills, and you have to do that in a context [um] of very busy clinics.*
>
> ***Mm, yeah***.
>
> *And so it's not terribly surprising" CN01*

The neurologist quoted above suggested that neurologists do not receive the communication training required to facilitate SDM and put this down to resourcing. She also implied that there were time limitations during 'very busy clinics'. Other neurologists also suggested limited time during consultations made comprehensive conversations difficult.

"[During consultations] I talk about frequency. Do you travel a lot? Do you have access to a fridge? Do you have a real preference in terms of wanting to take a pill or an injectable? Do you have children in your house?

Yeah.

So, all these things are things that I discuss, but in the end, it's more the, an opportunity to flag to the patient the issues surrounding the drugs, […] because the details are too difficult to grasp in, in a short consultation. And then giving the patient with that foundation the ability to go and probe further with regards to those options I've outlined. […] I then refer them to websites,

Yeah

to [er] delve further into the nuances. Just because we're limited by time." CN05

The neurologist in the extract above appreciated the decision was difficult. For this reason he thought there was not enough time to deliberate in consultations and instead aimed to "flag" to patients important aspects to consider when making this decision independently.

11.4.2. Accommodation of involvement preferences

Efforts to increase patient involvement is not an issue in itself, but problems arise when patient involvement is encouraged (or imposed) indiscriminately in contexts where more nuanced approaches might be required. Neurologists' views as to whether they should provide advice differed widely, with some more willing than others to provide advice and make decision on patients' behalf. One neurologist said that he sometimes initiates discussion about involvement preferences at the point where it is apparent there is a decision to be made.

"I, sometimes I say, "People vary a lot in how they like to take these decisions, and there are some people who want to go away and look at everything on the internet, and the other people who want us to take the decision, and if you really wanted to do that we can do that."

Yeah.

"But what's important is we [er] we help, we support you to take a decision that feels the right decision to you." CN04

This initial explicit engagement in the topic of involvement preference appeared to be unusual, as it was not evident in the other neurologists' accounts nor was it apparent in analysis of MS participant interview data.

All the neurologists interviewed said they tried to assess and respond to preferred levels of involvement (to varying extents). However, how they did this was not clear. Consistent with findings in chapter 9, the neurologists said that they first set a context where the patient had decisional responsibility by initially giving information and instructing them to make the decision.

"But if you have to be seen to be going through a fair process [by giving the decision to patients], then if the patient comes back "What would you do Doctor?" Most of us would feel comfortable saying, "Well my experience probably in your shoes it would be one of these two, or that one."

Mm.

And then you feel more comfortable. But you couldn't start off by saying that.

No.

That would be such… that would be going against what's, what's recognised as being right for patients [um] from all the evidence." CN03

Based on the reasons detailed above in '11.2.2 Aims for best practice', the neurologist in the extract thought patients ought to make the decision in the first instance. Similar to other neurologists interviewed, she said she would only provide input later, if the patient

had had a period of time deliberating and asked for guidance. One neurologist interviewed was more rigid and felt there were some instances when she could not decide on patients' behalf.

> *"The injectable DMT's, there are three different ones which you know you will choose them based on which company you like, type of thing.*
>
> ***Right.***
>
> *In situations like that I will not take a decision, [er] then the patient will have to, irrespective of whether they like it or not." CN06*

In the two extracts above, the neurologists equating patients making the decision with best practice (as discussed in section 11.2.2) seems to make them reluctant to be involved in the decision. The extract suggests they consider decisional responsibility dichotomously, as they talked about either the patient or the healthcare professional making the decision. However, findings presented in chapter 9 suggest that MS patients do not necessarily want healthcare professionals to make the decision *for* them, but they did want more discussion and advice (see chapter 9).

11.4.3. Assessing preferred levels of involvement

In addition to deliberating how (and whether) to accommodate involvement preferences, neurologists also discussed how to gauge them. In reference to DMD decisions Heesen et al. (2007a) recognised the need to be aware of involvement preferences:-

"Certainly, not every patient wants to share every medical decision, but we think at least patients should make their preferred interaction model explicit, i.e. if they want to share the decision or want the doctor to decide. We think it is essential that a shared decision making process based on validated evidence-based information should be available for those who want it." (Heesen et al., 2007a)

Heesen et al.'s suggestion that "patients should make their preferred interaction model explicit" positions patients as responsible for making their preferences known, but this may be unrealistic; analysis of interviews with people with MS showed they are often reluctant to put forward their perspective (see chapter 9) and this reluctance has also been identified in broader literature (Barry et al., 2000). As healthcare professionals typically lead consultations, in order to assess patients' preferences, healthcare professionals often need to work to elicit them. However the best way to do this is unclear; some SDM theorists argue involvement preferences should be explicitly addressed during consultations (Towle and Godolphin, 1999), whereas others suggest that is too intrusive and that intuitive assessments are more appropriate. This ambiguity was also evident in neurologists' accounts as they discussed their uncertainty over the best approach and found it challenging.

During interviews, one neurologist said he tries to intuitively assess patients' involvement preferences from the first consultation. He then adapts his approach accordingly.

> *"But I do, I'm aware that I have this conversation with them in different ways, depending a bit on my gut feeling about what their decision-making style is. But I'm, so, but I completely accept it's very much a gut feeling thing,*
>
> ***Yeah***
>
> *and it might therefore be being influenced by all sorts of prejudices I bring to the table, CN04*

Despite talking candidly about his approach, he recognised a potential weakness that it was based upon his intuition and influenced by his own prejudices. Another neurologist discussed using personality tests.

"There was a personality questionnaire, it was a very passive versus pro-active that had a five-point score,

Okay

and that patients could fill in and then they could bring it in to the clinic and you could tell what personality type they were. And we did talk about whether we should be doing this so that we could assess what type of decision making process our patients were." CN03

The extract above suggests that gauging patients' involvement preferences is a challenge already recognised and discussed amongst colleagues. None of the other neurologists interviewed suggested ways to assess involvement preferences and although the two suggestions posed (in extracts above) differed from one another, they both entailed inexplicit or indirect assessments, as they would infer involvement preferences (either from personality tests or behaviour during consultation).

11.4.4. Reducing decisional burden

Some of the neurologists thought MS patients feel particularly burdened by this decision due to the perceived seriousness of the decision. They thought patients may feel concerned about making the 'wrong' choice. Neurologists talked about various ways in which they tried to "depressurise" the decision to enable patients to feel more comfortable about making it.

"If I feel that somebody is finding the decision burdensome when we're conversing about it, I will have the conversation in which is aimed at trying to de-pressurise the situation.

***Yeah**.*

And one of the things I might say as part of that is, "Don't feel you're committed to this [um] forever,"

Yeah

"By choosing to start this. If you hate it we can change to something else, or we can stop,"

***Mm**.*

It's not a fixed decision." CN04

"So I think that there has to be a context that's built that's appropriate when discussing drugs. It doesn't mean that the discussion of drugs has to be generically altered, I think it's how it's presented at the time it's presented. Cos if a patient is newly diagnosed with MS,

Yeah

and they haven't even come to grips with that, then giving them an entire [er] list of choices for which medicines may be appropriate for them, should they qualify for MS criteria probably is not the most effective time because they're just trying to understand [er] what's happening to them, and adjusting their feelings, their ideas, their functions, what their expectations are." CN05

"You can pre-empt these concerns by explaining to them that the only way that, that they, I think that the reason that they worry is because they make the wrong choice at the end of the day. And so one needs to explain why actually it doesn't really matter

Right

which one they have" CN02

Some neurologists said they told patients the reason they have a choice is because the DMDs are so comparable in terms of effectiveness and so there was no *wrong* option as such. Some also said that they would let the patient know that they can later stop taking, or change, a DMD if it does not suit them, with the aim of making the decision seem less permanent, and therefore, less serious. There was some appreciation that the timing of the decision had an impact and one neurologist thought it was unfair to expect patients to make a decision soon after diagnosis or within a short time frame. The approaches that neurologists suggested seem reasonable, but contrasted with findings from MS participant interview data, which suggested little effort was made to reduce the burden of the decision. Most MS participants said that they faced the decision soon after diagnosis; some were given little time to make it and none understood reasons why they were being asked to make the decision. Although this discrepancy may simply be because the MS participants were unlikely to have consulted the same neurologists as those who were interviewed, it also highlights an opportunity to ensure MS patients choosing a DMD are informed about these aspects of decision context.

In summary, patients could be given some pointers in order to reduce the burden of the decision and to enact their involvement preferences. Based upon these findings, box 11.1 summarises the information that healthcare professionals could communicate to people with MS who are choosing a DMD.

Box 11.1: Information for people with MS choosing Disease Modifying Drugs when clinical effectiveness is comparable

It's up to you how and whether you make this decision. Some people would like to make this decision independently; some would like to share this decision with healthcare professionals and/or friends and family; and others would prefer the neurologist or MS nurse to make the decision on their behalf. All of these options are fine and the MS nurse or neurologist can accommodate your preferred level of involvement. You may choose to delay making this decision until a later date. In this case, a discussion with your neurologist or MS nurse about potential outcomes of no treatment in the interim period would be recommended.

At this stage there are no 'right' answers. In situations where there is a most appropriate drug, the neurologist or MS nurse will let you know. For this particular decision the evidence suggests the DMDs you have been asked to choose between are as effective as one another. You will have access to information about the difference between DMDs and your decision may depend on your preferences e.g. practicalities of storage or preferred way of taking the drug. You can discuss the options with a healthcare professional and/or friends and family (if you wish to).

You can change your mind. Although research studies tell us about how well drugs work across whole groups of the population, people cannot know how a DMD will affect them as an individual until they have tried it. If you find the side effects difficult or you continue to relapse, you can discuss stopping or trying a different DMD with your MS nurse or neurologist at a later stage.

11.5. Discussion

Interviews with neurologists further reinforced the concept of 'imposed consumerism' that emerged from analysis of MS participants' interviews. Most of the neurologists said they tended to *initially* give decisional responsibility to patients, and then later responded to patients' preferred levels of involvement *if* they appeared to require guidance. This approach largely corroborated the MS participants' descriptions of their neurologist's approach, although none of the MS participants said that their preference for guidance was later accommodated, as most of the neurologists interviewed suggested they intended to. We cannot make direct comments about MS participants'

neurologists' approaches, but if similar to these neurologists, this lack of accommodation may be because patients' unease went undetected.

MS participants were unaware of the reasons why they were given decisional responsibility. All the neurologists agreed that a key factor was the comparability of DMDs in terms of their effectiveness. They said this meant that the decision was based upon more practical issues such as storage or administration methods, and consequently the best option depends upon patients' preferences and lifestyles. In addition to this, all the neurologists discussed how they aimed for best practice by giving decisional power to the patient. They referred to literature in support of patient involvement and used terms such as 'paternalism' to describe an outdated approach and SDM when describing patient involvement. Some also referred to literature in support of patient involvement for people with MS specifically, and based on this, argued that patients who are involved in decisions have better compliance and are less likely to regret their decisions.

Much of the literature focused on decision making in MS, argues for increased patient involvement. For example, one survey recorded that 79% of people with MS surveyed, prefer more active roles (autonomous, informed choice, SDM) than less active roles (professional as agent or paternalism) (Heesen et al., 2007a). But these survey responses are to hypothetical situations, and discrepancies between hypothetical and real situations have been demonstrated in other health decision contexts (Entwistle et al., 2001). This may explain why MS participants' judgements about their preferred role in a decision they had really made contradict what previous literature suggests patients hypothetically prefer.

As explored in chapter 1, there is a body of literature and policy documents advocating patient involvement and recommending SDM as the best interactional style to support

involvement. But much of the discourse around this is over-simplified and does not address many of the complexities and implications of enacting such partnerships in reality (Cribb and Gewirtz, 2012). One area that is often overlooked is how to respond to patients' preferred level of involvement. Literature discussing SDM claims that patients should only be involved to the level they are comfortable with, but this is often presented in response to concerns that SDM processes may impinge on the agency of patients who prefer less involvement. More commonly, SDM literature focuses on presenting increased involvement equating to good practice and little examination of how involvement preferences ought to be accommodated. For example, a narrative review identifies patients' preferences for less involvement as a barrier to SDM (Joseph-Williams et al., 2014), reflecting an SDM discourse that does not accommodate the patients' preferences when they are for reduced involvement. If SDM aims to accommodate patients' preferred level of involvement, then theoretically it can exist in varied forms, some of which appear at odds with the archetypal SDM interactional style. This is illustrated in the table below:-

Table 11.1 Shared decision making as both an archetypal interactional style and a concept that accommodates all styles

Decisional power dynamics	**Consumerism**	**Shared Decision Making**	**Paternalism**
	Patients have high levels of decisional control and healthcare professionals have low. Patients make the decision (Stewart and Roter, 1989).	Patients and healthcare professionals have equal levels of decisional control. They make the decision collaboratively (Coulter and Collins, 2011).	Healthcare professionals have high levels of decisional control and patients have low. Healthcare professional makes the decision (Stewart and Roter, 1989).
Patients have power to influence decisional power dynamic adopted	← **If an SDM approach accommodates patients' preferred style of interaction, it may span the spectrum of styles in terms of decisional power** →		
	The existence of clinical equipoise between options may mean healthcare professionals have little to offer in terms of professional recommendations. Although discussion of preferences through a SDM process may occur, patients have power to choose between options.	In an archetypal SDM consultation, healthcare professionals and patients discuss factors from a clinical and personal perspective; in light of these, agree to a course of action.	In contexts where patients prefer not to be involved, healthcare professionals may make decisions on patients' behalf. This is congruent with paternalistic decisional power dynamic, but the key aspect is that patients have chosen this approach. A set-up where healthcare professionals consider patients preferences alongside clinical knowledge has been termed "physician as perfect agent" (Gafni et al., 1998)

SDM not only describes an archetypal style of interaction, but conceptually it can also accommodate a spectrum of interactional dynamics if responding to patients' involvement preferences. Yet this premise is problematic: the very term *shared decision making* may appear to be at odds with accommodating patients' preferences not to be involved in the decision process. Thus, healthcare professionals may face challenges

when following professional advice to involve patients whilst also responding to a patient's preferred level of involvement (if it is to not be involved). In this thesis, the dynamic – in terms of decisional power – has been interpreted as akin to the consumerist model because patients are provided with information and given power to decide independently. However, some of the neurologists described this decisional set-up as SDM, further demonstrating the nebulousness and confusion surrounding such terms.

Aiming for patient involvement is not a problem in itself, but accounts from the interviews suggest problems arise when patient involvement is encouraged (or imposed) indiscriminately in contexts where more nuanced approaches are required to accommodate patients' preferences. As the table above demonstrates, a consumerist style of interaction could be encompassed under an SDM definition. But a vital step appears to be missing; the data from both sets of interview data suggests patients' involvement preferences are not typically explored or responded to (with some neurologists showing reluctance in doing so, arguing that patients ought to make the decision).

In terms of preventing a feeling of 'imposed consumerism', explicit discussion with patients about their involvement preferences seems like a simple method to ensure patients understand they can be involved at a level they prefer. But this may conflict with some of the neurologists' stances who gave patients decisional responsibility as default, arguing that this was best in terms of compliance and later satisfaction, despite difficulties they may experience making the decision. To what extent should *healthcare professionals' preferences* for patient involvement be enacted if judged to be the most effective way to make the decision? Despite arguments for patient empowerment and mutuality, healthcare professional power persists within the dynamic as neurologists

appear to ultimately decide the level to which patients ought to be involved and those that accommodate patient preference do so on their terms i.e. patients' experience of involvement is likely to be largely determined by the particular neurologist they consult.

Some neurologists discussed the prospect of working through the decision with patients and helping them to consider their preferences. Leading authors in SDM, Elwyn et al. (2000) outlined guidance for healthcare professionals in contexts of clinical equipoise as follows:

"1) implicit or explicit involvement of patients in the decision-making process; 2) explore ideas, fears, and expectations of the problem and possible treatments; 3) portrayal of equipoise and options; 4) identify preferred data format and provide tailor-made information; 5) checking process: understanding of information and reactions (e.g. ideas, fears, and expectations of possible options); 6) acceptance of process and decision making role preference; 7) make, discuss or defer decisions; 8) arrange follow-up."

This approach aims to involve patients (point 1) but also is responsive to the level in which a patient prefers to be involved (point 6). Participants' surprise as to why they were asked to make the decision may be alleviated if they were aware of existence of clinical equipoise (point 3). The burden of the decision could also be lessened through opportunities to defer decision (point 7) or knowing that decision may be changed through follow up (point 8). They also suggest working through patients' perspectives (point 2) and implications (point 5) on a personal level. Taking such a comprehensive approach could alleviate many of the issues raised by MS participants and prevent a sense of 'imposed consumerism'. Many of the neurologists said that ideally they would work through this decision, but time limitations and lack of communication training was consistently reported as a barrier to this. This echoes findings from chapter 10, where neurologists said that ideally they would have comprehensive discussions about complementary and alternative therapies if patients required this in order to appear less

dismissive. However, limited resources in terms of time available and communication training were identified as major barriers to such discussions.

The perceived importance of this decision is likely to differ between patients and neurologists, for whom seeing patients make this decision is routine. If we consider a context where healthcare professionals judge the treatments are of equal effectiveness and the time limits of 'very busy clinics' mean they are unable to deliberate with the patient, an MS patient may feel rushed and/or alarmed by professional uncertainty when making – what is likely to feel like – a major decision. Based on the findings presented here and in chapter 9, some pointers have been developed (box 11.1) in order to lessen the burden felt by MS patients when choosing a DMD. Although how it would be received is unknown, it aims to alleviate distress felt by patients based upon combined findings from interviews with people with MS and neurologists. However, it is unclear whether all healthcare professionals would be willing to potentially make this decision on patients' behalf if they opted for this (as the pointers suggest).

Section five
Discussion

Chapter 12

Discussion

12.1. Chapter summary

The aims of this doctoral study were to explore the information and decision support needs of people with MS, and consider personalised care planning as a suitable approach for them. Using a range of methods (outlined in chapter 3), this research adopted a broad gaze to identify and examine decisional support and information needs relevant to people with MS. The chapter begins with a discussion of the principal findings, then each research question is addressed in turn. This is followed by reflections on the strengths and limitations of this research, and finally the chapter considers the implications for practice and directions for future research.

12.2. Discussion of principal findings

12.2.1. Literature gap examining personalised care planning for people with MS

This doctoral research identified a clear gap in the literature examining the effectiveness of personalised care planning for people with MS. The systematic review which aimed to assess the effectiveness of personalised care planning interventions for people with MS specifically (presented in chapter 6) identified only two eligible studies, despite including a broad range of study types. Furthermore, methodological issues and unclear presentation of data meant that no conclusions about the interventions' effectiveness could be made. This evidence gap also appeared to exist more broadly, as none of the studies included in the Cochrane review (summarised in chapter 4) tested interventions

aimed at people with long term *neurological* conditions (Coulter et al., 2015). This gap is noteworthy when we consider the promotion of personalised care planning for people with long term conditions (LTCs), and specifically that the National Service Framework for people with LTCs identified personalised care planning as a priority specifically for people with long term *neurological* conditions (Department of Health, 2005).

In terms of the effectiveness for LTCs more broadly, findings from the Cochrane review suggest interventions improve physical and psychological health and people's abilities to self-manage, compared with usual care (Coulter et al., 2015). However, these favourable effects were only moderate. Also, there was very little evidence demonstrating favourable effects for subjective health status or reduction in use of healthcare services. In theory, forward-planning, preventative approaches with activated patients is advocated as an ethical and economical solution to healthcare systems under strain (Wanless, 2002, Wagner, 1998), but in reality, it is unclear whether this approach is effective or feasible.

The lack of evidence assessing effectiveness of personalised care planning for people with MS (and long term neurological conditions) is striking in relation to diabetes, which has much more evidence demonstrating its effects. For example, 12 out of the 19 studies included in the Cochrane review assessed interventions for people with diabetes. There are potential reasons why personalised care planning interventions for diabetes appear to be more popular, compared with those for MS. It may be part of wider movements to promote self-management for and by people with diabetes, reflected in the development of diabetes self-management groups such as DAFNE (dose adjustment for normal eating) and DESMOND (diabetes education and self-management for ongoing and newly diagnosed) that provide self-management training and advocate patient education (DESMOND, 2015, DAFNE, 2016). There may be more actions

people with diabetes can plan, such as daily insulin dosage and monitoring diet, compared with MS where triggers are often ambiguous. Or it could be that health status indicators are easier to measure in diabetes (blood sugar levels) compared to neurological conditions, where symptoms are varied and often difficult to measure or sometimes detect. This makes design of effectiveness trials problematic and defining suitable outcomes in MS difficult. As Goldman put it:

"Measurement of MS disease progression and treatment response is further complicated by: individual patient heterogeneity; population variability in disease course and tempo of progression; subclinical MRI changes of uncertain impact on delayed disability progression; multifaceted neurological deficits with varied abilities for individual patients to compensate; and patient comorbidities. Independently and collectively, these issues have proved challenging for the design of MS therapeutic trials. In addition, cost limitations and practicalities have resulted in studying a chronic disease in snapshot increments, with clinical trials on average occurring over a 2-3 year period in a disease that lasts decades […] A single MS outcome measure of sustained disease progression may remain elusive." (Goldman et al., 2010, p.230-235)

The unpredictability and uncertainties within MS are common features that run throughout this thesis, and it stands to reason that this poses challenges in research settings. If we couple the complexities of MS with the likelihood that personalised care planning is a complex approach, it is unsurprising that there appears to be little research published in this area.

12.2.2. Implications of the uncertainties inherent in MS

Three key areas of uncertainty (existential, embodied and interventional) featured in MS participants' accounts and these appeared to factor into their experiences and the ways they used and accessed information. Existential uncertainty refers to how future effects of MS are ambiguous due to the unpredictability and variability of potential prognosis. The analysis showed how, in a context of existential uncertainty, some people with MS

initially imagine their prognosis as one where they will be severely affected by MS. In turn, this can lead to a perception of their future as being substantially altered from what they had previously imagined.

Bury's seminal work on 'biographical disruption' conceptualised the presence of chronic illness as a "disruptive experience" in people's lives, which is unexpected and changes their previously imagined life course (Bury, 1982). Various theorists have examined and developed this concept (Charmaz, 1983, Williams, 2000, Locock et al., 2009). Locock et al. (2009) coined the term "biographical abruption" to describe experiences of people with motor neurone disease (MND). This concept is distinguished from Bury's "biographical disruption", because rather than a "disturbance and unwelcome change, 'abruption' is intended to convey a sudden ending, literally a 'breaking off'" (Locock et al., 2009, p.1047). Analysis of the interview data showed how people with MS may initially experience something closer to "biographical abruption" than "disruption" when thinking about their futures.

Participants talked about how this initial period where they imagined a severe prognosis led to feelings of powerlessness, fear and anxiety. Not only are these undesirable in themselves, but low mood is also linked to reduced functional ability in people with MS (Smith and Young, 2000), and some participants linked a worsening of symptoms to the shock they felt around this time. The findings also showed how, for some, this perception shaped life decisions, such as those about child bearing, marriage and career plans.

Perceptions of severe prognosis seemed to be further reinforced by information accessed around the time of diagnosis. A recent literature review examining healthcare experiences of MS patients in the UK, identified a lack of timely and credible

information at the time of diagnosis, which was linked to feelings of anxiety and fear (Methley et al., 2015). This corroborates findings presented in chapter 8, which suggest information provided at the point of diagnosis is often insufficient and sometimes non-existent. Furthermore, the findings suggest the information gap at diagnosis is often filled with negative representations of MS, some of which were accessed during independent information searches, and some were encountered by chance. This appeared to instigate and/or reinforce negative perceptions, and in turn exacerbate unnecessary and unfavourable consequences of these.

However, many participants described how such initial perceptions were temporary, exaggerated and later adjusted, as they later reimagined their prognosis would not be as severe as previously thought. In turn, a perception of their future closer to what they envisaged *before* diagnosis seemed to be restored. Such restoration (or repair) of biographies appeared to be aided by their own experiences of a milder version of MS, and by accessing information containing a broad range of MS trajectories. Elsewhere, theorists have observed how people with LTCs may 'reconstruct' or 'repair' disturbed biographies by adapting and incorporating illness effects into new perceptions their futures (Charmaz, 1985, Williams, 1984). Some MS participants did recount elements of such repair; but these findings also demonstrated how some participants later restored their previous imagined futures, without incorporating (much, if any) existential change, implying a notable disparity between initial perceptions of MS and reality.

As outlined in chapter 2, MS is a condition with a broad range of potential symptoms (Dua et al., 2008) and a lack of instrumental objective measures that reliably record disease activity (Lublin et al., 2014). This context appears to have led to people with MS experiencing confusion over whether symptoms are caused by MS or something different. The term 'embodied uncertainty' was used to define this confusion in chapter

7. This embodied uncertainty has implications in clinical settings, as causes of problems may be misattributed either to MS, or something other than MS. The findings presented in chapter 9 demonstrated how this can result in misdiagnosis and subsequent barriers to appropriate treatments, or cause frustration and concern if patients are concerned that true causes are overlooked.

Difficulties in establishing symptom causes mean that MS is a condition that typically takes a long time to diagnose (Dua et al., 2008, Methley et al., 2015). During this liminal stage, many participants said that they were given little information and felt frustrated and distressed. Some suspected they had MS and did not understand why they were not diagnosed sooner, and others were surprised to receive a diagnosis, because they thought MS had previously been ruled out.

The unpredictable relapsing nature of prognosis and uncertainties around how MS manifests meant that participants often found it difficult to judge the effectiveness of interventions they used. They talked about the impossibility of knowing whether apparent recovery was due to an intervention's true effects or a period of remission. Also, some participants were reluctant to judge an intervention as ineffective if they experienced no improvement, due to the potential that their symptoms may have worsened further without it. A context where there are limited clinically proven treatments available, and uncertainty regarding intervention effects, seemed to foster interest in, and the adoption of, a broad range of interventions outside mainstream medicine.

12.2.3. Perceived importance of bespoke experiential information and peer support

The findings presented in chapter 8, suggest that peer support and experiential information were perceived as being particularly valuable. Some participants

distinguished these from more general information accessed via official sources (such as healthcare professionals and charities), that they often found did not apply to them personally. Instead, they sought out others' experiences that were particularly relevant to their personal circumstances and decisions they faced. Greenhalgh et al. (2015) recognised this distinction between mainstream medical and experiential bodies of information:

> "EBM's accumulating body of (explicit, research-based) knowledge and the (informal, tacit, and socially shared) knowledge actually being used by people managing their condition are developing separately rather than in dialogue with one another." (Greenhalgh et al., 2015, p.7)

Similar to the accounts of MS participants, Greenhalgh et al. position experiential information sources as being perceived as particularly useful, compared with more official sources, and suggest that EBM discourses tend to devalue or overlook more 'informal, tacit, and socially shared' sources of information. In a condition like MS that is highly variable, the development of information that is both general and reliable is challenging, and with such heterogeneity, it is unsurprising that participants instead used others' experiences to identify personally bespoke information. The findings also showed how some participants tailored their peer support. They described how initial peer contact was difficult because they struggled to relate to others in such groups, or seeing others affected by MS caused distress regarding potential futures. In these cases, such 'identity tension' (Mazanderani et al., 2012) appeared to be overcome by organising or accessing peer support more relevant to themselves.

The findings presented in chapter 8 demonstrate how own experiences were considered a vital source of information for people with MS. The heterogeneity of MS in terms of symptoms, relapse triggers and treatment effects, and lack of objective measures meant

the only way to understand these elements was by experiencing them. However, this also had limitations, as embodied uncertainties meant making such judgements was sometimes considered to be impossible.

12.2.4. Feeling "dismissed"

Findings presented in chapter 9 demonstrated how some MS participants appeared to opt out of future consultations (either about specific topics, with particular healthcare professionals or with healthcare services more generally) when they perceived healthcare professionals as being dismissive about information they presented. In contexts where MS patients want to discuss complementary and alternative medicine (CAM) approaches, this poses a particular dilemma. If healthcare professionals treat the prospect of trying such approaches as worth serious consideration, this may risk endorsing them to some extent. But, if they assert their professional judgement – that they are not worth pursuing – this may close conversation and patients may opt out of future consultations (without useful discussion).

Consultant neurologists were presented with this dilemma during structured interviews. The findings presented in chapter 10, showed there was consensus that discussion of such topics could be interpreted as endorsement to some extent. However, there were mixed responses concerning the best way to respond to patients' wishes to discuss CAM, ranging from some who thought it was part of their role to engage with these topics, to others who thought this was outside their remit.

Neurologists considered ways to overcome this dilemma. They talked about how ideally healthcare professionals could explore the reasoning and motives behind patients' interest in CAM, during which they could help patients to translate the evidence and critically appraise the information they have accessed. Additionally, they could identify

other relevant support during such discussions e.g. CBT or mindfulness. But such solutions were presented by neurologists as ideals. Various barriers such as lack of communication training and time limitations during consultations (for discussion) and outside consultations (for research), meant that providing such support (if done responsibly) was perceived as unrealistic. Interviewed neurologists who thought discussions about CAM were not part of their remit, suggested that such topics would be better accommodated by other healthcare professionals, such as MS nurses or GPs. However, given that not everyone has access to MS nurses (Morse, 2011), and GP services are under strain (Hobbs et al., 2016), it is likely the same barriers may persist.

In turn, this raises questions about what is deemed appropriate content for healthcare consultations. Arguably, from a patient-centred care perspective, content of consultations should be broad and potentially include anything related to patients' wellbeing (Entwistle and Watt, 2013). CAM relates to health and wellbeing in that it may be judged on a personal level as beneficial in varied ways (Shinto et al., 2005, Esmonde and Long, 2008), or conversely it might be costly, time consuming, invasive, used instead of proven strategies, or risk side effects and interactions with other medications. If healthcare professionals are not engaging in such discussions, then patients are left to access (and could be convinced by) less regulated sources (Mazanderani et al., 2013). Furthermore, if content of consultations were limited to discussions about officially recommended interventions only, it is unclear how patients would understand the difference. We can assume that healthcare professionals have a clear idea about the differences between CAM and mainstream medicine, but this distinction is likely to be less clear to patients seeking advice.

12.2.5. Imposed consumerism

The findings suggest that when deciding which type of disease modifying drug (DMD) to start, people with MS are typically provided with lots of information, but little guidance. This scenario was conceptualised as 'imposed consumerism' in chapter 9, because although patients adopted a consumerist role, where they had high levels of control and healthcare professionals had low in regard to the actual decision, this decisional power was *imposed* upon them, and typically not welcomed. Paradoxically, this is arguably the reverse consumerist power dynamic, as patients have low levels of power and healthcare professionals have high power in deciding who had decisional responsibility.

The findings presented in chapter 11 showed that the neurologists interviewed recognised that MS patients are typically given a choice of DMDs to choose between; however, some also thought that it was unlikely that patients would be dissatisfied with making this decision independently. The analysis revealed two main motives behind giving the patient control of this particular decision. Firstly, DMDs are so comparable in terms of effectiveness and side effects, neurologists felt they had little advice to offer, and that the decision ought to be based upon personal preferences (in regard to administration). This may reflect a perception of their roles as information purveyors, rather than extending to decision facilitators. Secondly, neurologists showed appreciation for the literature and policy that promotes patient involvement, and often considered clinical equipoise an opportunity to enact it.

It seems reasonable that neurologists would encourage patients to make decisions where possible, as patient involvement is largely advocated by official bodies, academia and policy makers (as outlined in chapter 1). Movements away from paternalism were born

out of an imperative to respect and attend to patients' rights to be involved (Foot et al., 2014, Morgan, 2008, Edwards and Elwyn, 2009), and some of the neurologists interviewed justified the approach as a way to avoid 'outdated' paternalism. Some described patients making this decision autonomously as 'shared decision making', illustrating how interpretations of 'informed choice' and SDM can become confused when used interchangeably (Moumjid et al., 2007). The trend advocating increased patient involvement is also apparent when focusing on MS literature specifically. The Association of British Neurologists' (ABN) DMD guidelines recommend that neurologists "recognise the central importance of patient choice" (ABN, 2009, p.4), and much of the literature about decision making in MS argues for increased patient involvement. Heesen et al. (2011, p.192) argue that "[f]or patients with multiple sclerosis (MS) involvement in treatment decisions becomes ever more imperative", and studies recording MS patients' preferred roles in decisions about DMDs have shown that the majority of participants included in these studies prefer autonomous roles, compared with under half who prefer to share decisions, and a small minority who prefer healthcare professionals to make decisions on their behalf (Kasper et al., 2008, Heesen et al., 2007b). Previous research has also shown that when MS patients are involved in decisions, they are more likely to continue with the chosen DMD in the long term, and be satisfied with their decision (Koudriavtseva et al., 2012). Thus, it is unsurprising that, although neurologists recognised that not all patients wanted to make this decision, this was perceived to be rare and motives for 'best practice' by enacting patient involvement appeared to outweigh such concerns.

In contexts of clinical equipoise, some have argued that patient involvement becomes particularly relevant because there is no obvious clinically 'right' option (Coulter et al., 2011). But these findings and research elsewhere (Ziebland et al., 2006) have shown

how such uncertainty can be burdensome to patients, arguably making patient involvement less suitable. Some have recommended guidance for healthcare professionals to encourage patients to become involved, but also accommodating their preferences for less involvement in contexts of equipoise (Elwyn et al., 2000). However, such guidance is detailed and is likely to require time and skilled discussions. All the neurologists said that, ideally they would work through these decisions with patients, but in reality they felt they neither had time available during consultations, nor the communication skills to do this sufficiently. Others have identified a gap in communication training, inhibiting adoption of SDM (Petrova et al., 2006, Stiggelbout et al., 2012). Some neurologists interviewed suggested that MS patients felt distressed during this decision, due to its perceived importance and potential timing of the decision. If MS patients were aware the DMDs were comparable in terms of effectiveness, that they could change their mind, and the decision was timed appropriately, this could reduce the perceived burden of this decision, and ameliorate some of the frustration and distress felt.

Another issue highlighted in the findings was ambiguity in how to accommodate involvement preferences. Firstly, neurologists appeared to lack confidence in how to gauge involvement preferences. Secondly, although they agreed they should respond to involvement preferences, this was to varied extents. Some were more willing to be guided by patients' involvement preferences and make decisions on patients' behalf (when favoured by patients), whereas others said that the imperative to adhere to best practice meant patients ought to make this decision, despite it being difficult.

12.3. Question 1: What does the literature tells us about the effectiveness of a personalised care planning approach for people with MS?

Based upon the findings presented in both systematic reviews (presented in chapters 4 and 6), it remains unclear whether personalised care planning is an effective approach for people with MS. There have been various studies suggesting that people with MS do prefer to be involved in decision making (Koudriavtseva et al., 2012, Hamann et al., 2007, Heesen et al., 2004). However, these focus on MS patient involvement in discrete decisions, rather than broader preventative forward-planning approaches. The lack of evidence assessing personalised care planning approaches does not necessarily mean such approaches are ineffective for people with MS, but rather it may reflect that designing research assessing the true effects is challenging.

12.4. Question 2: In which contexts are decision support and information needs particularly relevant for people with MS?

12.4.1. Information around the time of diagnosis

The period during diagnosis has been identified as a particularly distressing time for people with MS (Solari et al., 2014, Methley et al., 2015), and this was reinforced by the findings presented in chapter 8. This study identified two stages where participants felt particularly distressed; firstly, a liminal stage where they were having unexplained symptoms and diagnosis was not yet confirmed, and secondly, the period shortly after an MS diagnosis was confirmed.

There are various criteria required to confirm an MS diagnosis (Polman et al., 2011), and difficulties in assessing symptom causes means diagnosing MS typically takes

prolonged periods of time (Dua et al., 2008, Methley et al., 2015). Some participants talked about how their neurologist seemed to rule out the possibility of MS, and reluctance to acknowledge potential for an MS diagnosis has been evidenced elsewhere (Heesen et al., 2003). However, these findings showed how lack of acknowledgement can lead to either frustration that concerns are not taken seriously, or shock when eventually diagnosed. Appreciation of the complexity of diagnostic processes was not evident in participants' accounts; instead frustrations felt during this period were often aimed at neurologists, who were perceived as being evasive and not taking concerns seriously. This suggests a potential mismatch between patients' *expectations* and the *reality* of diagnostic processes. People with MS may benefit from clear information about the diagnosis processes and acknowledgement that they may have MS. Heesen et al.'s (2009) patient information aid specifically for diagnostic stages (discussed in chapter 8) could address these issues, as it provides a comprehensive overview of diagnostic stages and implications of receiving diagnosis along with potential disease trajectories. The information aid was positively received by people with suspected MS and those recently diagnosed considering it retrospectively. It did not appear to cause additional anxiety (contrary to authors' expectations) and respondents identified the period *before* diagnosis as the best time to receive it.

At the point of diagnosis, a lack of information (from official sources), and negative representations of MS (accessed independently), led some to imagine they would be severely affected by MS. However, with experience and access to more information, participants often judged these perceptions to be exaggerated, and thus subsequent distress and affect in life choices unnecessary. In order to aid adjustment to illness, having a positive and accurate view of self and a perception of illness that matches the reality of illness have been identified as important aspects (Sharpe and Curran, 2006).

In light of these findings, people recently diagnosed with MS could benefit from information about the broad range of potential MS trajectories, including positive representations. Such information may avoid particularly negative perceptions of MS prognoses and aid adjustment.

There are existing information resources that present a range of potential MS trajectories and people living positively with MS. These include: information aids developed in Germany (Heesen et al., 2009) and Italy (Borreani et al., 2014); a UK based social networking website that hosts information and short films depicting the varied experiences of MS (SHIFT MS, 2016); and a leaflet developed by charity MS-UK with examples of people living positively with MS (MS-UK, 2016). But, despite the existence of such resources, the findings suggest this type of information was not readily accessed by those interviewed. People recently diagnosed with MS may benefit from being provided with – or signposted to – this type of information. Some healthcare professionals have voiced concerns that providing information about milder trajectories to people with more severe forms of MS would be inappropriate (Borreani et al., 2014). With this in mind, care ought to be taken not to present an overly positive view of MS, but rather a balanced and realistic representation of potential trajectories, with examples of the potential to live well with MS.

12.4.2. Complementary and alternative medicine

MS is potentially debilitating; regulated treatment options available are limited; and intervention effects are difficult to judge. In this context, it is unsurprising that people with MS commonly use CAM (Apel et al., 2005, Nayak et al., 2003), and that many participants seemed open to trying such approaches. The findings presented in chapter 10, revealed how neurologists had strong concerns about the appeal of *some* alternative

approaches that promised dramatic outcomes, were expensive and judged to be dangerous.

Analysis showed how people with MS often find out about CAM from others' experiences, and, although most participants had a clear understanding of the limitations of experiential information, some considered others' accounts as enough to convince them that an approach was effective. Interviewed neurologists suggested that people with MS might benefit from guidance to critically appraise the reliability of information they access. Recognising the challenge of assessing reliability of information, a joint Austrian and Italian study developed a website, specifically designed to provide evidence-based information for people with MS (Colombo et al., 2016); however, content was limited to licensed treatments. The charity 'Sense About Science' (2013) developed a tool that aims to help members of the public to assess the reliability of information and implications of trying untested treatments; such a resource could be potentially useful in making decisions about alternative treatments.

12.4.3. Support in decisions about disease modifying drugs

The findings suggest that people with MS starting DMDs are often expected to choose a DMD autonomously, as they are given lots of information, but no advice from healthcare professionals. This decisional responsibly seemed to cause distress and confusion as participants said they felt burdened and would have like more guidance. Although interviews with neurologists revealed reasons behind why MS patients are expected to make this decision, analysis of MS participants' interviews suggest that people with MS are often unware of these. If they understood decisional contexts and implications, and their involvement preferences were accommodated, this could reduce

the perceived burden of this decision. In light of this, box 11.1 was developed in chapter 11 as an information aid for people with MS facing this decision.

Box 11.1: Information for people with MS choosing Disease Modifying Drugs when clinical effectiveness is comparable

It's up to you how and whether you make this decision. Some people would like to make this decision independently; some would like to share this decision with healthcare professionals and/or friends and family; and others would prefer the neurologist or MS nurse to make the decision on their behalf. All of these options are fine and the MS nurse or neurologist can accommodate your preferred level of involvement. You may choose to delay making this decision until a later date. In this case, a discussion with your neurologist or MS nurse about potential outcomes of no treatment in the interim period would be recommended.

At this stage there are no 'right' answers. In situations where there is a most appropriate drug, the neurologist or MS nurse will let you know. For this particular decision the evidence suggests the DMDs you have been asked to choose between are as effective as one another. You will have access to information about the difference between DMDs and your decision may depend on your preferences e.g. practicalities of storage or preferred way of taking the drug. You can discuss the options with a healthcare professional and/or friends and family (if you wish to).

You can change your mind. Although research studies tell us about how well drugs work across whole groups of the population, people cannot know how a DMD will affect them as an individual until they have tried it. If you find the side effects difficult or you continue to relapse, you can discuss stopping or trying a different DMD with your MS nurse or neurologist at a later stage.

The box contains key information points about aspects of the decision that – based on the thesis findings – may clarify decisional context and alleviate some of the burden felt when choosing a DMD.

12.5. Question 3: How can healthcare professionals most effectively support people with MS?

The previous section discussed areas that were identified as particularly relevant in terms of information and decision support needs for people with MS. Developing this further, this section draws on findings from chapters 9, 10 and 11 to consider how healthcare professionals can effectively support people with MS.

12.5.1. Engagement in uncertainty

Popular discourse of biomedicine as deterministic and authoritative (Adamson, 1997) means patients often expect certitude from healthcare professionals (Hatch, 2017). However, in reality, uncertainty is present in much of medical knowledge, and it has been argued that healthcare professionals ought to be more open about this in order to foster trust from patients (Hatch, 2017). The findings suggest that healthcare professionals may support people with MS by engaging with uncertainties inherent in the condition. For example, participants felt frustrated with neurologists during diagnostic stages and perceived them as being evasive, rather than appreciating the complexity of MS diagnosis. It is possible this may be alleviated if healthcare professionals acknowledge the potential for MS, and explain the complexity of diagnostic processes. The prospect of informing patients that MS is suspected raises concerns that it may cause patients anxiety, and some neurologists have reported withholding such information during investigations in order to protect them from difficult information (Heesen et al., 2003). However, concerns about causing unnecessary anxiety have been mainly voiced by healthcare professionals rather than patients, and there is some evidence suggesting early provision of such information is preferred by people with MS (and suspected MS), and it does not cause additional

anxiety (Heesen et al., 2009). Additionally, withholding information about MS could be seen as prioritising or protecting those who do not develop clinically definite MS, which is estimated to be 18% of those with clinically isolated syndrome (first sign suggestive of MS) (Novakova et al., 2014).

Discussing cause uncertainty may be particularly useful for people with MS. The findings presented in chapter 9 showed how participants recounted times when healthcare professionals seemed to misattribute (or potentially misattribute) symptoms they experienced either to MS or something else. This could lead to feelings of distress, or in some cases, seemed to inhibit access to appropriate treatments. Ensuring that uncertainty is discussed could provide a context for people with MS to voice concerns and provide opportunities for reassurance through subsequent explanations or investigations.

12.5.2. Discussions about CAM

It is unclear whether healthcare professionals can accommodate useful discussions about CAM when patients request it. Various other studies (mostly in cancer and paediatric care) have identified barriers to discussions about CAM within consultations (Davis et al., 2012), and similarly found that previous negative reactions from healthcare professionals appeared to make patients less likely to disclose their CAM use or bring up these topics up in future consultations (Tasaki et al., 2002, Schofield et al., 2010). In order to overcome these issues, many have recommended ways in which healthcare professionals can engage in more open, encouraging and non-judgemental ways about CAM (O'Keefe and Coat, 2010, Tasaki et al., 2002, Schofield et al., 2010). Based upon fulfilling the needs of patients already interested in CAM, such comprehensive approaches seem sufficient; however, when we consider this in broader

contexts, problems arise. Firstly, during structured interviews with neurologists they identified time limitations as a barrier to discussions about alternative approaches. They agreed that the lack of time to keep updated with current alternative approaches and time pressures within consultations led to a lack of knowledge base and prioritisation of other content respectively. Therefore, in real contexts it may be unlikely they could adhere to such comprehensive guidance. Secondly, none of the studies advising best ways to engage in CAM discussions address the risk that engagement in such topics may be interpreted as endorsement to some extent, which was a concern for the neurologists interviewed. For example, one study suggested that CAM ought to be part of medical school training and that leaflets about CAM could be left in the waiting rooms to encourage discussions (Tasaki et al., 2002). Such accommodation might be useful for patients already interested in alternative approaches. But this may be interpreted as heightening legitimacy, not only for those already interested, but also for patients more broadly. Therefore, it is likely to be met with unease from evidence-based medicine, epistemological, and public-spending perspectives.

The best way to engage in discussion about CAM with patients is unclear, but these findings demonstrate how merely stating judgement that it is not worth considering, can be perceived as dismissive and have potentially damaging consequences. In chapter 9, it was argued that in aiming for patient-centred care, the content of consultations ought to be shaped by patients' priorities, which can include discussion of approaches outside mainstream medicine. But feedback from neurologists suggests that this may not be realistic in current healthcare systems. It seems a balance must be struck between taking seriously the patients' view that an approach is worth considering, whilst also communicating a professional stance (that it is not worth considering).

12.5.3. DMD decisions

The neurologists interviewed judged that most people with MS would be satisfied when choosing a DMD autonomously, and this perspective is supported by literature about DMD decisions (Kasper et al., 2004, Heesen et al., 2011). But findings presented in chapter 9 demonstrated how people with MS may prefer more guidance from healthcare professionals. In the light of these findings, healthcare professionals could support people with MS by firstly recognising that deciding which type of DMD to start can be distressing and challenging for their patients. Providing information about aspects and implications of the decision (detailed in box 11.1) may help to reduce decisional burden.

Healthcare professionals could additionally expand upon the point entitled: 'You can change your mind' (included in box 11.1) (i.e. the decision is not permanent) by ensuring that experiences of the chosen DMD are monitored and reviewed. Not only does the monitoring of treatment effects on an individual level adhere to evidence-based principles (Greenhalgh et al., 2015, Gray, 2004), clarifying this to patients may also provide reassurance. The first point in box 11.1: 'It's up to you how and whether you make this decision' may be contentious as some neurologists interviewed showed a reluctance to be involved in decisions. The pointer is justified as accommodating a patients' preferred level of involvement can adhere to SDM approach (Towle and Godolphin, 1999, Elwyn et al., 2000) and the ABN DMD guidelines (ABN, 2009) recommend the 'central importance of patient choice' is recognised when making decisions *with* neurologists. However, if accommodating a patient's preferred role conflicts with some neurologists' priorities, then implementing is likely to be inhibited (May and Finch, 2009).

12.6. Question 4: What elements are important to consider in personalised care planning for people with MS?

12.6.1. Organisational support

The Cochrane review found that interventions which were embedded within and supported by organisations were most successful (Coulter et al., 2015). This finding supports arguments made by others who emphasise the importance of organisational support in successful of implementation of planned, proactive and collaborative approaches in healthcare (Batalden et al., 2015, Wagner, 2000, Coulter et al., 2013).

There are various examples of how personalised care planning can fail when organisational support is lacking. For example, a comprehensive primary care based intervention incorporated all the various stages of Coulter et al.'s personalised care planning model (Bower et al., 2012), but the intervention was unsuccessful, and the authors demonstrated how its implementation was impeded by organisational features, such as healthcare professionals having priorities that conflicted with the execution of the intervention (Kennedy et al., 2014). This example demonstrates the relevance of 'collective action' i.e. if an intervention's impact on facilitators' other responsibilities is deemed unacceptable, then it is unlikely to be implemented successfully (May et al., 2009). In the systematic review assessing the effectiveness of personalised care planning interventions for people with MS specifically (chapter 6), one of the included studies trialled an intervention which relied on community resources such as recreational activity centres and public meeting rooms (Block et al., 2010). In an accompanying ethnographic study (Block et al., 2006) they reported on the difficulties in carrying out the intervention, as locations were inaccessible and participants were sometimes unable to take part. This again highlights the importance of anticipating the

organisational support available when designing and implementing personalised care planning.

Additionally, there were two connected concerning elements of Block et al.'s (2010) study: firstly, those who were assessed as *unable to participate* were allocated to the control group, and, secondly, the authors claimed the intervention was successful due to the difference in self-efficacy scores being in favour of the intervention group, but this was in the context of an overall decrease in self-efficacy scores from baseline. This suggests that taking part in this study may have lowered self-efficacy scores, especially for those that the intervention was inaccessible for (those allocated to the control). Although claims cannot be made about direct causality, the potential for personalised care planning interventions (that by design aim to engage patients in their own healthcare), to damage people's feelings of self-efficacy if they are unable to participate, should not be ignored. When implementing personalised care planning – in addition to ensuring that organisational features are supportive – it is also vital that the intervention is designed so all those it is aimed at can participate fully. Otherwise, not only is there a danger that the intervention will be ineffective, but it may risk damaging effects also.

12.6.2. Setting, monitoring and reviewing goals

A comprehensive approach to personalised care planning not only involves collaborative planning between patients and healthcare professionals, but also ongoing monitoring and reviewing. When considering these processes in the context of MS, its uncertainties and unpredictability may pose potential opportunities and difficulties. Arguably, the heterogeneity of MS means that individual monitoring is crucial in interpreting the condition, identifying triggers and assessing intervention effectiveness. But equally, difficulties in assessing presence of disease activity (and subsequent

confusion over causes and intervention effects) and its unpredictable course may present challenges in monitoring for some people with MS.

The findings in chapter 7 also showed that some people with MS experience short term fluctuations in their condition that are unpredictable and affect their capabilities on a daily basis. Many participants talked about difficulties in making plans and (although they were referring to social events) short-term fluctuations and difficulties in planning are worth considering in the context of personalised care planning. Ensuring goals set and actions planned are adaptable, with contingencies to accommodate daily fluctuations during monitoring stages may be particularly useful, especially for those who find their MS typically has fluctuating patterns.

12.6.3. The appropriate healthcare professional to facilitate personalised care planning

A crucial element to consider is which healthcare professional could facilitate personalised care planning for people with MS. Literature and policy documents use generic terms such as clinician or healthcare professional, rather than specifying who ought to be responsible for facilitating such an approach (Coulter et al., 2013, Department of Health, 2011, Coalition for Collaborative Care, 2015, Department of Health, 2005). So despite the mandate that personalised care planning should be offered to everyone with a LTC, lack of clarity about who facilitates it may potentially hinder its adoption.

People with MS may consult a broad range of healthcare professionals, and it is unclear which is best placed to facilitate personalised care planning. Neurologists' expertise is focused in neurology, potential prognoses and treatments (NHS Health Careers, 2016b), rather than broader aspects of health and wellbeing. Analysis of neurologists' interviews suggests that time limitations, lack of communication training and knowledge base

hinder comprehensive discussions with MS patients. Thus specialism, time limitations and relatively infrequent neurology appointments mean that neurologists are unlikely to be suitable to facilitate personalised care planning in current healthcare systems. The role of MS nurses may be more suitable in facilitating this approach. Specialist knowledge in MS means they can provide condition-specific advice and they can also coordinate services (MS Trust, 2016). However, availability of MS nurses varies widely throughout the UK and not everyone has access to them (Morse, 2011).

GPs are a first point of contact and everybody has access to them. A generalist approach means they can consider a broad range of strategies and coordinate services (NHS Health Careers, 2016a). The findings presented in chapter 7 and recent research (Methley et al., 2016), demonstrate that causes of problems are often unclear in the context of MS. Previous studies have also shown that people with MS often have hidden comorbidities (Marrie et al., 2009b, Marrie et al., 2009a). Thus, a generalist gaze might accommodate the breadth of potential problems. However, current GP services have been shown to be under strain and unsustainable (Hobbs et al., 2016), and some GPs have voiced concerns that they lack experience and knowledge because they serve few patients with MS and the condition is particularly complex (Methley et al., 2016). Additionally, other existing priorities are likely to act as competitors for focus. For example, the Quality Outcomes Framework has been shown to reinforce paternalistic dynamics, limit content of consultations to be more bio-medically focused and prioritise target conditions (not including MS) (Chew-Graham et al., 2013). Such existing priorities could act as a barrier to an approach like personalised care planning for people with MS in general practice.

Although none of the participants talked about visiting occupational therapists, much of their role appears to complement a personalised care planning approach, as they assess

patients' priorities and help them to overcome barriers to living independent fulfilling lives (College of Occupational Therapists, 2011). Goal setting appears to be a strategy used frequently, and it features in much of the rehabilitation literature (Kielhofner and Barrett, 1998, Garvey et al., 2015, Mroz et al., 2015). Congruence between personalised care planning and occupational therapy was evident in the MS systematic review (presented in chapter 6), as one of the studies compared a personalised care planning intervention with usual occupational therapy care, but it transpired that factors distinguishing personalised care planning from usual occupational therapy were unclear. Thus, occupational therapists seem well placed to facilitate personalised care planning for people with MS. There is some evidence that occupational therapy as a part of primary care is feasible with promising outcomes (for people with comorbidities) (Garvey et al., 2015), but in current healthcare systems MS patients are likely to be referred to occupational therapists to overcome problems. This means relatively few people with MS consult occupational therapists, and when they do it is often in reaction to problems, rather than prevention of them, which is a defining feature of the personalised care planning approach.

12.7. Concluding remarks

Although there appears to be some evidence demonstrating that personalised care planning is effective for people with long term conditions, such favourable effects were not demonstrated in the context of MS. Based on the findings from the systematic reviews, it is unclear whether personalised care planning is effective for people with MS, and there is a clear gap in the literature examining this. Findings from the interview stages suggest there are key areas which are lacking in terms of information and decisional support. Such areas included the type and amount of information around the time of diagnosis, support when choosing disease modifying drugs and discussions

about approaches outside mainstream medicine. Findings from neurologist interview data corroborated those from MS interview data, but through examination of issues raised, it also highlighted some of the complexities and challenges of involving patients, and enacting shared decision making, in reality.

12.8. Strengths and limitations

Prior to the qualitative research studies, two systematic reviews were conducted, both of which adhered to the Cochrane Collaboration methods and guidance which are widely regarded as the gold standard for systematic review methods. The first systematic review was peer reviewed and published as a Cochrane review (Coulter et al., 2015), demonstrating its importance in the field. The second systematic review drew on and adapted methods used for the first review. Although little evidence was found demonstrating the effectiveness of personalised care planning for people with MS, the review did identify a clear gap in the literature.

In using a modified grounded theory approach, this research was able to examine elements that people with MS identified as important, without being too limited to predetermined topics. Interview data can only represent perspectives that participants recall or choose to tell. This poses particular problems when asking people to reflect on previous interactions; therefore data were interpreted with caution. However, it is worth noting that, despite the lack of direct evidence, the findings from interviews with people with MS were corroborated by findings from the second stages of interviews, and patient and public involvement members considered them relevant. Interview methods also have additional strengths. In direct observations, feelings and intentions are often hidden, but during interviews participants can reflect upon previous experiences. This allows examination of participants' interpretations and the consequences of these (which expand further than the consultation). For example, if we observed patients

being asked to decide which DMD to start, this may not appear to be contentious, but interview data revealed it can be problematic.

Although the types and severity of symptoms varied greatly between those who took part in the interviews, it is possible that those with particularly severe trajectories were less likely to volunteer, thus certain experiences may be absent in the data. For example, although carer support could be inferred from interview content, it was not discussed in depth. This may reflect how family members can adopt carer roles, but resist or reject the label of 'carer', as they identify as a relative first (Hughes et al., 2013), or could be that those more dependent on carers were not included in the sample.

Further examination of findings from MS interview data during structured interviews with neurologists adhered to grounded theory principles, that recommend iterative processes are used to further scrutinise findings (Charmaz, 2006). This iterative process allowed identification of barriers and potential solutions, which have implications for practice. A unique approach was developed and used during structured interviews with neurologists. Infographic representations depicted findings from interviews with people with MS. This novel method provided a tool to convey key content concisely, enabling focused examination of the findings, in interviews that were limited to 30 minutes.

A patient and public involvement group provided advice throughout all stages of the research (as detailed in chapter 3). They took part in various tasks, including identification of important outcomes for the systematic reviews, improving participant-facing documents, ensuring the research design and findings were relevant to patients, and that important areas were not overlooked (see appendix E).

12.9. Implications for practice

12.9.1. Reconsideration of the promotion of personalised care planning

Findings from the systematic reviews have demonstrated that it is unclear whether personalised care planning is an effective approach in improving the health and wellbeing for people with MS, and that research into this area is considerably lacking. Although government policy identified personalised care planning as a priority for people with long-term neurological conditions (Department of Health, 2005), a review 5 years later demonstrated there is little evidence of its adoption (National Audit Office, 2011), and a survey has shown that a majority of people with MND, Parkinson's disease and MS do not report having a care plan (Peters et al., 2013). This doctoral research suggests it is unclear whether personalised care planning is an approach that works or is feasible in present healthcare contexts. At this stage, further work is needed to examine its feasibility, value and form, alongside resource availability, before further policies promoting its implementation are made.

12.9.2. Examination of how patient involvement is enacted

Two particularly problematic areas were identified during analysis of MS interviews, both of which exemplify some of the challenges of involving patients. Firstly, it appears that when neurologists expect MS patients to choose a DMD independently, they are motivated by an imperative for 'best' practice. Based on literature, policies and guidelines, this approach seemed reasonable. However, problems arise when patient involvement is encouraged (or imposed) indiscriminately in contexts where more nuanced approaches are required.

In the context of decisions about DMDs, there may be some confusion in the way concepts are translated into practice with apparent conflation of consumerist and SDM styles. As explored in chapter 1, terms such as SDM, informed patients and consumerism may lead to confusion when under the umbrella of patient involvement (Moumjid et al., 2007). To further the complexities around SDM, some argue it should accommodate patients' preferred styles of interaction (Elwyn et al., 2000, Towle and Godolphin, 1999). Thus, theoretically SDM can exist in varied forms, some of which appear at odds with archetypal SDM interactional style (depicted in Table 11.1 in chapter 11).

The second area exemplifying a challenge in enacting SDM was the dilemma of how to accommodate patients' preferences for discussion of content that lies outside (or is counter to) mainstream medicine. Ideally, equal partnerships would provide a forum to discuss a broad range of content, expanding further than biomedical factors to other areas deemed important from patients' perspectives (Entwistle and Watt, 2013). Healthcare professionals face challenges in facilitating such discussion without endorsing approaches (Beauchamp and Childress, 2009), or in asserting their judgement, without appearing dismissive. The findings showed that avoiding discussion about such content may be interpreted as dismissive by patients, and in turn discourage future consultations, thus inhibiting useful discussion. In a context where understandings are contested and people with MS are interested in using CAM (Apel et al., 2005, Nayak et al., 2003), it is worth considering the ways to effectively overcome this dilemma.

12.9.3. Information provision

This research has shown that diagnostic stages can be particularly stressful for people with (suspected) MS, and a lack of useful information appears to exacerbate this distress. NICE guidelines (2003) at the time of the interviews recommended that patients' information needs are assessed and reviewed *during* diagnosis investigations. However, this recommendation has been omitted from recently updated NICE guidelines (2014); although they recommend people with MS should be well informed and supported *after* receiving their diagnosis, they do not provide guidance on information provision during the pre-diagnosis period, as previous guidelines did (albeit vague). Based on this doctoral research, early information about the diagnostic processes and acknowledgement of the potential for being diagnosed with MS would be recommended, along with information about the broad range of potential MS trajectories.

People with MS can find choosing which DMD to start burdensome. The findings demonstrate that the provision of information about the decision context and more guidance and advice when required (detailed in box 11.1) might help to reduce the burden felt.

12.10. Directions for future research

As the value and feasibility of personalised care planning for people with MS is unknown, further research is needed to explore this approach in terms of its form, feasibility, suitability and effectiveness for people with MS. Factors identified in section 12.6 provide useful information to design such a feasibility study. A narrative literature review exploring the experiences of people with MS in relation to various stages of personalised care planning may be useful when considering its feasibility.

Although this research recommends that those with suspected and recently diagnosed MS are provided with information about diagnostic processes and a broad range of potential trajectories, it would also be useful to examine the effects of this. As it has been suggested that having information about mild forms of MS might be insensitive and inappropriate for those with particularly active MS, it may be particularly useful to examine how such information is received by those with more active forms of the condition.

Based on the findings of this research, an information aid (see box 11.1, page 261) was designed to reduce the burden felt by people with MS facing decisions about DMDs. Further research is required to assess its effects. This research also demonstrated the difficulties in discussing topics outside mainstream medicine and how the aim to assert professional opinion without appearing dismissive appears to be challenging. In light of this, it would be beneficial to research ways to overcome this challenge responsibly.

Section six
Appendices and Bibliography

13. Appendices

Appendix A – Participants' documents

A.1. MS participant information sheet

Department of Primary Care Health Sciences
Radcliffe Observatory Quarter, Woodstock Road, Oxford, OX2 6GG
E-mail: abi.eccles@phc.ox.ac.uk
Tel: +44 (0)7587 142597

Participant Information Sheet - Interviews
Exploring Experiences of Decision Making for People with Multiple Sclerosis

Researcher: Abi Eccles, doctoral student.
e-mail: abi.eccles@phc.ox.ac.uk Phone: 07587 142597
Post: Department of Primary Care Health Sciences, University of Oxford, Radcliffe Observatory Quarter, Woodstock Road, Oxford, OX2 6GG

Information last updated: December 2012(a)

Why me?
Thank you for expressing an interest in taking part in this study. You have been invited to take part because you have been diagnosed with MS and are over 18 years old. The insights you can share about your experiences will be invaluable to the research. I am looking for people with a range of different experiences, so whether you have had MS for many years or received a diagnosis recently your contribution will be very useful.

What does it involve?
The research process will involve an interview about your treatment and care where we will discuss your experiences of having MS and making decisions. The interview is likely to last approximately one hour. The interview can take place in your home or another venue which is convenient for you. The interview will be audio recorded, then later transcribed and analysed by the researcher.

Additionally, the researcher may invite you to take part in an optional follow-up telephone interview to explore issues further at a time which convenient for you. The follow-up telephone interview is likely to last approximately 30 minutes and will be audio recorded, then later transcribed and analysed by the researcher.

What will my interview be used for?
The interviews will be analysed and findings will be written up as a research document entitled, 'Exploring Experiences of Decision Making for People with Multiple Sclerosis'. This doctoral research aims to identify decisional support needs and improve quality of care provided for people with MS. The findings will also inform research papers submitted for publication in academic journals and will be included in the findings of the Quality and Outcomes of person-centred Care Policy Research Unit (QORU) funded by the Department of Health.

After completion of the research your interview will stored in the ESDS (Economic and Social Data Service) Qualidata UK Data archive. Here the interview will accessible for reanalysis by ESDS approved researchers following their guidelines which are designed to protect the participants' identities. Any content which may identify you will be removed.

Please turn over

Page 2 - Participant Information Sheet

Will my interview be confidential?

Yes. Your interview will be made anonymous and any identifying content, such as names and places, will be removed. The interview transcript and recordings will be kept securely on a password protected hard drive. Direct quotes may be taken from your interview and used to illustrate points; however any content in which you may be identified will be removed in all of the research outcomes.

Can I access the research findings?

A lay summary of the findings will be created and distributed to participants. On successful submission of the research, it will be deposited both in print and online in the University archives with open access. The researcher aims to make findings available before January 2015.

What is the benefit of taking part?

Taking part will enable you to be involved in an exciting research project which may help to improve services for people with MS and more broadly people with long term conditions. The researcher is dedicated to keeping participants informed and will keep you updated on research outcomes. Taking part is voluntary so you will not receive any payments. It is unlikely that you will need to travel, but if you do, travel expenses will be reimbursed.

Are there any risks involved?

The risks involved in this research have been assessed as minimal. It is possible you may feel fatigued during or after the interview. Distress may be caused if there is discussion of stressful previous experiences. You can choose not to answer any interview questions you'd prefer not to, and can stop the interview at any time. Taking part in this research will have no effect on the care you personally receive.

What if I have questions or change my mind?

You will have opportunities to ask questions throughout the research process. If you wish to withdraw from the research, you are free to do so at any stage leading up to and during the interview. There will be no penalty for withdrawing and no explanation is required.

How do I take part?

If you would like to take part in this research project, please complete the reply slip and return it in the prepaid envelope enclosed. Alternatively, you can request an electronic version to complete, send a scanned version of the reply slip or send an email to abi.eccles@phc.ox.ac.uk with the relevant details as requested on the reply slip. Once your reply slip is received the researcher will contact you to arrange the interview.

Concerns about this research?

If you have a concern about any aspect of this project, please speak to the researcher (Abi Eccles, abi.eccles@phc.ox.ac.uk, 07587 142597) who will do her best to answer your query. If you remain unhappy and wish to make a formal complaint, please contact the Research Ethics Committee at the University of Oxford (ethics@medsci.ox.ac.uk; Medical Sciences Inter-Divisional Research Ethics Committee, Medical Sciences Divisional Office, Level 3 John Radcliffe Hospital, Oxford, OX3 9DU, UK).

This project has been reviewed by, and received ethics clearance through, the University of Oxford Central University Research Ethics Committee.

UNIVERSITY OF OXFORD

Department of Primary Care Health Sciences
Radcliffe Observatory Quarter, Woodstock Road, Oxford, OX2 6GG
E-mail: abi.eccles@phc.ox.ac.uk
Tel: +44 (0)7587 142597

Reply Slip

Exploring Experiences of Decision Making for People with Multiple Sclerosis

If you would like to take part in the research entitled: Exploring Experiences of Decision Making for People with Multiple Sclerosis, please complete the form below in block capitals and return it to the researcher in the prepaid envelope. Alternatively, if you would prefer to submit the form electronically, please request an electronic version of the form via email, or you can send a scanned version, or send an e-mail which includes the information requested below. Once you have returned this form the researcher will contact you to arrange an interview.

Name: ……

Address: ……

…… **Post Code:** ……

Phone number(s): Day: …… **Evening:** ……

Email address: ……

Preferred time to receive calls: ……

Year of MS diagnosis: …… **Age:** …… **Gender:** ……

Ethnic background:…… **Occupation:** ……
(It is important to include perspectives from a range of backgrounds in this research)

Do you have any additional needs you feel the researcher should be made aware of?

……

……

……

……

Where did you hear about this research? ……

Please return this form using the prepaid envelope to:-
Abi Eccles, Nuffield Department of Primary Care Health Sciences, University of Oxford
Gibson Building 1st Floor, Radcliffe Observatory Quarter, Woodstock Road, Oxford, OX2 6GG, or via email, abi.eccles@phc.ox.ac.uk

The Department of Primary Health Care is a member of the NIHR National School for Primary Care Research
Head of Department
Professor Richard Hobbs, FRCGP, FRCP, FESC, FMedSci

A.2. MS participant consent form

Department of Primary Care Health Sciences
Radcliffe Observatory Quarter, Woodstock Road, Oxford, OX2 6GG
Tel: +44 (0)7587 142 597
E-mail: abi.eccles@phc.ox.ac.uk

Interview consent form for the project entitled:
Exploring Experiences of Decision Making for People with Multiple Sclerosis

Researcher: Abi Eccles, doctoral student **Contact details:** abi.eccles@phc.ox.ac.uk or 07587 142597

Purpose of the study: To explore and identify decisional support needs of people with MS, with an aim to improve quality of healthcare services provided for people with MS and long term conditions.

Please tick the appropriate boxes

Taking part	**Yes**	**No**
I confirm that I have read and understand the participant information sheet dated December 2012(a).	☐	☐
I have been given the opportunity to ask questions, and I have received satisfactory answers to any questions I have asked.	☐	☐
I understand that the project has been reviewed by, and received ethics clearance through, the University of Oxford Central University Research Ethics Committee.	☐	☐
I understand who will have access to personal data provided, how the data will be stored; and what will happen to the data at the end of the project.	☐	☐
I understand the research will be written up as a student's thesis, and I understand how personal data included in that thesis will be published and stored.	☐	☐
I understand how to raise any concerns or complaints I have about the project.	☐	☐
I agree to take part in the project. Taking part in the project will include being interviewed and audio recorded.	☐	☐
I understand that my taking part is voluntary; I can withdraw from the study at any time without penalty. In order to withdraw I inform the researcher of my decision and I do not have to give any explanation for my decision.	☐	☐

After the interview	**Yes**	**No**
I agree to take part in a follow-up audio recorded telephone interview to explore points raised further. I understand that this is optional and will only take place if I agree to it and at a time convenient to me. There will be no penalty if I decide not to agree to this.	☐	☐

Name of Participant (**block capitals**)	Signature	Date
Researcher	Signature	Date

Reference number: _____

A.3. MS participant copyright form

Department of Primary Care Health Sciences
Radcliffe Observatory Quarter, Woodstock Road, Oxford, OX2 6GG
Tel: +44 (0)7587 142 597
E-mail: abi.eccles@phc.ox.ac.uk

Copyright form for project entitled:
Exploring Experiences of Decision Making for People with Multiple Sclerosis

Please tick the appropriate boxes

Use of the information I provide for this project only	**Yes**	**No**
I understand my personal details such as name, phone number and address will not be revealed to people outside the project.	☐	☐
I understand that my words may be quoted in publications, reports, web pages, and other research outputs. All content which identifies me with be removed.	☐	☐
Use of the information I provide beyond this project	**Yes**	**No**
I agree for the data I provide to be archived at the ESDS (Economic and Social Data Service) Qualidata, UK Data Archive.	☐	☐
I understand that other ESDS approved researchers will have access to this data only if they agree to preserve the confidentiality of the information as requested in this form.	☐	☐
I understand that other ESDS approved researchers may use my words in publications, reports, web pages, and other research outputs, only if they agree to preserve the confidentiality of the information as requested in this form.	☐	☐
So we can use the information you provide legally	**Yes**	**No**
I agree to assign the copyright I hold in any materials related to this project to the researcher, Abi Eccles.	☐	☐

Name of Participant (**block capitals**)	Signature	Date
Researcher	Signature	Date

Reference number: _____

A.4. Neurologist participant information sheet

Nuffield Department of Primary Care Health Sciences
Radcliffe Observatory Quarter, Woodstock Road, Oxford, OX2 6GG
E-mail: abi.eccles@phc.ox.ac.uk
Tel: +44 (0)7786 927834

Participant Information Sheet – Feedback Interviews
Exploring Experiences of Decision Support and Information Needs for People with Multiple Sclerosis

Researcher: Abi Eccles, doctoral student.
e-mail: abi.eccles@phc.ox.ac.uk Phone: 07786 927834
Post: Nuffield Department of Primary Care Health Sciences, University of Oxford
Gibson Building 1st Floor, Radcliffe Observatory Quarter, Woodstock Road, Oxford, OX2 6GG

Information last updated: August 2016

Why me?
Thank you for expressing an interest in taking part in this interview. You have been invited to take part because you a clinician with an interest in this area. The insights you can share will be invaluable to the research.

What does it involve?
The research process will involve taking part in a feedback interview. The researcher will present findings from earlier stages of the research and specific subjects and issues will be explored. The interview is likely to last approximately 30 minutes and can take place in venue convenient to you. The interview will be audio recorded, then later analysed by the researcher.

What will my interview be used for?
The interview will be analysed and findings will be written up within a doctoral thesis entitled, 'Exploring Experiences of Decision Making for People with Multiple Sclerosis'. This doctoral research aims to identify decisional support needs and improve quality of care provided for people with MS. The findings will also inform research papers submitted for publication in academic journals.

Will my interview be confidential?
Yes. The interview recording will be kept securely on a password protected hard drive. Any content in which you may be identified will be removed in all of the research outcomes.

Can I access the research findings?
On successful submission of the research, it will be deposited both in print and online in the University archives with open access. The researcher aims to make the findings available before August 2017.

What is the benefit of taking part?
Taking part will enable you to be involved in a research project that may help to improve services for people with MS and more broadly people with long term conditions. The researcher is happy to update you on any research outcomes, please let her know if you would like her to do so. Taking part is voluntary so you will not receive any payments. It is unlikely that you will need to travel, but if you do, travel expenses will be reimbursed.

Please turn over

Nuffield Department of Primary Care Health Sciences
Radcliffe Observatory Quarter, Woodstock Road, Oxford, OX2 6GG
E-mail: abi.eccles@phc.ox.ac.uk
Tel: +44 (0)7786 927834

Are there any risks involved?
The risks involved in this research have been assessed as minimal. You can choose not to answer any interview questions you'd prefer not to, and can stop the interview at any time.

What if I have questions or change my mind?
You will have opportunities to ask questions throughout the research process. If you wish to withdraw from the research, you are free to do so at any stage leading up to and during the interview. There will be no penalty for withdrawing and no explanation is required.

How do I take part?
If you would like to take part in this research project, please let the researcher Abi Eccles know via email abi.eccles@phc.ox.ac.uk or phone 07786927834. She will then arrange the interview.

Concerns about this research?
If you have a concern about any aspect of this project, please speak to the researcher Abi Eccles, 07786 927834, who will do her best to answer your query. The researcher should acknowledge your concern within 10 working days and give you an indication of how she intends to deal with it. If you remain unhappy or wish to make a formal complaint, please contact the chair of the Research Ethics Committee at the University of Oxford (using the contact details below) who will seek to resolve the matter in a reasonably expeditious manner: Chair, Medical Sciences Inter-Divisional Research Ethics Committee; Email: ethics@medsci.ox.ac.uk; Address: Research Services, University of Oxford, Wellington Square, Oxford OX1 2JD

This project has been reviewed by, and received ethics clearance through, the University of Oxford Central University Research Ethics Committee.

A.5. Neurologist participant consent form

Nuffield Department of Primary Care Health Sciences
Radcliffe Observatory Quarter, Woodstock Road, Oxford, OX2 6GG
Tel: +44 (0)7786 927834
E-mail: abi.eccles@phc.ox.ac.uk

Feedback interview consent form for the project entitled:
Exploring Experiences of Decision Making for People with Multiple Sclerosis

Researcher: Abi Eccles, doctoral student **Contact details:** abi.eccles@phc.ox.ac.uk or 07786927834

Purpose of the study: To explore and identify decisional support needs of people with MS, with an aim to improve quality of healthcare services provided for people with MS and long term conditions.

Please tick the appropriate boxes

Taking part	**Yes**	**No**
I confirm that I have read and understand the participant information sheet dated August 2016.	☐	☐
I have been given the opportunity to ask questions, and I have received satisfactory answers to any questions I have asked.	☐	☐
I understand that the project has been reviewed by, and received ethics clearance through, the University of Oxford Central University Research Ethics Committee.	☐	☐
I understand who will have access to personal data provided, how the data will be stored; and what will happen to the data at the end of the project.	☐	☐
I understand the research will be written up as a student's thesis, and I understand how personal data included in that thesis will be published and stored.	☐	☐
I understand how to raise any concerns or complaints I have about the project.	☐	☐
I agree to take part in the project. This will involve taking part in an audio recorded feedback interview.	☐	☐
I understand that my taking part is voluntary; I can withdraw from the study at any time without penalty. In order to withdraw I inform the researcher of my decision and I do not have to give any explanation for my decision.	☐	☐

_____	_____	_____
Name of Participant (**block capitals**)	Signature	Date
_____	_____	_____
Researcher	Signature	Date

Reference number: _____

A.6. Neurologist participant copyright form

Nuffield Department of Primary Care Health Sciences
Radcliffe Observatory Quarter, Woodstock Road, Oxford, OX2 6GG
Tel: +44 (0)7786 927834
E-mail: abi.eccles@phc.ox.ac.uk

Copyright form for project entitled:
Exploring Experiences of Decision Making for People with Multiple Sclerosis

Clinician Feedback Interviews

Please tick the appropriate boxes

Use of the information I provide for this project only	**Yes**	**No**
I understand my personal details such as name, phone number and address will not be revealed to people outside the project.	☐	☐
I understand that my words may be quoted in publications, reports, web pages, and other research outputs. All content which identifies me with be removed.	☐	☐
So we can use the information you provide legally	**Yes**	**No**
I agree to assign the copyright I hold in any materials related to this project to the researcher, Abi Eccles.	☐	☐

Name of Participant (**block capitals**)	Signature	Date
Researcher	Signature	Date

Appendix B – Recruitment documents

B.1. Recruitment poster

School for Primary Care Research

Increasing the evidence base for primary care practice

Personal Experiences of Multiple Sclerosis (MS)

Do you have MS?
Are you over 18 years old?
Do you live in the UK?

Are you interested in taking part in a study which may help improve services for people with MS by sharing your experiences?

This research explores the personal experiences of people with MS. We are looking for people with a range of different experiences, so whether you have had MS for many years or received a diagnosis recently your contribution will be very useful.

Taking part in the project would involve being interviewed about your experiences of MS, including what your thoughts and feelings have been at different stages, how you have got information and how you have made decisions.

For more information on taking part, please contact researcher:
Abi Eccles via e-mail: abi.eccles@phc.ox.ac.uk Or phone: 07587 142597

This project has been reviewed by, and received ethics clearance through, the University of Oxford Central University Research Ethics Committee

UNIVERSITY OF OXFORD
DEPARTMENT OF PRIMARY CARE HEALTH SCIENCES
QORU policy research unit in quality and outcomes of person-centred care
NHS National Institute for Health Research

B.2. Guest blog on Shift.MS

Exploring MS complexities and decision uncertainties

There's not much we know about MS. There are some things we think we know, with plenty of research being carried out into causes, risks and treatments, but the findings are often inconclusive or do not apply to everybody. One thing we can say about MS is that it is unpredictable. An individual's symptoms, their reactions to treatments and patterns of MS progression may differ widely from another individual's, or even at different stages of their own condition.

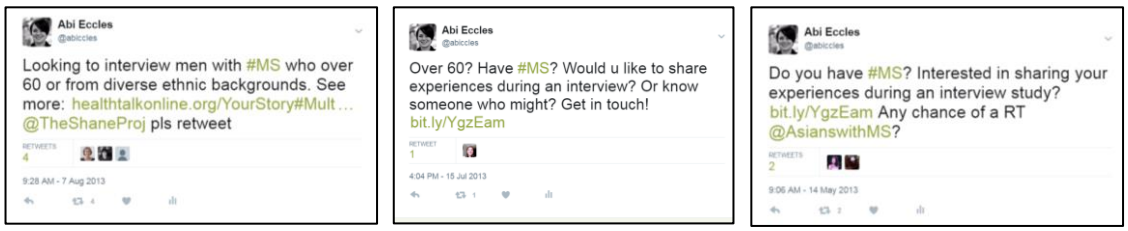
An MS diagnosis is usually received between the ages of 20 and 40; a time which is often associated with establishing life choices and pursuing future plans. Due to its unpredictable nature, people are faced with many uncertainties about how they might expect MS to affect them. There are countless choices about therapies, information use, self-management and support. There are even decisions to be made about the timing of when to make such choices. These decisions are all likely to be shaped by various factors, including a person's values and priorities, social circumstances, the research evidence, availability of choices, side effects and symptoms. Such factors may also be further affected as people live with and learn about what is particular about their own MS experience.

As a doctoral researcher I am interested in understanding how decisions about MS are made, with an aim to identify ways in which decision making support can be provided for individuals. I am carrying out an interview study exploring personal experiences of MS and decision making. If you are interested in taking part by sharing your experiences, please get in touch via phone (07587 142 597) or e-mail (abi.eccles@phc.ox.ac.uk) and I will send you further information.

Abi Eccles

Link to recruitment advert: https://www.phc.ox.ac.uk/research/health-experiences/research-projects/qoru

B.3. Focused Twitter posts



Appendix C – Participants' backgrounds

C.1. Participants' demographic details and short biographies

Table: Information recorded in their participant information sheet

ID code	Pseudonym	Ethnicity	Year diagnosed	Years since diagnosis	Age	F/ M	Employment
MSDM01	Janet	white British	2005	8	50	F	Disability benefits
MSDM02	James	white Italian British	2007	6	33	M	Football consultant
MSDM03	Katy	White	2007	6	41	F	Health and wellbeing
MSDM04	Paul	white British	2010	3	27	M	Project and planning manager
MSDM05	Mary	White	2000	13	61	F	Retired due to MS
MSDM06	Donna	White	2009	4	49	F	Actor
MSDM07	Anna	white Scottish	2012	1	39	F	Manager
MSDM08	Chris	White	2008	5	38	M	Architect
MSDM09	Miles	white British	2012	1	44	M	Supervisor in wholesale
MSDM10	Robert	white Scottish	1998	15	53	M	Community development consultant
MSDM11	Adam	white British	2005	8	36	M	parliamentary research
MSDM12	Oliver	white British	2002	11	57	M	Retired
MSDM13	Melissa	white British	2006	7	46	F	Housewife
MSDM14	Ulrike	white British	2003	10	62	F	retired assistant principle
MSDM15	Ava	white British	1990	23	63	F	Retired typesetter
MSDM16	Ian	white British	2007	6	42	M	Retired due to MS. IT manager.
MSDM17	Sadie	white British	2002	11	50	F	Sales assistant
MSDM18	Joanne	white Irish	2005	8	28	F	Public relations
MSDM19	Laura	white Irish	2009	4	30	F	Journalist
MSDM20	Hannah	black Caribbean/African	2000	13	49	F	PA/admin
MSDM21	Kamila	white Czech	2012	1	29	F	Community support assistant
MSDM22	Clem	white British	2003	10	35	M	Project support officer for a bank

Notes

Biographies written as their status at time of the interview

Information such as type of MS, marital status, living arrangements not recorded in participant information sheets, but may have been discussed during interviews and referred to in biographies.

Acronyms

RRMS: Relapsing remitting MS

SPMS: Secondary progressive MS

PPMS: Primary progressive MS

SDM: Shared decision making

DMDs: Disease modifying drugs

LDN: Low dose nitroxone

LTCs: Long term conditions

CCSVI: Chronic cerebrospinal venous insufficiency

UTIs: Urinary tract infections

CBT: Cognitive behavioural therapy

MSDM01

Janet is 50 years old and receives disability benefits. She was diagnosed with SPMS 8 years ago, but believes she started getting MS-caused symptoms 15 years previous to this. She also has epilepsy and depression. Janet is very interested in medical research and appreciates it when clinicians recognise her knowledge. At one stage, Janet was developing UTIs every other week and participated in a trial to prevent them. On two occasions, Janet brought up the possibility of starting DMDs with neurologists, but was not eligible. Janet is familiar with the archetypal models of patient-clinician interactions, and although her preference is for SDM, she does not believe this approach would suit everyone. She is an avid user of social media and considers herself a "disseminator of information".

MSDM02

James is a football consultant. He has dual Italian British nationality and lives in the UK. He is 33 years old and was diagnosed with MS 6 years ago. Before diagnosis, James described his frustration when his GP referred him to a physiotherapist after he had requested to see a neurologist. This led James to seek help from healthcare services in Italy; here he received an MS diagnosis and started DMDs. Currently, James travels to Italy every 6 weeks to receive treatment but this is becoming difficult to maintain financially, so he would like to share care between Italy and UK. This set-up would involve him alternating 6-weekly treatment sessions between Italy and UK, allowing him continue relationship with consultants in Italy without having to travel there as often. James has written to, and been refused by, five different Healthcare Trusts in the UK.

MSDM03

Katy is 41 years old and self-employed. She was diagnosed with MS 6 years ago after experiencing blurred vision. Katy explained how she did not anticipate her diagnosis and feels her neurologist was insensitive when revealing this to her. At the time, she did not know anything about MS and was devastated when accessing information online. Her neurologist recommended that she should start DMDs, however Katy prefers not to use medication and decided against it. Instead, she sought advice online and discovered that many people reported that stress triggers relapses; this led to her making changing to lifestyle to reduce stress. Currently Katy is not effected too badly by MS and she described herself as 'the healthiest person I know'.

MSDM04

Paul is a 27 years old and was diagnosed with MS 3 years ago. He is married with young children and works as a project and planning manager. He also has epilepsy and is unsure if his seizures are MS-related. Following advice from his neurologist, Paul started DMDs earlier than guidelines recommended. He also drastically changed his diet after researching self-management strategies online. Consequently, he has lost a lot of weight, which worries family members. He chose not to consult clinicians about his diet because he considers the NHS to be 'drug treatment based' and does not wish to be in a situation where clinicians advise against the diet he has adopted.

MSDM05

Mary is 61 years old, retired and was diagnosed with MS 13 years ago whilst living in Spain. She is married, a grandmother and has two dogs. Mary described the fatigue she experiences as incredibly debilitating to the point where she rarely leaves her home. Mary adopts various strategies to manage her MS. She previously bought Sativex (a cannabis derivative) and found it very effective in reducing muscle spasms and stiffness, and anxiety. When it became licensed on NHS she requested it from her GP but was refused. She now, reluctantly, uses cannabis, which she buys via her son. Mary finds swimming relieves her pain, but found travelling to swimming pools impossible and so installed an indoor pool in her home. Mary really values her relationship with her MS nurse, who knows all about her family and she feels she can talk to her about anything.

MSDM06

Donna is a 49-year-old actor. She received her diagnosis 4 years ago whilst in hospital due to a severe relapse. When DMDs were offered to Donna, she did not hesitate as she felt the alternative was to 'do nothing'. The injections did not suit her, so when the opportunity to take part in a trial for an oral DMD arose, she took it. With hindsight, she now knows that she would not return to the injection mode of administration. Donna has delayed telling people she has MS and feels it may have implications for her acting career. However, she is finding it increasingly more difficult to hide. Donna has a positive attitude and carries out lots of fundraising activities for MS-related charities. She discussed end of life preferences and has asked her sister to make healthcare and financial decisions if a situation arises where she cannot.

MSDM07

Anna is 39 years old, works as a manager and was diagnosed with MS a year ago. She is divorced and has a teenage son. Anna described how she had to 'fight to get a diagnosis' and felt she was 'dismissed […] as being a hysterical female'. This led to her carrying out extensive research in order to present her case for diagnosis. She believes the experience of growing up with a dad who also had MS added to her anxiety around this time. After receiving a diagnosis, she had 24 hours to choose which of two DMDs she would like to start. Anna's son is her priority and she explained how the effect on him is a primary consideration when making any decisions. Anna blogs about her experiences of MS and believes this process has helped her to

adjust. She believes MS has made her stronger and enabled her to reassess elements of her life; she described MS as being 'one of the best things that happened to me'.

MSDM08

Chris is 38 years old and lives with his partner and two dogs. He was diagnosed with MS 5 years ago after suddenly experiencing various symptoms such as double vision; vertigo; fatigue; difficulty walking and slurred speech. This resulted in him taking a month off work. Chris works long hours as an architect, which he has reduced in order to manage fatigue. Chris believes he was previously eligible for DMDs and disagreed with his neurologist's assessment of his symptoms. This led him to transfer to a new neurologist, with whom he is much happier. Chris has a good relationship with his GP who listens carefully and spends time finding a solution tailored to him.

MSDM09

Miles was diagnosed with PPMS 1 year ago. He is 44 years old, married with children and works as a supervisor in wholesale. He previously served in the parachute regiment and retired due to a knee injury 25 years ago. He considers himself to have a very strong work ethic (like his dad). After receiving a diagnosis, Miles heard nothing from health professionals for 6 months because he had been lost in the system. During this time, he searched for information about PPMS online and felt shocked and devastated by what he found. Miles is open to try almost anything in order to manage his MS. He attends a rehab centre where he tries different self-management techniques and gains support from others with LTCs.

MSDM10

Robert was diagnosed with RRMS 15 years ago. He is 53 years old, self-employed as a community development consultant and lives with his wife and three daughters. Robert was infuriated when his neurologist told him that little is known about MS and nothing can be done. Various additional factors meant the period during diagnosis was particularly difficult for Robert and he feels he was close to a breakdown. This led to his wife consulting a GP friend and looking into alternative strategies. They decided to make major changes to his diet in order to prevent relapses, which he has found very effective and is now 'second nature'. Although he understood the reasoning, Robert was annoyed that his neurologist would not advise on strategies where evidence was lacking. This led to Robert fundraising £33,000 in order to start a clinical trial into diet effects on MS and it also allowed him to organise two conferences about alternative therapies attended by people with MS and clinicians.

MSDM11

Adam works in parliamentary research. He is 36 years old and was diagnosed with RRMS 8 years ago. He remembers receiving diagnosis during an uncertain period of his life as he was making lifestyle decisions about career, relationships and living arrangements, all of which having MS affected. Although Adam was at first reluctant, he was persuaded by his neurologist to start DMDs. He felt the process of starting DMDs was a sign that his condition was deteriorating and consequently exacerbated his depression. Adam was frustrated that when making a decision about which type of DMD to start because his clinician seemed unwilling to advise him and he felt alone during the process.

MSDM12

Oliver is retired from the car production industry. He is 57 years old, married and he has two grown-up daughters. Oliver was diagnosed with PPMS 11 years ago. He feels frustrated by the lack of information provided and options available to him through the NHS. He only attends 18 monthly check-ups in the hope there will be a medical breakthrough. Oliver found out about his local MS centre by chance, which has been a great source of support for him as it allows him to meet other people with MS and try alternative strategies. Oliver recently paid £10,000 to have CCSVI surgery in the hope that it would improve his condition. However, he is unsure how

effective it has been. Oliver suffers most with fatigue and assesses his capabilities on a day-to-day basis.

MSDM13

Melissa is a 46-year-old housewife who was diagnosed with RRMS 7 years ago. Initially, her neurologist decided not to confirm a diagnosis because he believed it would not be helpful. Melissa disagreed and therefore transferred to a new neurologist, from whom she received a diagnosis. Despite having strong anti-medication preferences, Melissa was persuaded to start DMDs by her neurologist, husband and friend. She soon decided to stop, which she did so without consultation and now feels abandoned by healthcare services. Melissa feels particularly let down my healthcare services after severe hip pain was frequently dismissed as MS-caused. After a year of suffering, it was discovered she had a broken hip. Melissa practises shiatsu and homeopathy. She is due to have CCSVI surgery, which she and her husband re-mortgaged their house to pay for.

MSDM14

Ulrike is 62 years old and was diagnosed with MS 10 years ago. She is a retired assistant principal. Before confirming diagnosis, Ulrike was frustrated when her neurologist interpreted her MRI results as inconclusive and told her to 'go away and wait for things to happen'. She transferred to a new neurologist and received diagnosis. In order to manage her MS, Ulrike maintains a healthy lifestyle by carrying out regular exercise, resting and reducing stress. She also takes DMDs. Ulrike described how she has had to "draw a new parameter about what is possible." Ulrike decided to disclose her diagnosis to very few people and 'recast [her] friendship group'; losing friends and staying close to a few old friends. She feels much happier because of this. Ulrike decided to take voluntary redundancy 5 years ago, became self-employed as an educational consultant for a few years until stopping work completely.

MSDM15

Ava is 63 years old and was diagnosed with MS 23 years ago. Upon receiving a diagnosis she had no idea what MS was; she tried to ignore it and did not seek further information. Around this time her relationship with her business partner began to break down, causing much stress which she believe led to a major relapse. Ava decided to disband the partnership enabling her to reduce stress and pace herself. She volunteered doing admin for a Carers Support group, which helped her regain confidence and socialise with other volunteers. Ava has had Manual Lymph Drainage for 20 years and is certain this has reduced severity of symptoms. She has not been offered DMDs, but would not like to use them anyway as she believes they are damaging. She did not like the idea of various adaptations, such as using a wheelchair, an automatic car and a kindle, but found them not to be too bad in practice. Ava enjoys horse-riding which gives her a 'sense of freedom', she explained how 'you can't run, and you can't dance, but on a horse you [can] actually gallop'. She also founded 'The Red Wheelies'; a synchronised mobility scooter dance group.

MSDM16

Ian is 42 years old, a retired IT manager and was diagnosed with MS 6 years ago. He also has asthma and arthritis. He suffers most with 'random chronic pain', which changes daily and is often severe. He manages his pain with morphine. Ian has many tablets and so his medical cupboard is well organised with a comprehensive schedule to follow. On one occasion, Ian had a major relapse and ended up in hospital. When he told staff it was a relapse, he felt they did not listen. His MS nurse called the hospital, but they still did not seem to listen. It was not until his neurologist called a few days later did they appear to take note and initiated the relevant tests. Whilst in hospital they also reorganised his medication schedule. Ian refused to follow the new schedule, until a clinician redraw it to comply with his normal regime. Through experience, Ian has learnt the best way for him to manage his fatigue is to arrange his days so fatigue hits him at an appropriate time. If he does not do this, it has knock effects the following day. Ian rides a

motorbike. He did consider stopping due to MS, but concluded that the positivity it brings is more important.

MSDM17

Sadie was diagnosed with MS 11 years ago. At the time she had never heard of MS and searched online to find out more. Sadie works as a Sales Assistant and feels she has to prove to colleagues that her MS does not affect the quality of her work. She feels that sometimes work colleagues do not understand and question her if she is sits down to rest or cannot complete certain physical tasks. The only health professional Sadie consults is her GP, whom she trusts very much. Her GP supported her through a difficult past with an abusive ex-partner and she feels she can tell her GP anything. However, Sadie worries that her presentations are always assessed as MS-related when they might be caused by something else. One day Sadie's pain was so unbearable, she overdosed on painkillers and became very unwell. An ambulance was called out to her. The paramedics suggested she could attend a 'pain clinic' as an alternative to taking medication. She dislikes taking painkillers and so requested a 'pain clinic' referral from her GP, but nothing has been arranged. Sadie does not feel she manages her MS, as she continues as normal until she relapses.

MSDM18

Joanne is 28, works in public relations and was diagnosed with MS 8 years ago. She believes she had numerous relapses throughout her teenage years when she lost her eyesight, but always put them down to exam stress. When Joanne first sought help from her GP she was told symptoms were due to being overweight or a new mum. She was concerned about consulting her GP in case they thought she wasn't coping and contacted social services. After eventually receiving a diagnosis she and her husband decided this would be a good time to have their second child, before starting DMDs. As soon as she finished breastfeeding she started her course of DMDs, but she found that they caused nightmares and depression, so she changed to a different type which suits her better. She recently relapsed quite badly, and her neurologist suggested that she may have change to a more powerful drug, which fears due to dangerous side effects. Joanne was keen to meet other similar people to herself with MS as she felt quite alone. She managed to connect with some people online and she is now vice chair of Young Women with MS group. Joanne is badly affected by fatigue, and manages this by taking regular naps and through acupuncture. Joanne looked into having CCSVI surgery and paid 500 euros to have primarily tests carried out, but decided against when her neurologist advised her not to.

MSDM19

Laura is 30 years old and recently had a baby. She works as a Journalist and was diagnosed with MS 4 years ago. Laura felt frustrated when her neurologist did not confirm her diagnosis. Everyone was telling her she was unwell because she was not looking after herself, so when she eventually received a diagnosis she felt relieved there was an explanation. Although Laura and her husband were planning to move house and have a baby, they probably did these things sooner, because of MS. Soon after having their first baby, Laura started DMDs. However, she suffered with side effects and changed to a different type. The new DMD also did not suit her either, therefore they decided this would be a good time to temporarily stop DMDs and have a second baby. During labour with her firstborn, Laura requested an epidural, but was told she could not have one because she had MS. When pregnant with her second child she discovered that it would be fine to have epidural and had it written in her birth plan. She ended up having an emergency caesarean and clinicians still appeared to be nervous about giving her a spinal block, but she was not worried because she had researched it. Although it is always a major consideration, Laura is adamant that MS will not limit her lifestyle. She focuses on the positives, is thankful for good days and tries not to think about the future too much.

MSDM20

Hannah is 49 years old, works as a PA and was diagnosed with MS 13 years ago. She received a diagnosis very quickly, after been referred to hospital on the same day she consulted her GP about tingling in her legs. Hannah felt very depressed after her diagnosis. She remembered feeling her MS was like the 'Sword of Damocles' because she was anticipating her next symptom, but did not know when it would happen or what it would be. Hannah was relieved to discover 'The Shane Project', a support group for people with MS from minority ethnicities. She believes it is important to have contact with people who are similar in order to feel less isolated. Hannah finds listening to others' experiences very useful; she gets ideas from this group and often consults members when making treatment decisions. She also volunteers as a counsellor for the group's helpline to support people newly diagnosed with MS. When having relapses, there is little Hannah can do except monitor symptoms, rest and wait for them to pass. Hannah has found that stress seems to worsen symptoms. In order to prevent stress she has reduced her work hours and changed her social life. She also ensures she rests to reduce fatigue. Previously, she opted not to start DMDs because she feared the side effects. However, she recently started them after a major elapse.

MSDM21

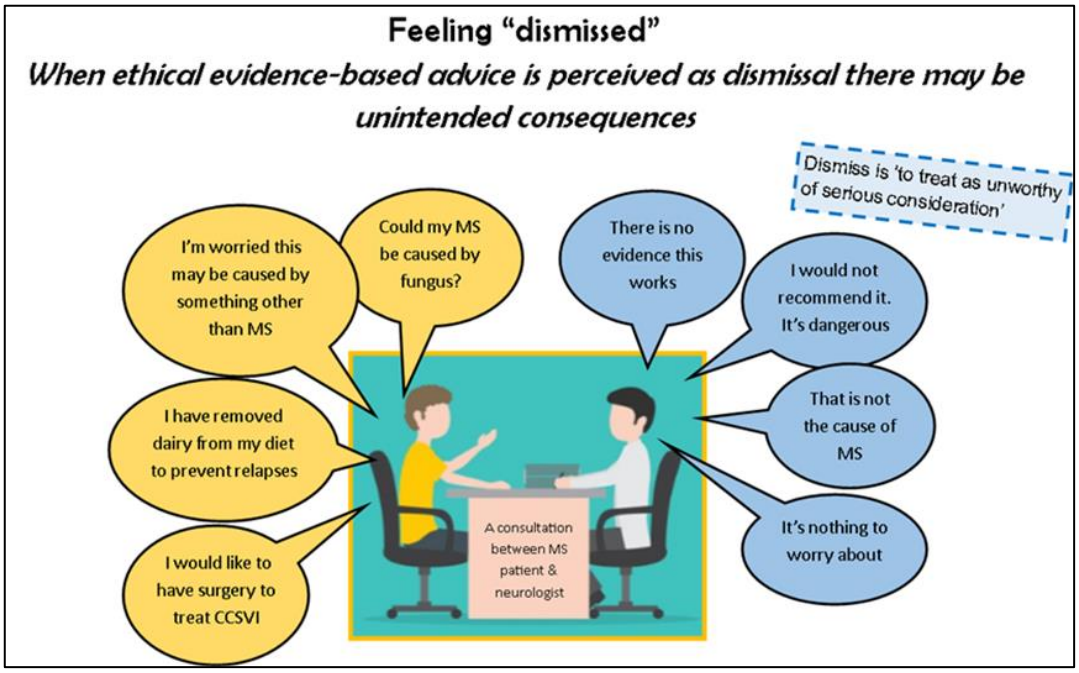
Kamilla is 29 years old and works as a full-time Community Support Assistant alongside being a part-time student. She was informed that she had MS one year ago via letter. The letter was very difficult to understand and contained no follow up information. Kamilla was disgusted at being told in such a way. She had to call the neurology department numerous times over the following weeks in order to arrange an appointment. Kamilla told her dad straight away, but delayed telling her mum because she thought she would make a big deal about it. She has decided not to tell her grandparents, as she does not wish to worry them. Kamilla started DMDs around 10 months ago and is currently finding the side effects difficult to cope with. She chooses not to consult her neurologist or MS nurse about this because of previous unsuccessful interactions. However she does get advice from the pharmaceutical MS nurse, who recommends she continue. Kamilla suffers with anxiety and depression, which was dismissed as unimportant by her MS nurse. She realised the worries she felt were not healthy and so sought a referral for CBT from her GP. Kamilla explained how she likes to be in control, which means the uncertainty of MS and the not being able to do much have been very hard to accept.

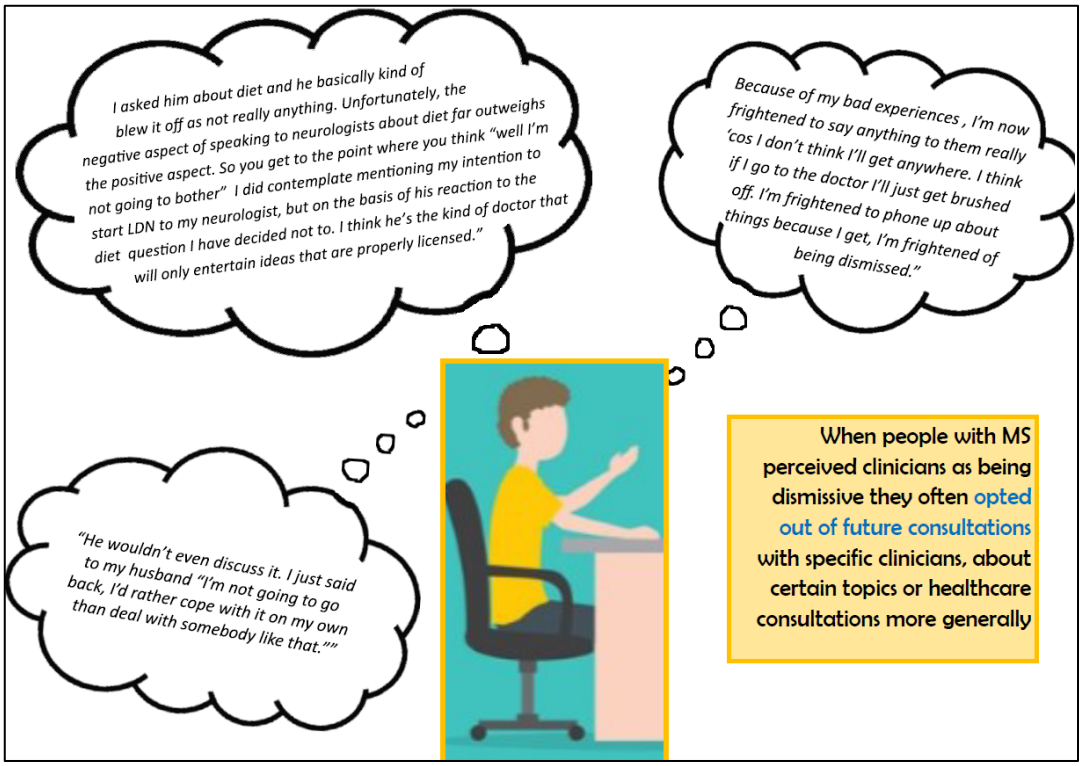
MSDM22

Clem is 35 years old and works as a Project Support Officer for a bank. He received his diagnosis 10 years ago and described it as 'a bolt out the blue'. MS scared him, so he avoided finding out any information and tried to continue as normal. Friends and family sent him information which he kept but never read. Clem's relapses were becoming more frequent and his condition was deteriorating but he did not ask for help. He became reclusive and lived upstairs because he did not have the strength to climb stairs. Clem had started DMDs by this point, but they were not effective and he suffered with side effects. Eventually, Clem got to a crisis situation where he could no longer stand up and was admitted to hospital. Once in hospital he gained access to lots of services he was unaware of. At this point also decided to stop DMDs. Whilst in hospital he also had to make a decision about where to live, because his current home was not suitable for a wheelchair. He decided to live alone in a flat. Soon after moving into his new home, Clem was readmitted to hospital with a distended bladder and learnt how to self-catheterise. Whilst in hospital he also started a new DMD, but unfortunately it caused liver damage. This led Clem to start another DMD which had a potentially deadly side effect. He has been on this DMD for the last 4 years and his condition has stabilised. He described how he was quite fatalistic in past and he thought there was nothing that could help, but has now learnt to ask for help sooner rather than later.

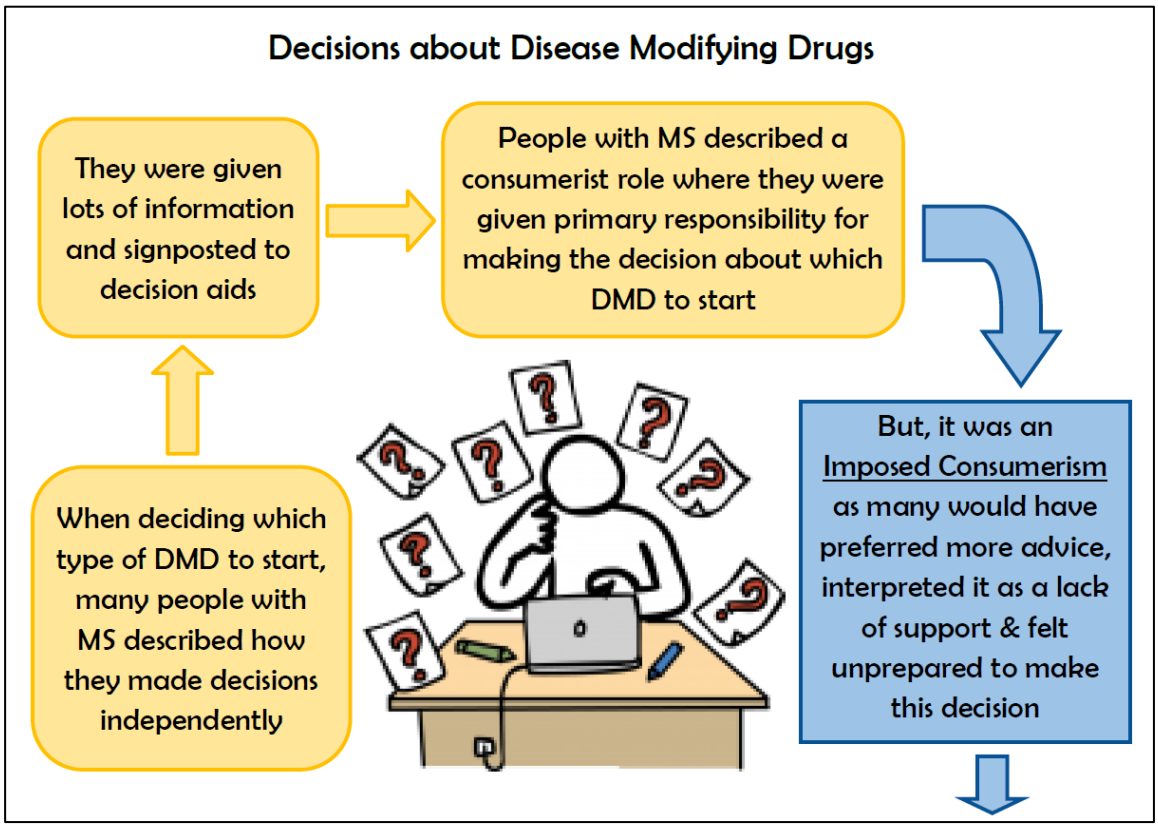
Appendix D – Infographics used during structured interviews

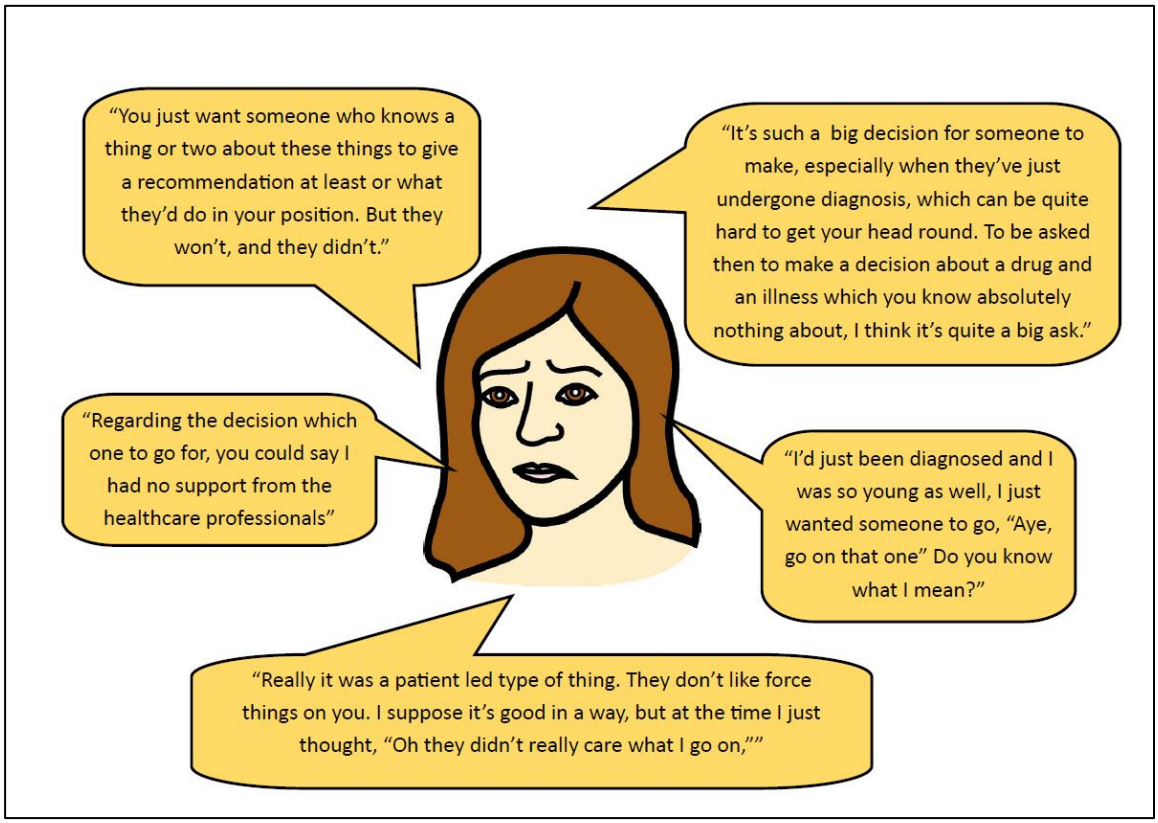
D.1. Infographic illustrating the theme: Feeling Dismissed





D.2. Infographic illustrating the theme: Imposed consumerism





Appendix E – Patient and public involvement

E.1. Role description

Role description for 'Expert Patients'

Members of 'Expert Patients' will share perspectives and carry out assignments to inform the design, analysis and dissemination of research about the support needs of people with Multiple Sclerosis (MS) and long term conditions (LTCs).

The group will exist online. A secure website will be created and will host information about the research and assignments available to volunteer for. Members will receive notification of each new assignment via e-mail and will be given instructions on how to access material, complete tasks and submit assignments online. Examples of tasks include: feedback on research design and interview schedule and reading and commenting on a couple of transcripts. The researcher will contact each member by phone to welcome them and answer any questions. Members are able to contact the researcher via the website, e-mail or phone with questions or support needs within working hours at any point during the research process.

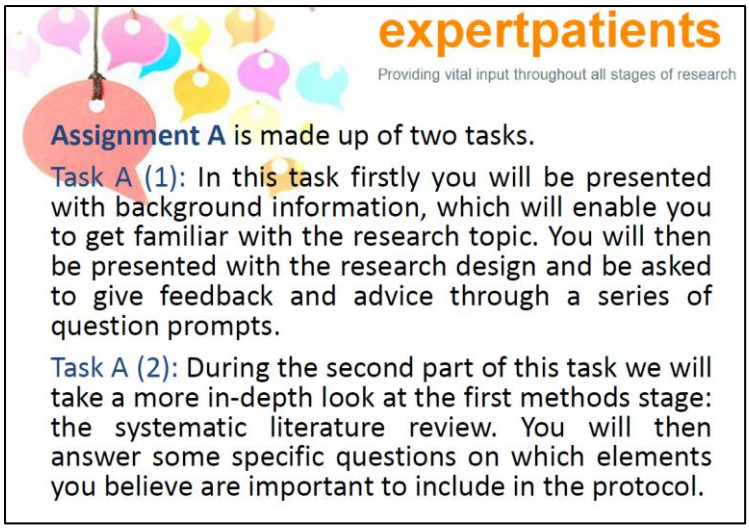
Members will be expected to devote half a day (3.5 hours) to each assignment and in return be paid £75. Some assignments may include more than one task and some tasks may span across more than one assignment. As 'Expert Patients' exist online it is unlikely that there will be a need to hold meetings, however if meetings are arranged then members will be paid for their time, at £75 per half day, and travel expenses will be reimbursed.

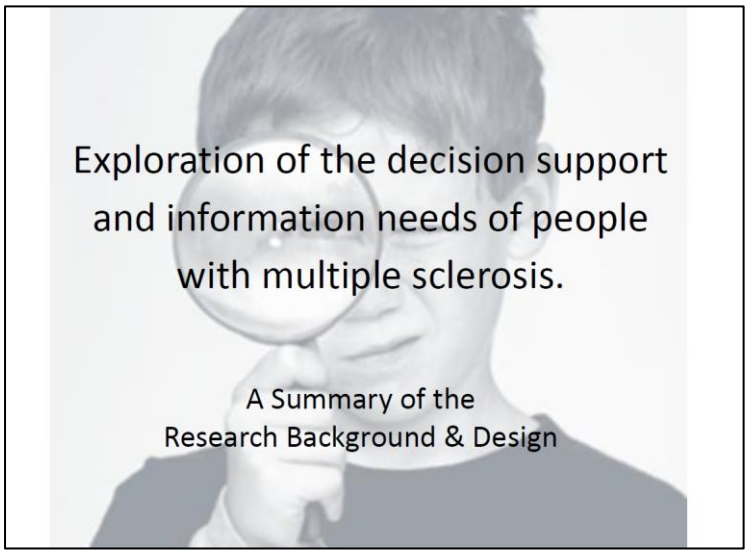
'Expert Patients' will form in November 2012. The assignments are not confirmed at this stage but it is envisaged there will be approximately 3-6 assignments available to volunteer for each year. Members of the group must have access to a computer and the internet, be over 18 years old and have experience of least one long term condition.

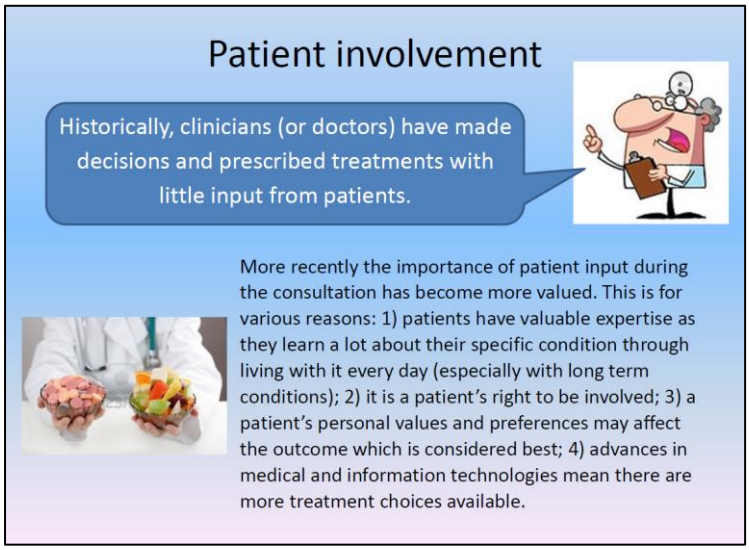
Criteria	**Essential attributes**	**Desirable attributes**
Condition experience	*Patient or carer experience of at least one long term condition.*	*Patient or carer experience of multiple sclerosis (MS). Such members are desirable because some of the assignments will be MS specific.*
Computer skills	*Good computing and internet navigation skills.*	*Excellent computing and internet navigation skills.*
Communication skills	*Competent written communication skills.*	*Good written communication skills.*
Personal qualities	*Self-motivated, analytical and able to give attention to detail.*	

The research is being carried out by Abi Eccles at the Department of Primary Care Health Sciences at the University of Oxford and is funded by the NIHR School for Primary Care Research.

E.2. Assignment A and C – Project feedback and prioritisation of outcomes







However, clinician input is still as valued as ever; increased patient involvement does not mean reduced clinician involvement. Clinicians are experts in diagnosis, prognosis, treatment options, causes and prevention strategies.



The concept of **'shared decision making'** is where patients and clinicians work together in equal partnership, sharing unbiased information and valuing one another's input when making decisions and planning care. This approach is argued to be the most ethical and effective approach to medical decision making, however it is yet to become the 'norm' to consultations which are still mostly led and controlled by clinicians.



Multiple Sclerosis

Multiple sclerosis (MS) is a neurodegenerative condition. MS affects people in different ways and symptoms may include pain, fatigue and motor loss. Typically people with MS experience gradual long term impairment, alongside episodes of symptoms (relapse) and recovery (remission).

However, MS is a highly variable condition as symptoms, relapse triggers and treatment effectiveness vary between and within individuals.



Decision making in multiple sclerosis (MS) is characterised by **uncertainty** due to the condition being highly **complex** and **variable**

Research Aim

This research aims to explore the decision making experiences of people with MS. It aims to identify the ways in which clinicians can support patients in decision making and care planning at different stages of the condition.

Research Methods

There are three methods stages within this research.

1. A **systematic literature review** will be carried out to assess the effects of personalised care planning.

Personalised care planning is a process where patients and clinicians work together to set priorities and goals and plan ways to achieve these. During this phase we aim to understand the effects of personalised care planning by reviewing all the existing research evidence.

2. **In depth interviews** will be carried out with people with MS to find out about their own personal experiences.

3. The findings from the first two stages (above) will be presented in **discussion groups** with people with MS and clinicians.





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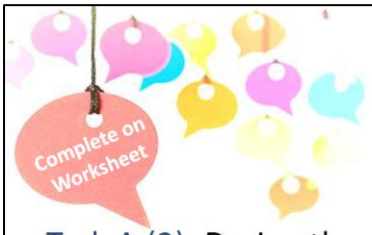
Task A (1)

Spend some time thinking about the research presented on the previous pages.

a) What are your thoughts about this research? Do you have any general feedback?

b) Do you have any advice or suggestions? Is there anything missing? Anything you would change?

c) Is there anything which is unclear?



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Task A (2)

Task A (2): During the second part of this task we will take a more in-depth look at the first methods stage: the **systematic literature review**. You will then answer some specific questions on which elements you believe are important to include in the protocol.

Further Details on the Systematic Literature Review

What is the review question?

'What are the effects of personalised care planning for patients with long-term conditions as compared to usual care?'

What is personalised care planning?

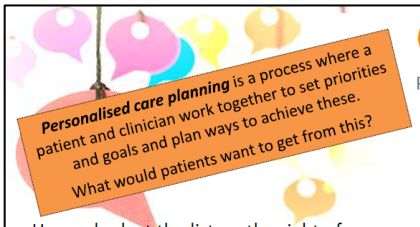
It is a process where a patient and clinician work together to set priorities and goals and plan ways to achieve these. It is promoted by government policy, but it is uncertain how effective this approach is in reality.

How will the review be done?

The researchers will carry out an extensive search with an aim to find all the existing research which tests or trials personalised care planning. We will then review these studies and summarise the findings.

How can the expert patients group help?

Within the literature there are many outcome measurements to assess the effectiveness of personalised care planning. We are unsure which we should assess as being important. We would like to hear your opinions on these.



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Task A (2)

Have a look at the list on the right of possible outcomes from personalised care planning.

Think about what you would want to get out of personalised care planning? Which do you think are important for people with long term conditions?

Now rank them in order of importance with (1) being the most important outcome and (4) being the least important outcome. Please feel free to add any extra notes and comments to explain your choices.

Do you have any further comments ?

List of outcomes:-

- **Improvement in health and wellbeing**

(e.g. changes in levels of fatigue and pain, body weight, self-esteem, anxiety, depression, coping, quality of life, biomedical indicators of condition progression)

- **Capabilities for managing condition**

(e.g. Improved understanding of health and information, self-management skills, ability to access relevant support)

- **Change in health–related behaviours**

(e.g. improved diet, exercise, use of relaxation techniques, condition-relevant self-monitoring, self-medication)

- **Change in use of formal health services**

(e.g. reduced hospital admissions, effective use of healthcare services)

E.3. Assignment B – PPI task giving feedback on recruitment documents

Worksheet - Assignment B

Please spend no longer than 3.5 hours on this assignment. Once completed please submit the assignment to researcher Abi Eccles before 14th December 2012

During the second phase of the research we will be carrying out in-depth interviews with people with MS. In task B (1) you will be asked to give feedback on paperwork which is being prepared for potential interviewees. Then in task B (2) you can provide any further advice you may have for recruitment.

Task B (1) – Recruitment documents

There are 4 pieces of paperwork we would like you to give feedback on:-

1) **Recruitment advert**

The recruitment advert will be used to encourage people with MS to volunteer to be interviewed.

2) **Patient information sheet**

The patient information sheet will be distributed to potential interviewees who are interested in finding out more about the project. Interviewees must read and understand this before taking part.

3) **Consent form**

The consent form will be filled out immediately before the interview. It is to confirm that the interviewee has consented to be interviewed.

4) **Copyright form**

The copyright form will be completed after the interview. It gives the researcher Abi Eccles copyright of the interview and allows the anonymised transcripts to be used in other research projects.

Take some time to have a look at each of these 4 documents and consider them in terms of:

- How understandable it is. Is anything unclear?
- How attractive it is. Do you like the look of it? Would you change anything?
- How easy it is to use. Can you foresee any difficulties potential interviewees may have?
- Any other general comments. Do you have any suggestions?

Complete the table below each document to record your evaluation and make any comments and/or suggestions.

1. Recruitment advert The recruitment advert will be used to encourage people with MS to volunteer to be interviewed.		
	Yes/No	Comments and/or suggestions
Understandable		
Attractive		
Easy to use		
General comments		

2. Participant information sheet

The participant information sheet will be distributed to potential interviewees who are interested in finding out more about the project. Interviewees must read and understand this before taking part.

	Yes/No	Comments
Understandable		
Attractive		
Easy to use		
General comments		

3. Consent form

The consent form with be filled out immediately before the interview. It is to confirm that the interviewee has consented to be interviewed.

	Yes/No	Comments
Understandable		
Attractive		
Easy to use		
General comments		

4. Copyright form

The copyright form will be completed after the interview. It gives the researcher Abi Eccles copyright of the interview and allows the anonymised transcripts to be used in other research projects.

	Yes/No	Comments
Understandable		
Attractive		
Easy to use		
General comments		

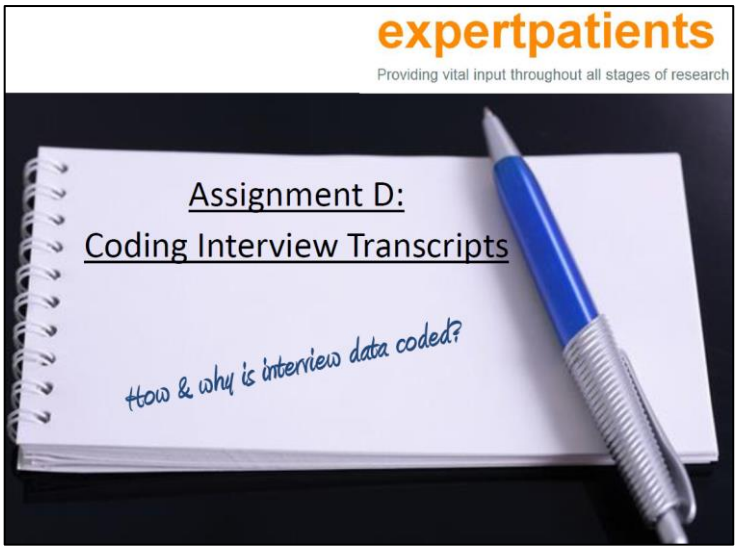
Task B (2) - Recruitment tips:

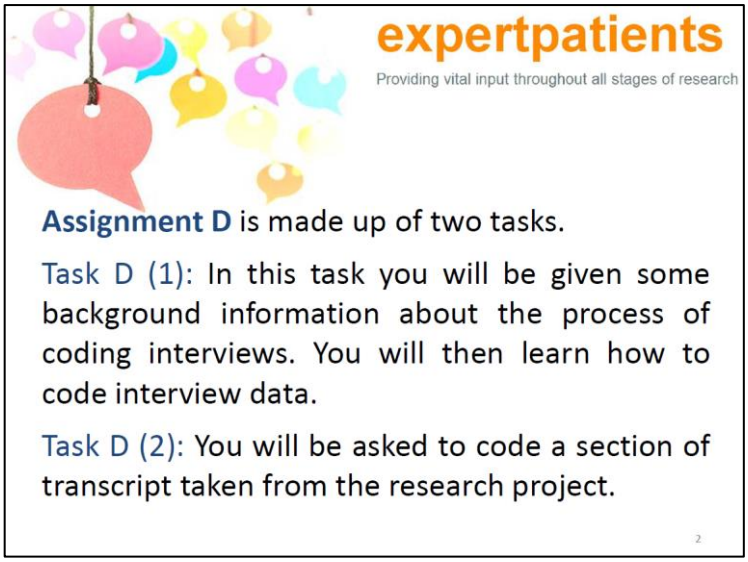
Do you have any advice or suggestions on areas or places where we could aim recruitment? Where would be a good place to distribute adverts?

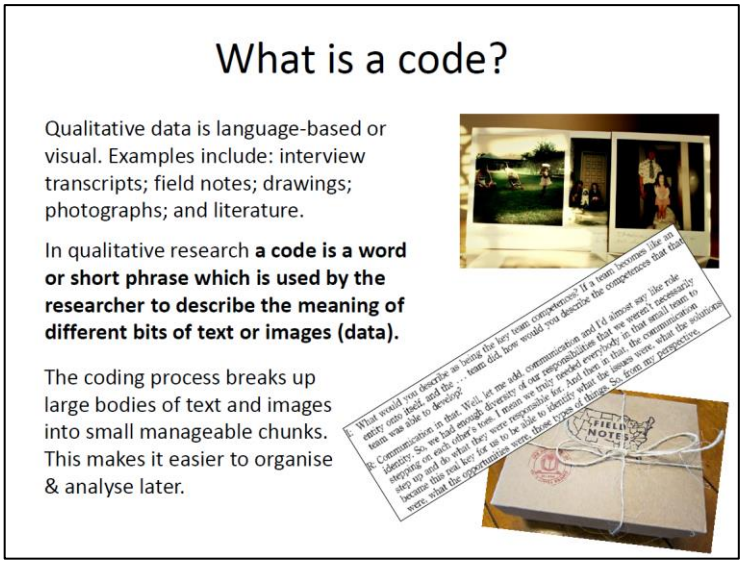
Are there any groups of people with MS who are often overlooked in research which we should try to aim recruitment at?

Do you have any other recruitment tips?

E.4. Assignment D – Coding task



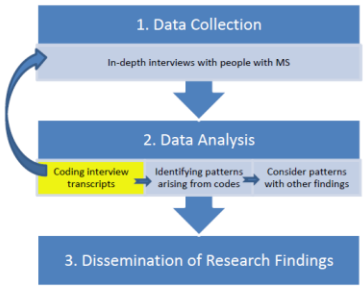




Where Coding Appears in the Research Process

Once interviews have taken place they are transcribed. The researcher then has a text version or transcript of the interview to analyse.

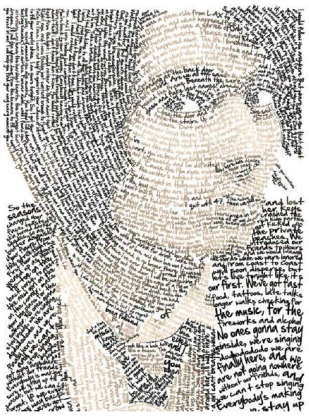
The researcher then codes the interview transcripts. This process involves looking at the content of the transcript in detail. Then words and phrases (codes) are developed to describe each bit of text (datum).



The coding of interviews starts as soon as the first interview is collected and continues alongside the collection of new data (future interviews). On-going data collection is shaped by on-going data analysis as emerging patterns are explored further during interviews. Data collection will only stop when the researcher is certain that no new codes are left to explore. This is called 'Data Saturation'.

4

Why are interviews coded?



The data collected from in-depth interviews is language-based. It is transcribed and presented in a verbatim text format. At this stage it is unwieldy and difficult to make sense of.

This raw data needs to be systematically processed into a format which can be analysed in a meaningful & practical way.

Coding enables data/interview content to be organised and grouped with similarly coded data/interview content. Then, once the data has been organised patterns may be identified.

5

How to Code Interview data

You are now going to learn how to code interview data. This is in order to prepare you for Task 2 where you will asked to code a section of interview from the main research project.

Guide to Coding

1. Carefully read through the interview transcript.
2. Whilst doing this think of codes (phrases or words) to describe or symbolise the content of the text.
3. Make a note of these codes and assign them to the relevant part of the text.
4. Much of the text may have more than one code.
5. The same codes are often frequently repeated within and between different interviews.
6. Reread the text until you feel happy you have covered all the descriptions or codes and assigned them to the text.

(This guide will be outlined again later)



On the next page you will see an example of some coding.

6

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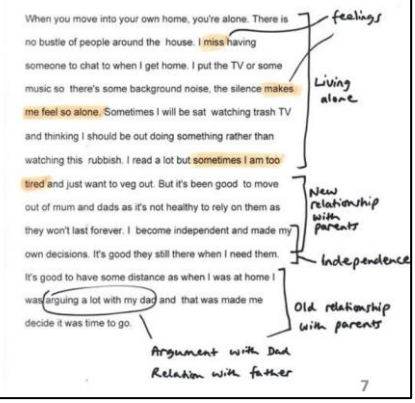
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Coding Example

In this example the researcher has coded a transcript where someone discusses moving out from their parents' home to live alone.

Spend a couple of minutes looking through this and thinking about the codes which have been assigned.

Different researchers might assign different codes. Can you think of any other codes you might add? If so, make a note of these on the worksheet.



7



Task D (2)
Coding Interview Data

BACKGROUND: **In Task D (2) you will be asked to code a section of interview which has already been coded**

The purpose of this task is to carry out a 'coding audit'. The codes you generate will be checked against the codes generated by the researcher. This checking process will confirm the relevance of some codes and it may identify important new codes which haven't yet been considered.

Your perspectives and interpretations of the interview data can provide a new way of looking at the data and will help to ensure that the interview data (expanding further than the text you code) is analysed effectively and rigorously.

Coding sometimes seems like a difficult and confusing task, but it can be enjoyable too. Please approach this with an open-mind; don't worry about it too much and do note down **any** codes you think might be relevant. There are no 'silly' or 'wrong' codes.

8

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Complete on Worksheet

Task D (2) - Coding Interview Data

Three short extracts from interview transcripts can be found in your worksheet. It is important that you do not share this transcript or information from this transcript with any one else. Carry out coding using the guide below. Note the codes you identify the column alongside the text it refers to.

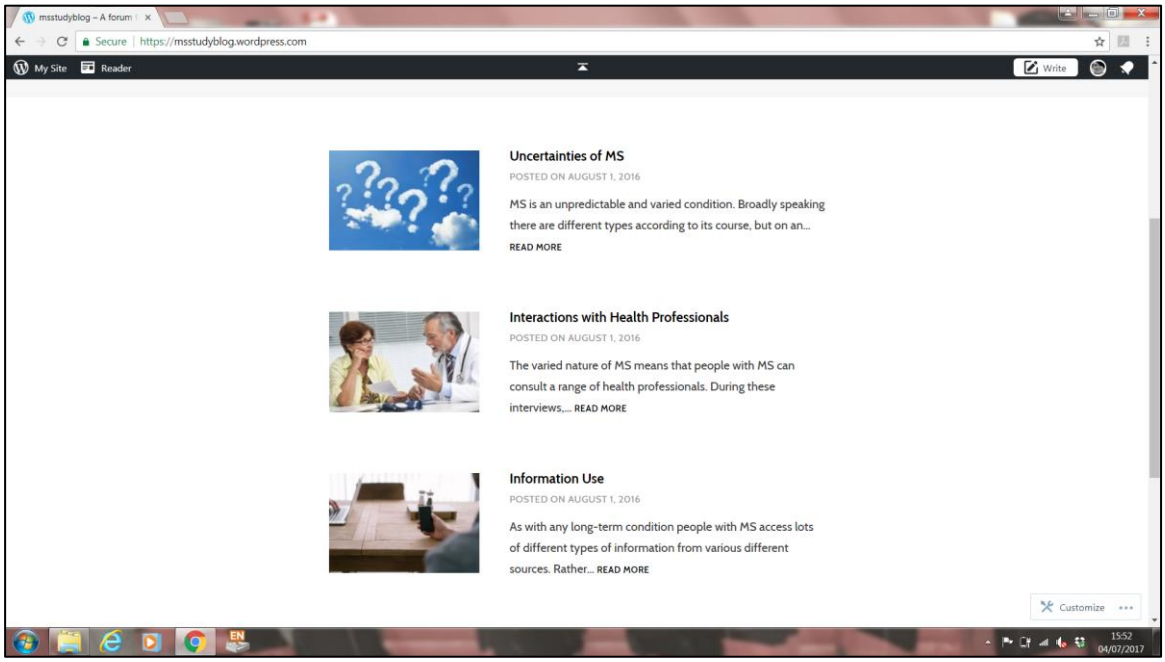
Guide to Coding

1. Carefully read through the interview transcript.
2. Whilst doing this think of codes (phrases or words) to describe or symbolise the content of the text.
3. Make a note of these codes and assign them to the relevant part of the text.
4. Much of the text may have more than one code.
5. The same codes are often frequently repeated within and between different interviews.
6. Reread the text until you feel happy you have covered all the descriptions or codes and assigned them to the text.

Once you have finished this return the worksheet to Abi Eccles in the prepaid envelope. 9

E.5. Assignment F – Feedback on findings

Screenshot of study website



Blog posts which PPI members discussed

How do people with MS discuss their perceptions and use of information in relation to managing their condition?

Introduction

As with any long-term condition people with MS access lots of different types of information from various different sources. Rather than presenting all the ways in which those interviewed perceived and used information, this blog presents the findings that were particularly interesting and relevant to the experiences of people with MS.

Information around the time of diagnosis

The period before, during and after diagnosis was a particularly important according to people interviewed, in terms of the information they accessed. MS is difficult to diagnose; after first symptoms, other causes need to be ruled out and more evidence is needed (from tests and further symptoms) to confirm a formal diagnosis. This means there is often a period of time where the person with suspected MS does not know what is wrong with them and may feel concerned they have MS or something else serious.

Many people interviewed talked about how they were told they probably had MS, but nothing could be done until they were diagnosed. This often led to feeling of frustration and distress. Although this waiting period may be unavoidable, frustrations were often aimed at health professionals; some thought they were insensitive and wanted more information during this phase and some thought it was in the health professional's power to diagnose MS and could not understand their reluctance to do so. Other participants said when they did receive an MS diagnosis they were particularly shocked, because it seemed like MS had previously been ruled out.

At the time of the interviews, NHS guidelines stated that the information needs of people with (suspected) MS are important to consider during the time around diagnosis and that individual information needs should be assessed and responded to. Interestingly, this recommendation is missing from the recently updated version of these guidelines. Although they recommend people with MS should be well informed and supported *after* receiving their diagnosis, they do not provide guidance on information provision during the *pre-diagnosis* period, as previous guidelines did. The findings from these interviews provide an argument for reintroducing this recommendation. However, it is likely reintroduction of such guidance alone will not be enough (as the recommendation was in place at the time of the interviews). There needs to be careful consideration about information provided during the diagnostic stages to ensure that information needs are responded to and distress is keep to a minimum.

Even once diagnosed, many people interviewed said they felt frustrated because they were not given enough information. This meant that they often carried out their own research instead and typically found information they accessed distressing. As shown in the uncertainties blog post, MS is unpredictable and potentially severe, so when people find out they have MS it's common to feel shocked and uncertain about what the future holds. Soon after diagnosis participants said they came across negative examples of MS, sometimes accessed by chance (in waiting rooms or in media stories) or sometimes during their searches for more information about MS. Such negative examples of MS made them feel worse and fear what their futures might hold. However, the people interviewed found that with time, these perceptions changed and many imagined their futures as being more similar to what they imagined before diagnosis. If people recently diagnosed with MS were provided with information, which presented a broad range of MS examples (including milder courses) then it may help people to have more measured views of how MS may affect them and alleviate some of the distress experienced during this time.

Avoiding stress is important in itself, as most of us would prefer not to be stressed. However, it may be particularly important for people with MS. Not only did several of the people interviewed directly link distress experienced around the time of diagnosis to a worsening of their condition, but the link between stress and MS relapses is increasingly recognised by researchers and academics. This provides further support that aiming to reduce distress during this stage through carefully considered information, might improve health and wellbeing for people with MS.

Information suitable for consultations

The people interviewed talked about how they considered some types of information more appropriate than other types to be discussed during healthcare consultations. They recognised that there were various factors which affected which topics were appropriate that included politics, drug company interests and professional stances. For example, some talked about how it was more likely that research is carried out into strategies where money can be made

(such as medication) than other approaches (such as diet change), therefore because there is less research, there is less evidence in support of diet change. This meant that many said they did not discuss self-management strategies and how to live with MS in consultations and looked elsewhere for advice on these topics. Many said that they wanted information about the practicalities of living with MS and found largely medical-focused information gained from health professionals less useful. Some (especially those who drugs were not suitable for) went as far as to say they got nothing from health professionals, as they only monitored their conditions and sometimes made them feel worse. In a condition like MS, where treatments are limited and do not suit everyone, then it may be particularly useful for them to discuss self-management and alternative approaches with health professionals.

Experience based information

MS is a condition that is particularly difficult to measure with technological instruments; there are MRI scans, but their direct relationship to MS activity is unclear and they are not regularly available. MS activity is normally assessed through experience and so it was unsurprising that participants found information based on their own experiences or experiences of others very useful.

As a condition that varies between individuals, most learn about *their* MS through experience. There are some well-known relapse triggers, but the people interviewed learnt which behaviours caused problems for them personally through experience and often changed behaviour in light of this. Although participants relied on their own experiences to understand their condition, they also talked about its limitations. Sometimes it was difficult to work out whether an intervention really worked or whether the act of *doing something* made them feel better i.e. they were unsure whether they were experiencing a placebo effect. Interestingly, this did not seem to bother most as they said what mattered was that they had found something that they believed in.

The nature of MS creates a context where few clinically proven treatments are available and there seems to be an openness to try alternatives, but impossibility to assess interventions' effects on an individual level. The variable nature of MS may mean that treatment effects really do vary between individuals to the point where there are lots of methods, i.e. they all have true effects, that work differently for different people's MS. Or alternatively, the difficulties in assessing effectiveness coupled with openness to try alternatives (due to lack of mainstream solutions) has led to confidence in a wide range of approaches.

Others with MS

As expected these interviews showed how people with MS use others' experiences to help with the real life practicalities of managing MS. Others' experiences provided signposting to services, tips on every day challenges and helped them to think through problems. Participants also said that sharing experiences was particularly useful compared with advice from health professionals, because they could not provide the same empathy as others with MS can. Participants found that much of the official information about MS was generic and did not seem to apply to them. This is likely to be because MS is so variable that generic information is difficult to create or often does not apply in reality. The people interviewed described how they tailored the information they used and sought out advice from others who had experiences which were particularly relevant to their situation.

If used to assess the effectiveness of treatments, then most would consider anecdotal accounts alone as poor evidence, because factors in addition to the treatment might affect the

outcomes. There is a danger that if approaches seem effective for individuals, it may convince others that they will work for them too. Much of the academic literature claims that patients do not tend to use anecdotal information in this way; instead anecdotal accounts are used as tools to think about situations, rather than evidence of effective approaches. This was apparent in the interview accounts as many did use others' experiences in this way, but there were also some who seemed to consider anecdotal accounts as evidence of a treatment's effectiveness. This becomes more concerning when the stakes are high. For example, people may invest a lot of money or hope in an approach, or there may be risky side effects associated with it. However, in a situation where regulated medicine offers very little and some feel concerned about how their MS will progress, it may reasonably be worth investing a lot and taking risks, based on poor evidence. In such instances, it may be useful for health professionals to engage people with MS in discussions about alternative approaches and the limitations of anecdotal accounts, to help them make well informed decisions.

Many of those interviewed found peer support from those with similar backgrounds particularly useful. (That is not to say that peer support from those with different backgrounds was *not* helpful, but it was clear that contact with people with similar backgrounds was particularly useful for some interviewed). After attending support groups and coming into contact with others they felt they could not relate to, they often looked elsewhere for people similar to themselves. For example, some joined groups whose members were made up of young adults who were leading busy active lives, another participant found a support group for those from ethnic minorities particularly useful. Seeking out those who they can identify may be a rejection of the negative examples of MS encountered during initial searches. Those who did find examples of people similar to themselves, said they wished they had discovered these sooner at around the time of diagnosis.

[COMMENTS]

How do People with MS describe their Interactions with Health Professionals and their Impact on their Relationships?

The varied nature of MS means that people with MS can consult a range of health professionals. During these interviews, people mostly talked about their interactions with GPs, MS nurses and neurologists. Their accounts varied widely and it was clear that they had experienced different approaches from the various health professionals they had seen. Patterns of similar experiences between accounts are explored below.

Attentive health professionals

People interviewed rarely talked about their health professionals being particularly attentive. This is not to say that health professionals are generally not attentive, but it seems that negative encounters (where healthcare professionals were less attentive) were brought up more readily, perhaps because they were more memorable.

As you might expect, people interviewed said they particularly appreciated times when health professionals seemed more attentive. This may have been by addressing their preferences,

asking for their opinion or acknowledging their knowledge about medical aspects of MS. Much is written about the importance of patients' values, preference and personal experiences in decision-making, but there is less recognition of their medical knowledge. However, in these interviews, instances where health professionals recognised patients' medical knowledge (and sometimes the limitations of their own knowledge) made people with MS feel empowered and respected. This recognition and subsequent empowerment could work to level out unequal power positions between health professionals and patients and allow discussion from more equal perspectives. However, there is a danger that some people with MS may feel pressured if health professionals assume they have medical knowledge (although there are no examples of this in this set of interviews).

Feeling "dismissed"

Many of the people interviewed talked about times when they felt health professionals had dismissed information they put forward in consultations (i.e. they seemed to treat information as unworthy of serious consideration). According to interviewees' accounts, this always had damaging effects to the point where they chose not to consult in future. This may have been about specific topics, with specific health professionals or healthcare services more generally.

In an ideal consultation, patients' views and clinicians' expertise alongside research evidence will all be discussed. In reality, the way in which these aspects are balanced is challenging, especially if they come in conflict with one another. Of course, we do not know why health professionals appeared dismissive, but they may be in difficult positions if patients suggest ideas that are unproven or go against professional understandings. Dismissing such ideas is perhaps ethical, because anything else could be seen as support for them. The question remains whether it is part of a health professional's job to discuss approaches arguably outside their profession. They may not feel confident in talking about subjects they are untrained in, or there simply may not be enough time available in a health system that is already strained.

But where does this leave the person with MS who would like professional advice? MS is an uncertain condition with many different theories about causes and treatments that people with MS may discuss with others or see in the media. Disease modifying drugs (DMDs) are not suitable for everyone and there may be little else regulated medicine can offer. In such a context, alternative approaches and theories about causes can be very appealing. When people interviewed felt dismissed, not only did this appear to make them less likely to consult in future, but they also did not seem to be put off; rather they went ahead without useful discussion with health professionals. Health professionals may be unaware of the potential for such consequences i.e. rather than putting people off an alternative approach, dismissal can instead put people off future consultations.

It might be more useful if health professionals refrained from appearing dismissive, explained reasons why they would not recommend such approaches and be open the possibility that people with MS may still choose alternative approaches in light of this advice. Perhaps topics that are worth serious consideration during consultations should be guided by preferences of patients rather than which treatments are licensed i.e. patients decide which topics are worth serious consideration.

Overlooked points of view

Some people interviewed talked about times when their point of view seemed to be overlooked or undervalued during consultations. Examples of this included individual preferences about medication use, personal experiences of MS and treatment effects. Health professionals can make incorrect assumptions about (or 'misdiagnose') patients' preferences which may lead to unfavourable outcomes. Although incorrect assumptions about patients' preferences can be as damaging as medical misdiagnoses, they are less obvious and often go unnoticed. Overlooking a patients' individualised experience of a condition or treatment effects could potentially lead to inappropriate treatment or flawed assessments. As MS is a condition that can take many different forms, it is not always clear whether health problems are MS or something else. This means people may get to know a form of MS that is specific to them. This could mean it is particularly important for health professionals to take note of information gained from patients' experiences of their condition and treatments.

Related to the issue of overlooked points of view, some people interviewed also said they did not always feel comfortable voicing their opinions during consultations. Even when they got on well with and described health professionals as approachable. When asked why they did not put forward their point of view, they explained that it was because health professionals were *the experts* and it was *their job* to make such judgements. As many academics have argued before, the traditional roles of health professionals and patients act as barriers against patients being involved in decision-making, i.e. patients typically do not expect, and are not expected, to put forward their opinions. Studies have shown patients' reluctance in voicing all of their concerns during consultations and some health professionals appear hesitant to encourage this due to time pressures and practicalities. As the things currently stand, if we want patients to feel comfortable in putting forward all their opinions, health professionals are the ones who have to instigate this, because as things currently stand they lead consultations.

Imposed choice

One of the most interesting findings from the interviews concerned decisions about which type of disease modifying drug (DMD) to start. There are 11 different types of DMDs and the decision about which one to start is complex. There are many things to weigh up such as benefits, side effects, different methods of taking and storing them and their effectiveness (which can differ between individuals and at different stages of one person's MS). Research into DMDs is ongoing with few studies that directly compare different types, so scientific evidence about how they compare is limited. All these factors mean the best DMD to start is not clear.

The majority of those interviewed who were eligible for DMDs said they chose which type to start independently. They remembered how they had leaflet-based or DVD information, were signposted to an online decision aid 'MS decisions' (on MS Trust's website) or open days, but were provided with no guidance from health professionals and were requested to make a decision about the type of DMD alone.

This imposed choice came as a shock to most people interviewed as they were used to medications being prescribed by health professionals and did not understand why they were told to make this particular choice. They talked about how they felt unprepared to make this decision by themselves and would have liked more guidance from health professionals, especially at a time when they were feeling vulnerable (perhaps soon after diagnosis or due to worsening condition).

Many policy makers, official bodies, academics and patients themselves advocate that patients should be involved in decisions about their health. However, in these cases, although interviewees had the *power to choose*, they were not *empowered to choose*, as we might expect in patient-centred care. Instead, they felt unsupported by health professionals and that this choice was imposed upon them.

We do not know the reasons why neurologists and MS nurses did not seem to provide guidance in these cases, but what is clear is that most people interviewed wanted more advice and preferred not to make the decision alone. These findings may encourage health professionals to provide more support to people with MS who are making decisions about DMDs or at least motivate health professionals to explain reasons why patients have to make these decisions alone.

[COMMENTS BOX]

How do Participants Discuss Uncertainties Related to Multiple Sclerosis?

Introduction

MS is an unpredictable and varied condition. Broadly speaking there are different types according to its course, but on an individual level its rate of progression and type of symptoms experienced are impossible to predict. MS does not have a cure and treatments available do not work for everyone or appear to slow overall progression. These factors result in a context of uncertainty for people with MS. This blog post presents how people interviewed felt about the various uncertainties they faced.

How will MS affect me in the future?

The unpredictable nature of MS meant that many of those interviewed said they felt uncertain and concerned about how they would be affected in the future. It was common to initially imagine worst-case-scenarios and assume they would be severely affected by MS. This caused fear and anxiety. People talked about how they imagined their life course would stop in its tracks due to MS. Some research suggests that chronic illness can cause 'biographical disruption' i.e. change people's imagined futures. This has been described as an unwelcome change or disturbance. However, these accounts suggest something more abrupt, i.e. an ending to a life previously imagined.

Previous research has shown something similar in the experiences of people with Motor Neurone Disease (MND); this abrupt ending to imagined futures has been put down to MND being a condition that is not only chronic, but terminal too. MS is not terminal, but people still seemed to experience something similar, when they imagined an abrupt ending. What differed with accounts of people with MS is that this point of view seemed to change. As time passed and people lived with their MS, most talked about how they regained a perception of the future closer to what they previously imagined. This period when people with MS seem to imagine worst-case scenarios and abrupt endings to their futures may be unrealistic and cause anxiety unnecessarily. The question remains as to whether we may avoid this happening with information and support during this time.

Some people said their MS fluctuated so frequently they faced uncertainties about their condition on a daily basis. This daily uncertainty meant that MS was always at the forefront of

their mind. Their plans changed accordingly, they felt concerned about whether they could fulfil plans made and some were reluctant to make plans at all. In addition to the physical difficulties of taking part in social events, a major concern was the social obligation; they worried they would "ruin it for others" or about the potential for embarrassment if they could not keep plans. Such limitations to plans can affect one's sense of self, as people with unpredictable chronic conditions no longer take part in events in the way they used to or people expect them to.

Which symptoms are caused by MS?

As MS can manifest in vast and varied ways, it is not always clear whether a problem is caused by MS or something else. There are instrumental measures such as MRI scans that record MS activity, but the way in which these directly relate to symptoms is unclear and people in the UK do not receive regular scans. Therefore, people rely on their own experiences of the condition when making sense of it. Many people talked about how they were unsure about whether various problems were caused by MS or something different. For example, one was confused as to whether visual disturbances were due to MS or just a normally aging process and another wondered whether his mood swings were caused by MS. This uncertainty about how MS manifests was felt so strongly that several people said there were occasions when they questioned whether they even had MS.

Such confusion has implications in healthcare settings too. Some of the people interviewed talked about times when they disagreed with health professionals' judgements and other times when health professionals differed in their opinions about causes. Health professionals' judgements are important as they act a gatekeepers to treatment and further investigations. If problems are misattributed to MS (or another cause), then they may be left untreated and get worse. Some participants described times when this had happened and others felt worried that this had happened. It may be important to carry out extra investigations to rule out other causes or for health professionals to have discussions with patients to explain reasons why other causes have been ruled out. This would ensure that other causes are ruled out, or help to lessen patients' concerns.

Is this treatment really working?

There are many different strategies and treatments used by people with MS in order to manage their condition, many of which are not available from mainstream medical professionals. Treatments that are available from mainstream medicine are relatively few, and their effectiveness differs between individuals and even at different stages of one person's MS. This means there are many options available, but it is unclear what works and what does not. Adding to this uncertainty, the unpredictable and fluctuating nature of MS makes it difficult, and sometimes impossible, to work out whether a treatment is working.

Many of the people interviewed talked about this confusion. They did not know whether the times when they had relatively few symptoms was due to a treatment working, or if it coincided with a period of good health. On the other hand, if their condition did not seem to improve, they thought a treatment may be working because their condition could have worsened further still without it. This uncertainty about

treatment effectiveness meant that many they often continued with treatments, sometimes suffering side effects, even when they appeared to be ineffective. People with MS might benefit from support and guidance in methods to assess and monitor treatment effects, so they can judge whether treatments are worth continuing.

[COMMENTS BOX]

Appendix F - Interview schedule

F.1. Original interview schedule

Interview Schedule – January 2013 - Abi Eccles

I am interested in the experiences of people with MS in terms their experiences of decision making for management and treatment.

Understand how decisions are made
Independently, with clinicians, or with others

I'm going to start the interview with a fairly open question and let you speak uninterrupted to begin with. I will then start asking you some follow up questions.

- I will be making notes, looking through my guide and time keeping throughout the interview, so please just try and ignore that.
- If there are any questions you would prefer not to answer or areas you don't want to talk about, that's absolutely fine, please just let me know.
- Please feel free to stop the interview at any time or take a break**.**

Can you tell me about your experiences of MS, but as you talk about this, can you also discuss how decisions have been made throughout you experiences?
When did you first notice there was a problem?
When did you seek help?
How do you manage your MS?

**Whilst the interviewee is responding to the initial question, the interviewer will be taking note of the decision points. Once the interviewee has finished the interviewer will explore each decision point further using 'section one' of the guide (below).*

Section one – Narrative

Decisions prompts:

- **Medications** (starting/stopping/changing dose)
- **Alternative therapies** (*hyperbaric oxygen therapy/acupuncture/herbal remedies/aromatherapy/reflexology)*
- **Clinical trials**
- **Caring help**
- **Adaptations and technical aids**

Can you tell me a bit more about how this decision was made?
Describe what happened.
What were your thoughts and feelings at different stages?
Who was involved in making this decision?

What was the problem that the decision was going to address?
What were your aims or priorities? / What was important to you?
What were the clinicians' aims or priorities?
How do you know?
    Did you discuss these? How did that play out?
    Were the issues important to you taken seriously?
How did you feel about the choice made?

Do you remember having any particular questions at this stage?
Did you get answers?
    How? / Why not?

What information did you use to make this decision?
Where did you access this information?
How do you assess the quality of this information?

What factors did you consider?
Are/were there any side effects?
    How did these affect you?
    Did your doctor understand how they affected you?

Can you describe the interaction during this consultation?
Who led the consultation?
Who made the decision?
How involved were you?
    Would you like to be more/less involved?
    Was your preference for involvement assessed?

Was anyone else (other than yourself and the clinician) involved in any way with this decision?
How were they involved?
What effect did that person's involvement have?
How important was it that they are involved?
How much say did they have?

Were there any misunderstandings?
Were any assumptions made?
Motives? Priorities?
Was anything confusing or unclear?
What were the consequences of these misunderstandings?

How was information exchanged between you and your [clinician]?

- **What kind of information did you provide?**

Did you volunteer knowledge and information you have learnt or discovered outside consultation?
Yes: How is this information received? No: Why not?
Were you encouraged to volunteer such information?

Did you give information about your own personal values and preferences during this exchange?
Yes: How was this received?
Did you feel confident in providing information about personal values? / Why not?
Were you encouraged to talk about personal values?

Do you think information about your own personal values was important during this exchange?
Do you think your [clinician] thought that personal values were important?

- **Were you completely honest information you shared with your clinician?**
  Did you adapt or hold back any information?
  Why?

- **What kind of information did the clinician provide?**

How did your [clinician] provide information about risks and options?
How was this information presented?
Stats? Graphs? Discussion?
Was this information clear?
What does it mean to you?
What type of information do you prefer?

- **Did the information provided by the clinician ever appear to be biased?**
  Did you trust all the information provided by the clinician?
  How do you know?
  Can you describe how you reacted?

Section two – MS Management

Can you describe ways in which you manage symptoms?

Which symptoms trouble you the most?

Why do they trouble you?
How do you manage these?

Which symptoms do you manage effectively?

How do you manage these?
How did you development these self-management skills?

Do you discuss self-management with any health professionals?

What is that like?
Can you describe the interaction?
Do you feel confident to be involved?

Do you have an explicit written overall care plan?

- YES:
- *What is it made up of?*
- *How was that created?*
- *Is it useful? Relevant?*
- *Why?*

- NO:
- *Do the terms 'action planning' or 'goal settings' mean anything to you?*
- *Are they relevant?*
- *Has anyone suggested this before?*
- *Do you think it would be useful?*
- *Why? Why not?*

Have you learnt what triggers relapses or symptoms?

How did you learn this?
Has this changed as you MS has progressed?
Have you discussed this with healthcare professionals?

Do you find that your diet affects your symptoms?

Have you changed your diet?

How did you make the decision to do this?
Where did you get information/advice about this?
Is there enough advice?

Do you have any other conditions?

How does this affect your MS? / How does your MS affect this?
How do you manage these together?
Do you discuss this with Health professionals?

Section three – General reflections and changes

Thinking back, throughout your experiences since diagnosis, in what ways has your approach to managing MS changed?
Has the style of interaction between you and healthcare professionals changed?
Has your role during consultations changed?
    Has your level of involvement changed?
    Has your preferred level of involvement changed?
Has the way information is exchanged changed?
Has your confidence and knowledge changed?

- **Can you give me any examples of these changes?**

Can you remember a decision you made, or a consultation you had, in the past which would differ if it was today?

- **How easy was it to make these changes?**

Have clinicians adapted to these?
Can you describe how they have/have not?

- **Have your preferences for types of information changed since diagnosis?**

Type? Subject areas? Amount? Format?
Has your abilities to access and assess information changed since diagnosis?

Does the interaction style differ according to the healthcare professional?
Why is that?
What are the effects of different interaction styles?

Can you remember any instances when you have disagreed with a health professional?
Can you describe what happened?
Did you discuss the disagreement?
    How was that received? Why not?

Does your [clinician] always take your issues seriously?
How do you know this?
How does that make you feel?
Has this delayed/led to help?
Are certain symptoms taken more seriously than others?

Have/would you discuss the emotional or psychological effects of MS with your clinician?
How has this played out?
Why not?

Have you ever gone to your [clinician] with a request for a specific treatment? Medication or other therapies?
Can you describe what happened?
What difficulties were you faced with? / How were you supported?

Section four – Concepts

**This section will only be explored if appropriate and if there is enough time*

There is a large body of literature around decision making and styles of interactions between patients and doctors. Three styles are commonly referred to:

1) Paternalism
2) Mutuality or Shared Decision Making
3) Consumerism

Have you heard of these concepts?
Can you describe them?
What do you think of them?
How do they relate to your experiences of decision making and interactions with Health Professionals?

What do you think about the 'shared decision making' approach?
Are there any problems?
What are the advantages?
Is it realistic?
How would it be achieved in reality?

How does this SDM approach relate to your experiences?

Have you ever heard of the term evidence based medicine?
Would you mind describing what this means?
Is it important to you?
How does it affect decision making for you?

Were you diagnosed with a certain type of MS?
What did this mean to you?
How useful was this?

What stage are you in now? How do you know?
How important is it to distinguish between types?

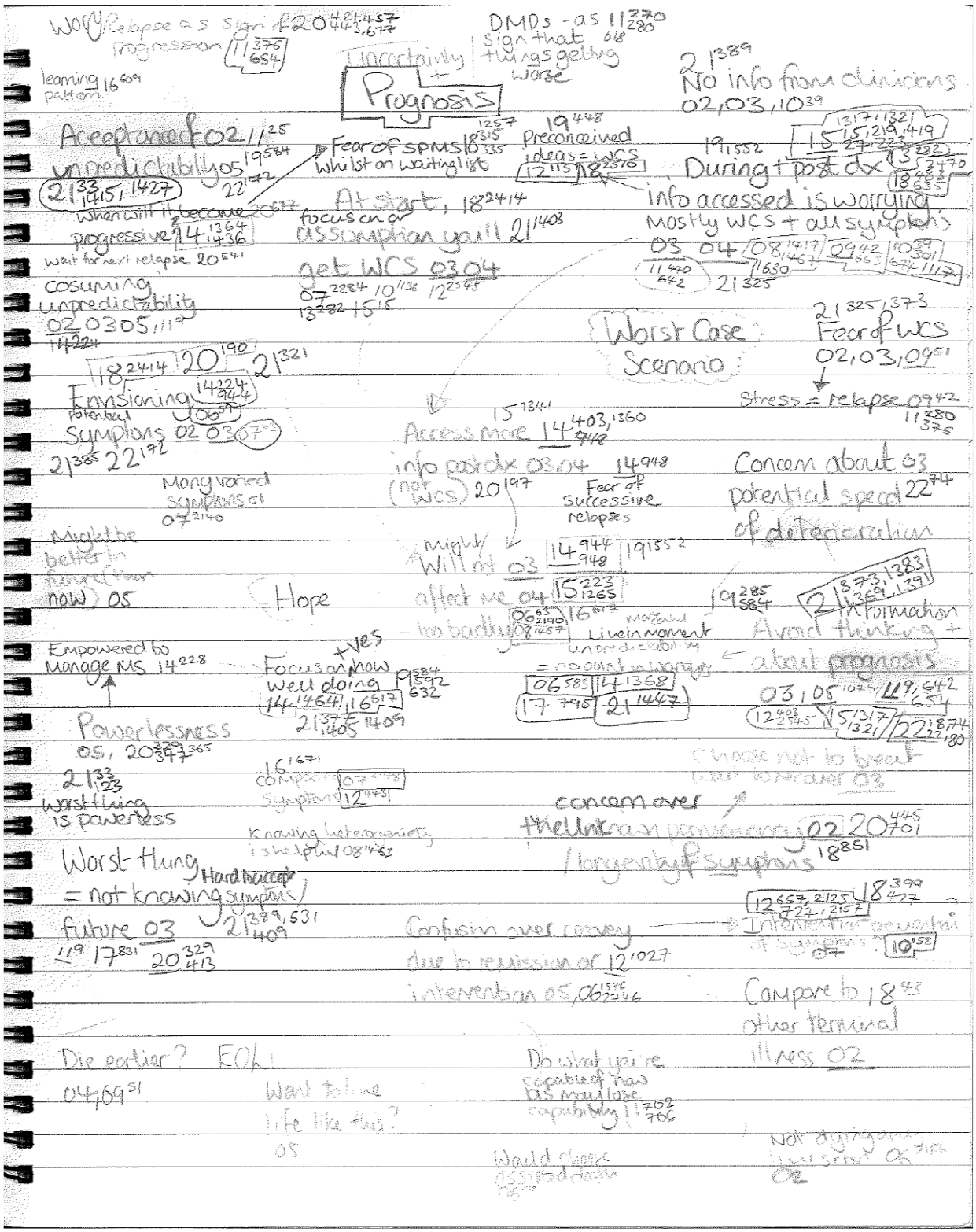
Closing Comments

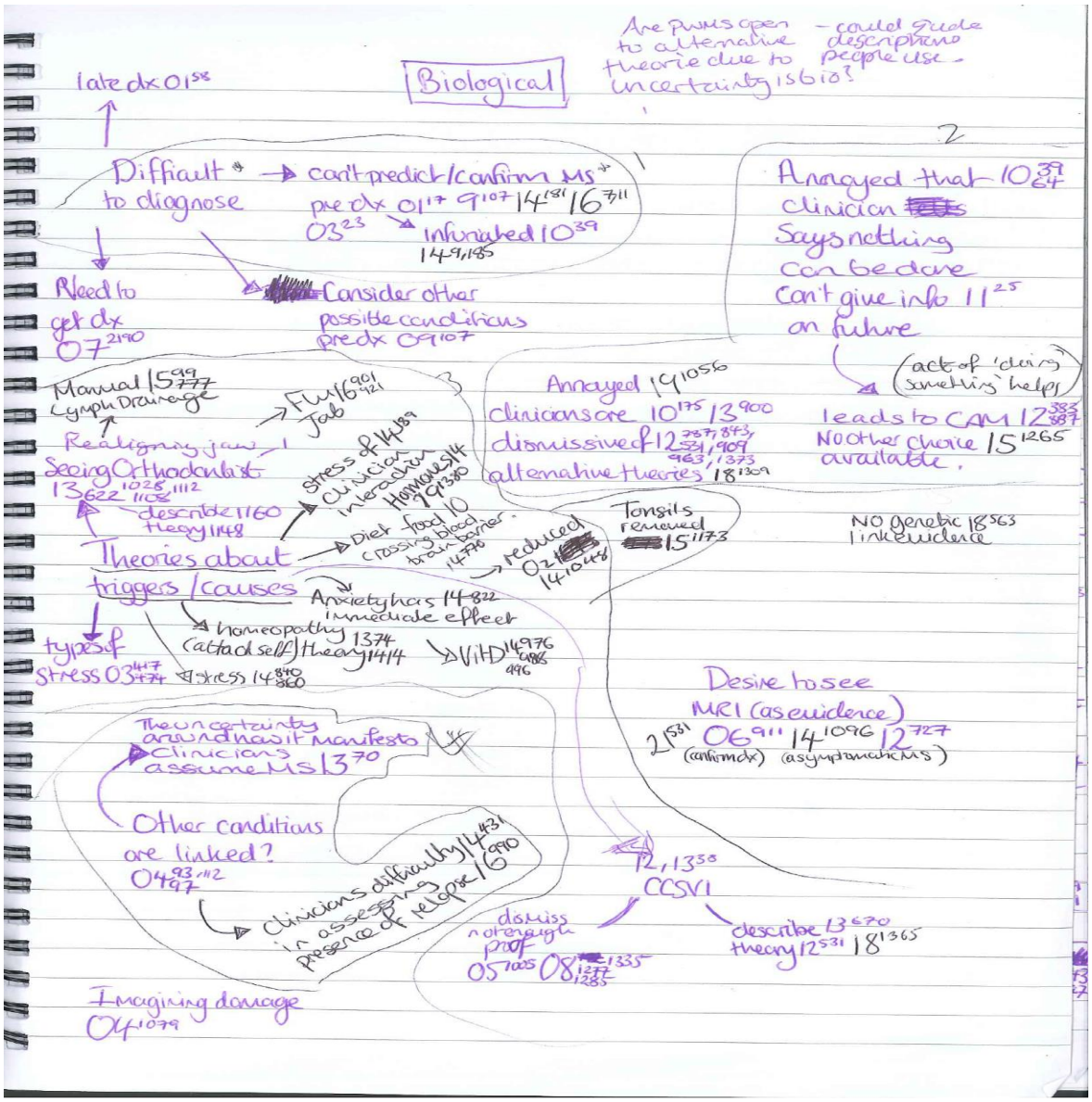
Well, I think we're coming to the end of the interview. But before we finish, is there anything you would like to discuss further, or something which we haven't covered which you would like to talk about?
Interesting experiences
Difficulties
Thoughts about decision making and information

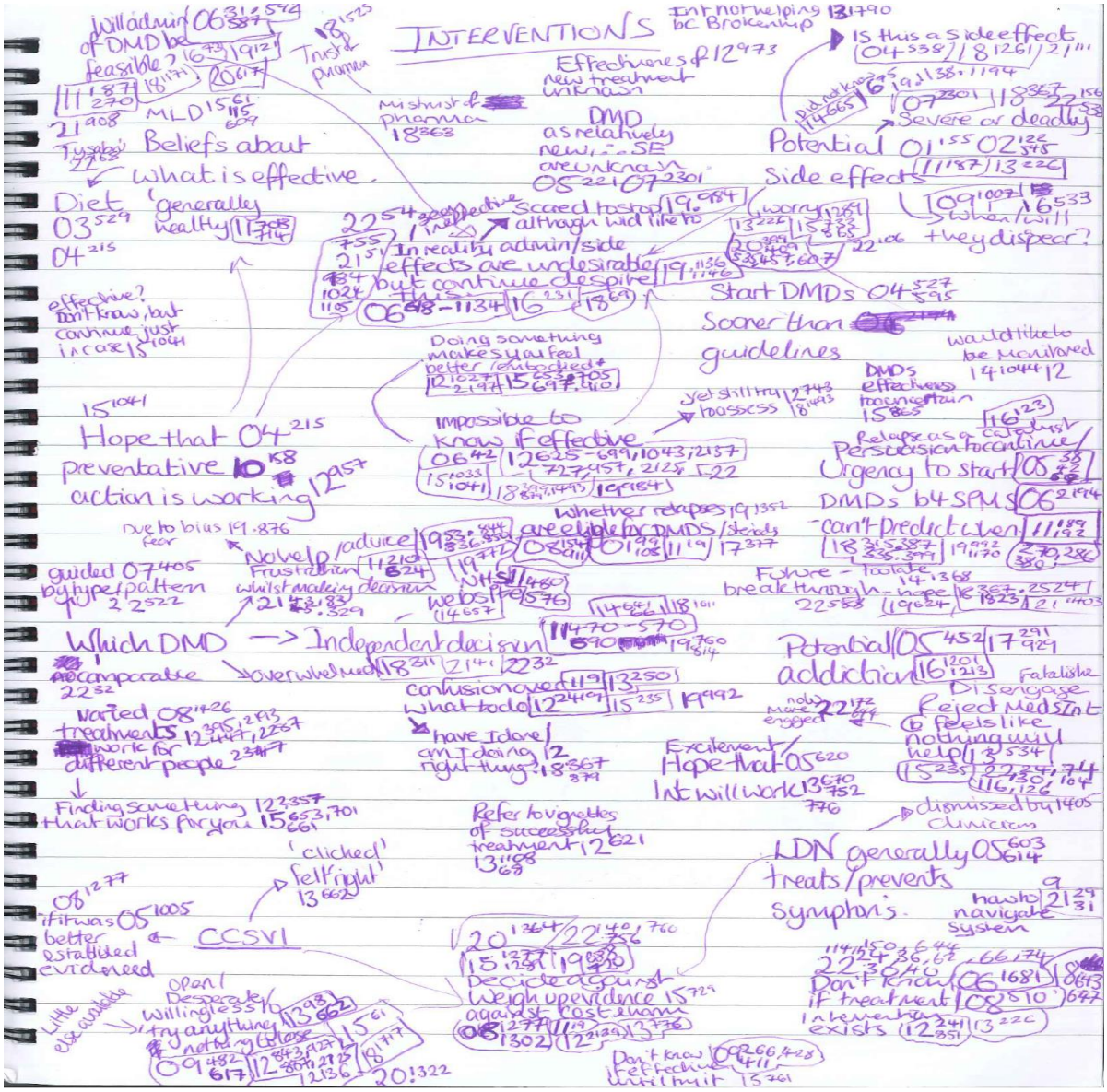
Do you have any decision making or self-management advice for others?
To someone with MS
People who know people with MS
To healthcare professionals

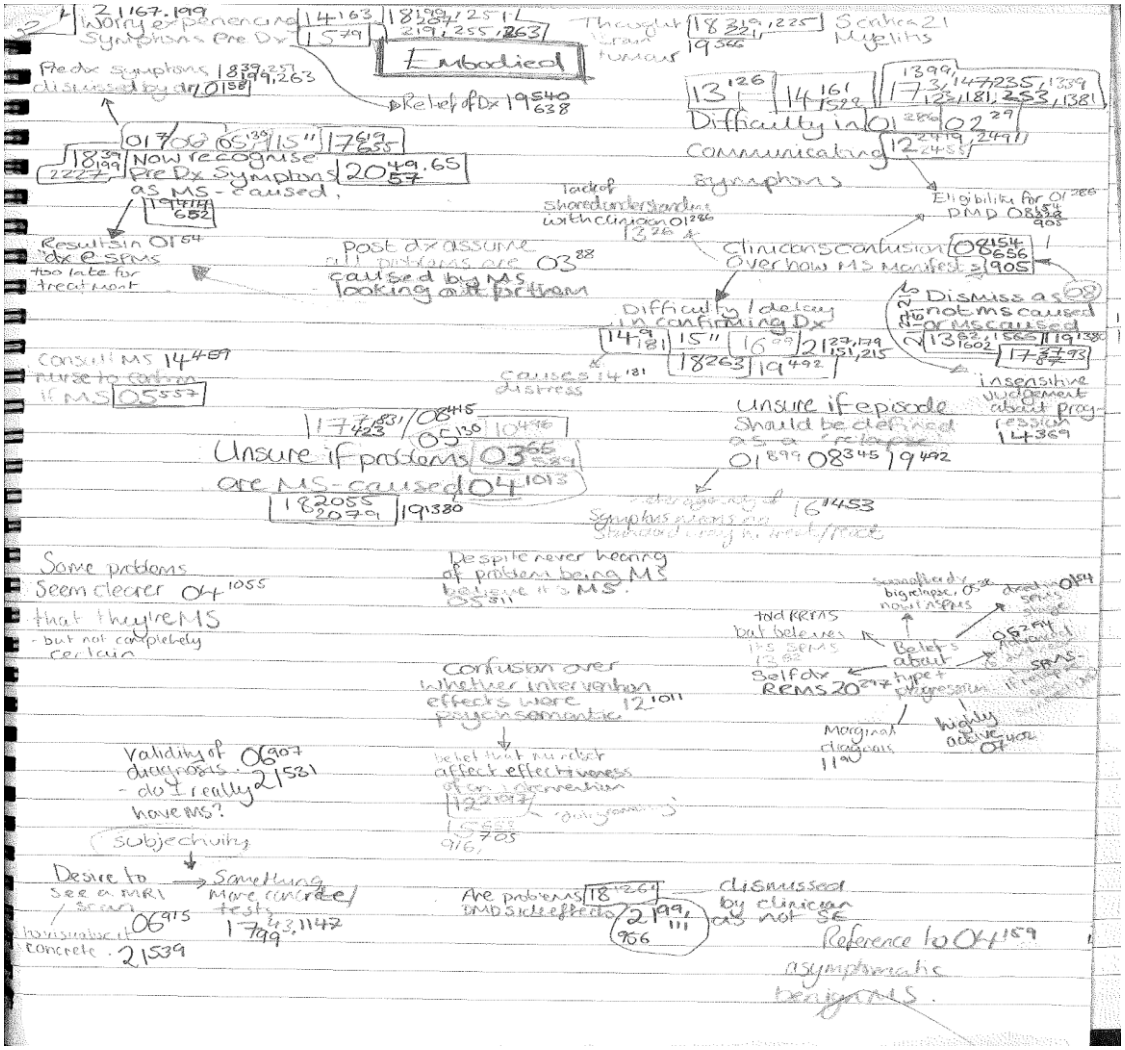
Appendix G - Examples of conceptual data maps

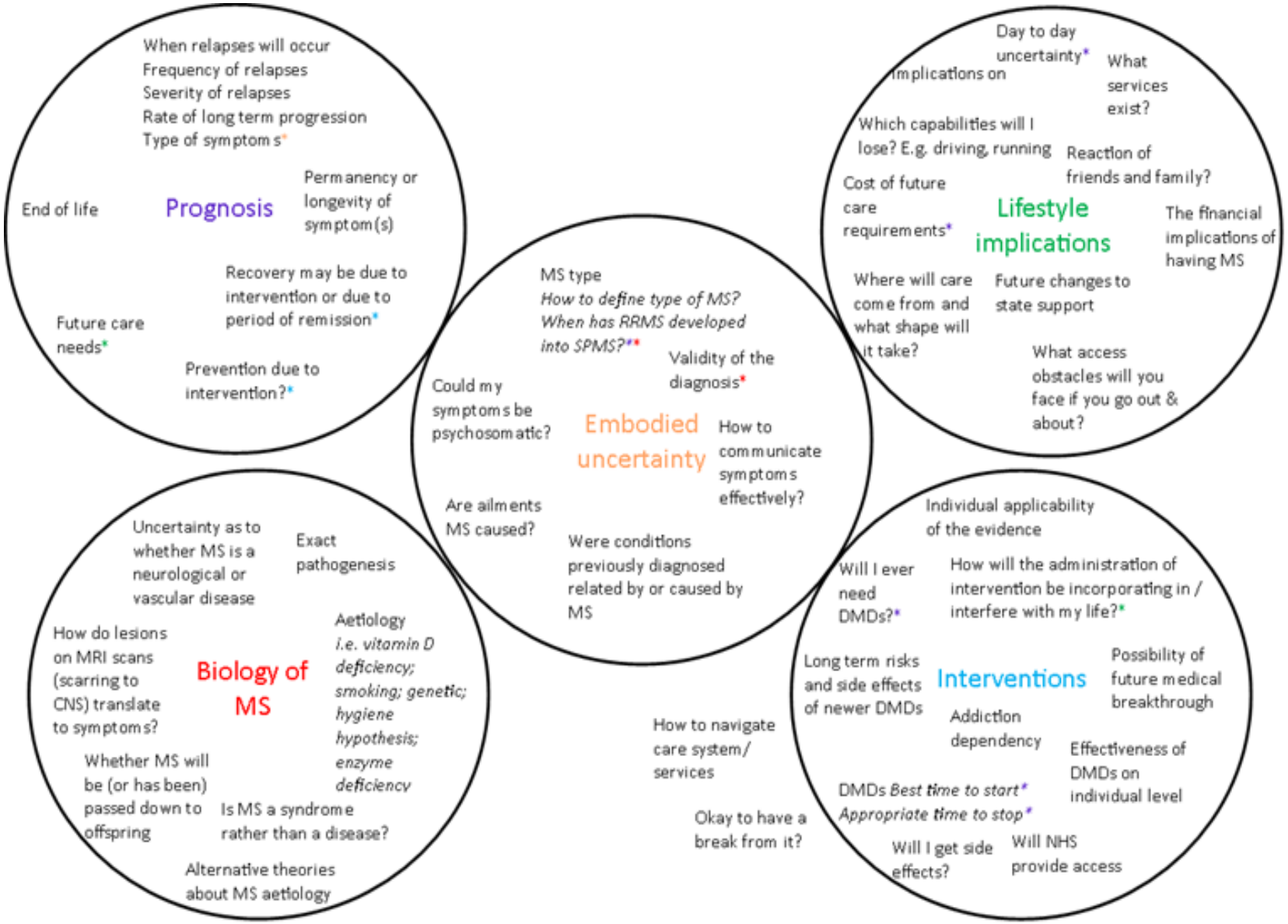
G.1. Uncertainties conceptual maps




Are PwMS open to alternative theories due to uncertainty is bio?
could guide descriptions people use.
Biological
late dx 01 58
Difficult to diagnose
can't predict/confirm MS pre dx 01 17 9 107 14 181 16 711
03 23
Infuriated 10 39 14 9,185
Need to get dx 07 2190
Consider other possible conditions pre dx 09 107
Annoyed that 10 39 64 clinician says nothing can be done
Can't give info 11 25 on future
(act of 'doing something' helps)
leads to CAM 12 385 387
No other choice 15 1265 available.
Annoyed 19 1056
clinicians are 10 175 13 900
dismissive of 12 787, 843, 531, 909, 963, 1373
alternative theories 18 1309
Manual 15 99 777 Lymph Drainage
Realigning jaw
Seeing Orthodontist 13 622 1025, 1112, 1158
describe 11 60 theory 11 48
Flu 16 901 421 Jab
stress 14 489
Clinical interaction
Homocysteine 14 1380
Diet food crossing blood brain barrier 14 776
reduced O2 14 1048
Tonsils removed 15 1173
NO genetic 18 563 link evidence
Theories about triggers/causes
Anxiety hers 14 822 immediate effect
homeopathy (attack self) theory 1374 11 14
ViTD 14 976 1486 496
types of stress 03 447 774
stress 14 840 360
The uncertainty around how it manifests
Clinicians assume MS 13 70
Other conditions are linked? 04 93, 112 97
Clinicians difficulty 14 431 990 in assessing presence of relapse 16
Imagining damage 04 1079
Desire to see MRI (as evidence) 2 581
06 911 14 1096 12 727
(confirm dx) (asymptomatic MS)
12, 1350 CCSVI
dismiss not enough proof 05 1005 08 1277 1285 1335
describe 13 670 theory 12 531 18 1365


INTERVENTIONS
Beliefs about what is effective
Diet
Generally healthy
Hope that preventative action is working
Which DMD
Independent decision
Incomparable
Varied treatments work for different people
Finding something that works for you
CCSVI
If it was better established evidenced
Like else available
Clear! Desperate/ willingness to try anything/ not trying to lose
Effectiveness of new treatment unknown
DMD as relatively new - SE are unknown
Mistrust of pharma
Int not helping bc Broke up
Is this a side effect
Potential Side effects
Severe or deadly
Scared to stop although would like to
In reality admin/side effects are undesirable but continue despite this
Start DMDs sooner than guidelines
Would like to be monitored
Doing something makes you feel better (placebo?)
Impossible to know if effective
Yet still try to assess
DMDs effectiveness uncertain
Relapse as a catalyst/ persuasion to continue
Urgency to start DMDs b4 SPMS
Can't predict when
Future - isolate, break through - hope
Whether relapses are eligible for DMDs/ steals
Due to bias, fear
Advice
Frustration whilst making decision
Guided by type/ pattern
Confusion over what to do
Have I done/ am I doing right thing
Refer to vignettes of successful treatment
Excitement
Hope that Int will work
Potential addiction
Fatalistic
Disengage
Reject Meds Int
(& feels like nothing will help)
Dismissed by clinicians
LDN generally treats/ prevents symptoms.
Has to navigate system
Decide against
Weigh up evidence against cost/harm
Don't know if effective until try it
Don't Know if treatment exists
Intervention


Embodied
Unsure if problems are MS-caused
Difficulty in communicating symptoms
Clinicians confusion over how MS Manifest
Difficulty/delay in confirming Dx
Unsure if episode should be defined as a relapse
Same problems seem clearer
Despite never hearing of problem being MS believe it's MS.
Confusion over whether intervention effects were psychosomatic
Validity of diagnosis - do I really have MS?
Desire to see a MRI scan
Are problems DMD side effects
Dismissed by clinician as not MS
Reference to asymptomatic benign MS.
Insensitive judgement about progression
Causes distress
Post dx assume all problems are caused by MS looking at bottom
Result in dx of SPMS too late for treatment
Consult MS nurse to confirm if MS
Pre dx symptoms dismissed by GP
Now recognise Pre Dx Symptoms as MS-caused
Relief of Dx
Throughout brain tumour
Scarica 21 Myelitis
Track of shared understanding with clinician
Eligibility for DMD
Dismiss as not MS caused
Belief about type + progression
Self dx RRMS
Believes has SPMS
Marginal changes
Widely active
Already big disease, doesn't matter if SPMS
Had RRMS but believes has SPMS
Worry experienced as Symptoms Pre Dx


When relapses will occur
Frequency of relapses
Severity of relapses
Rate of long term progression
Type of symptoms*
End of life
Prognosis
Permanency or longevity of symptom(s)
Recovery may be due to intervention or due to period of remission*
Future care needs*
Prevention due to intervention?*
MS type
How to define type of MS? When has RRMS developed into SPMS?**
Validity of the diagnosis*
Could my symptoms be psychosomatic?
Embodied uncertainty
How to communicate symptoms effectively?
Are ailments MS caused?
Were conditions previously diagnosed related by or caused by MS
Day to day uncertainty*
Implications on
What services exist?
Which capabilities will I lose? E.g. driving, running
Reaction of friends and family?
Cost of future care requirements*
Lifestyle implications
The financial implications of having MS
Where will care come from and what shape will it take?
Future changes to state support
What access obstacles will you face if you go out & about?
Uncertainty as to whether MS is a neurological or vascular disease
Exact pathogenesis
How do lesions on MRI scans (scarring to CNS) translate to symptoms?
Biology of MS
Aetiology i.e. vitamin D deficiency; smoking; genetic; hygiene hypothesis; enzyme deficiency
Whether MS will be (or has been) passed down to offspring
Is MS a syndrome rather than a disease?
Alternative theories about MS aetiology
How to navigate care system/ services
Okay to have a break from it?
Individual applicability of the evidence
Will I ever need DMDs?*
How will the administration of intervention be incorporating in / interfere with my life?*
Long term risks and side effects of newer DMDs
Interventions
Possibility of future medical breakthrough
Addiction dependency
Effectiveness of DMDs on individual level
DMDs Best time to start*
Appropriate time to stop*
Will I get side effects?
Will NHS provide access

Appendix H – Cochrane review

H.1. Search strategy for Cochrane review

1 MEDLINE search strategy

1. chronic*.mp.
2. ((persistent or long* term or ongoing or degenerative) adj3 (disease* or ill* or condition* or insufficienc* or disorder*)).tw.
3. long term care/
4. long* term care.tw.
5. exp cardiovascular diseases/
6. (heart disease* or heart failure or myocardial ischemia or coronary disease* or coronary artery disease* or myocardial infarction or hypertension or high blood pressure).tw.
7. sickle cell.mp.
8. exp lung diseases obstructive/
9. (obstructive lung disease* or obstructive pulmonary disease* or copd or asthma or bronchitis).tw.
10. exp emphysema/
11. exp pulmonary emphysema/
12. emphysema.tw.
13. (cystic fibrosis or respiratory distress).mp.
14. exp nervous system diseases/
15. (brain adj (disease* or damage* or injur*)).tw.
16. (cerebrovascular or brain ischemia or cerebral infarction or carotid artery disease* or stroke or epilep* or seizure*).tw.
17. (neurodegenerative or Huntington* or Parkinson* or amyotrophic lateral sclerosis or multiple sclerosis or motor neuron disease).tw.
18. (paralys* or quadriplegi* or tetraplegi* or paraplegi* or locked-in syndrome).tw.
19. ((communication or learning or consciousness or perceptual or speech or voice or vision or hearing or psychomotor) adj disorder*).tw.
20. (hearing loss or hearing aid* or deaf* or blind* or stutter*).tw.
21. down* syndrome.tw.
22. cerebral palsy.tw.
23. exp gastrointestinal diseases/
24. (gastroenter* or intestinal or bowel or colonic).tw.
25. renal insufficiency/
26. ((renal or kidney) adj (failure* or insufficienc*)).tw.
27. diabetes mellitus/
28. (diabetes or diabetic*).tw.
29. exp nutrition disorders/
30. (underweight or malnutrition or malnourished or overweight or obes*).tw.
31. exp arthritis/
32. exp rheumatic diseases/
33. (arthritis or osteoarthritis or rheumati* or fibromyalgia).tw.
34. ((back or neck) adj pain).tw.
35. exp thyroid diseases/
36. thyroid.tw.
37. exp hypersensitivity/
38. (hypersensitivit* or allerg* or intolerance or anaphyla*).mp.
39. exp neoplasms/
40. (cancer* or oncolog* or neoplasm* or carcinom* or tumo?r* or malignan* or leuk?emia).tw.
41. exp hiv infections/
42. (hiv infect* or hiv disease*).tw.
43. exp mental disorders/
44. exp behavioral symptoms/
45. ((mental* or psychiatr* or psychological*) adj (ill* or disorder* or disease* or distress* or disab* or problem* or health* or
patient* or treatment)).tw.
46. ((personality or mood or dysthymic or cognit* or anxiety or stress or eating or adjustment or reactive or somatoform or

conversion or behavior or perception or psycho* or impulse control or development* or attention deficit or hyperactivity or
conduct or motor skills or movement or tic or substance related) adj disorder*).tw.
47. (psychos#s or psychotic* or paranoi* or schizo* or neuros#s or neurotic* or delusion* or depression or depressive or bipolar or mania or manic or obsessi* or compulsi* or panic or phobic or phobia or anorexia or bulimia or neurastheni* or dissociative or autis* or Asperger* or Tourette or dyslex* or affective or borderline or narcissis* or suicid* or self injur* or self harm or adhd).tw.
48. (((substance or drug or alcohol) adj abuse) or "substance use" or "illegal drug use" or addict* or alcoholism or (problem* adj1 drinking)).tw.
49. or/1-48
50. patient care planning/
51. ((care or action or treatment) adj plan*).tw.
52. decision making/
53. choice behavior/
54. (patient* adj7 (decision* or choice*)).tw.
55. patient preference/
56. (patient* adj3 (preference* or priorit* or value*)).tw.
57. (treatment adj (option* or choice*)).tw.
58. goals/
59. (goal* adj2 (set* or plan*)).tw.
60. patient centered care/
61. (patient adj (cent*red or focus*ed or oriented)).tw.
62. individualised medicine/
63. (individuali#e? or individuali#ing or personali#e? or personali#ing or tailor or tailored or tailoring).tw.
64. or/50-63
65. cooperative behavior/
66. (patient* adj3 (participat* or involv*)).tw.
67. (negotiat* or agreement or concordan* or cooperat* or co-operat* or collaborat* or partnership).tw.
68. or/65-67
69. 64 and 68
70. patient participation/
71. (patient* adj2 (empower* or activat*)).tw.
72. ((shared or joint or informed or collaborative) adj2 decision making).tw.
73. ((involv* or participat*) adj3 (choice* or decision*)).tw.
74. (decision adj (aid* or support or tool*)).tw.
75. patient provider agreement*.tw.
76. decisional self efficacy.tw.
77. (personal budget* or direct payment*).tw.
78. (record access or patient held record*).tw.
79. ((self management or self care) adj2 support*).tw.
80. or/69-79
81. 49 and 80
82. randomized controlled trial.pt.
83. controlled clinical trial.pt.
84. randomized.ab.
85. placebo.ab.
86. clinical trials as topic.sh.
87. randomly.ab.
88. trial.ti.
89. or/82-88
90. exp animals/ not humans.sh.
91. 89 not 90
92. 81 and 91

H.2. Inclusion check-list for Cochrane review

We will use the following checklist to determine eligibility.

1. Does the paper present primary data? EXCLUDE if review article, commentary, protocol, etc. but flag for later reference scan.
2. Was this a randomised clinical trial or clustered randomised trial? EXCLUDE if not RCT, but flag for later reference scan.
3. Were participants aged over 18? EXCLUDE if participants were children or young people aged under 18.
4. Did participants have one or more chronic conditions? EXCLUDE if participants were healthy people or simulated patients.
5. Did trial include patient-based outcomes? If not, EXCLUDE, for example if outcomes related to clinicians only, or costs of intervention or care only. Trials of training programmes for clinicians that included measures of their effects on patients should be considered for inclusion in the review if training included personalised care planning.
6. Was the intervention concerned solely with planning for end-of-life care? If so, EXCLUDE.
7. Was personalised care planning with active involvement of patients in a shared decision making process an explicit component of the intervention?
    1. Were patients actively involved in planning their treatment or care with clinician(s)? INCLUDE
    2. Did the intervention include collaborative goal setting, action planning, shared decision making or decision support for patients? INCLUDE.
    3. Were patients offered a choice of treatment, support package or educational programme? INCLUDE.
    4. Were patients encouraged to set their own goals or priorities? INCLUDE.
    5. Was the care/action plan pre-prepared so patients had no opportunity to influence it? EXCLUDE
    6. Was the care/action plan simply a pre-prepared list of instructions about what to do in particular circumstances? EXCLUDE
    7. Is there any other evidence to suggest that the care planning process did not allow the patient to influence it? EXCLUDE

Appendix I – Data extraction table systematic review assessing the effectiveness of personalised care planning for people with MS

I.1. Data extraction from Block 2010

Extraction details:	
Form version/date	*Version 1.0 25/06/2015*
Review Title	*Personalised care planning for adults with Multiple Sclerosis*
Study ID	*Block 2010*
Name of trial or acronym	*Project Shake-it-up*
Other papers reporting on same study	*Block 2005, Block 2006*
Language	*English*
Name of review author completing this form	*AE*
Date form completed	*25/06/2015*
Notes *(Unpublished – for own use)*	www.projectshakeitup.org
Participants:	
Geographic location	*New york, USA*
Setting	*Community*
Start and finish date of trial	*Not stated but timeline shows it baseline measured 6mos before intervention, and measurement ended 12 months after intervention which ran: August 2002 -December 2003 (p.744). So February 2002 until December 2003*
Number (number randomised, number completing the study)	*There were a total of 35 participants in this study: 13 in the first treatment group, 13 in the second treatment group (of whom 9 also served as the wait-list control group), and 9 is the comparison (never treated) group. (taken from website)*
Age	*Ages (M = 44.2, SD = 13.3) ranged from 20-73 years. (from website)*
Sex	*22 males and 13 females*

Ethnicity	*Their ethnic groups were European-American (26), Asian- American (3), African-American and Hispanic- American (2 each), and other/missing (4) (from website)*
Principal health condition/s	*spinal-cord injury (16); multiple sclerosis (12); traumatic brain injury and stroke (2 each); and cerebral palsy, spina bifida, and aneurism (1 each) (from website)*
Other health problem/s (if relevant)	-
Stage of problem/illness (if relevant)	-
Treatment received/receiving	not stated
Literacy level	not stated
Cognitive or communication difficulties	not stated
Other social/demographic details	*their marital statuses were single (13), married (11), divorced (10), and missing (1). (from website)*
Interventions:	
Details of intervention, including theoretical basis, aim, content, format(s), media, source, setting for each arm of the study.	*In background they claim that setting achieving goals increase self efficacy which increases wellbeing. So the goal setting is part of the therapy. 'This project uses a disability studies framework of empowerment as the basis for designing the intervention' (p.742). Taking account of the social model of disability, seeks to use this understanding in order to address injustice and improve quality of life p.742. 'Project shake-it-up was designed to allow individuals to define and achieve individually indentified goals in a group setting that provides peer support, essential information and access to community resources and activities' (p.743 'participants [...] developed individually tailored project goals based on the topics that were particularly important to them, discussed strategies to for goal attainment with project staff and fellow participants, and engaged in role playing to practice self-advocacy.' (p/745) Project staff periodically monitored goal progress with participants. (No mention of changing goals or re-evaluating them (p.745)*
Stages of protocol' s model for personalised care planning cycle completed (elements A to G in Figure 1)	*A. Preparing (health info seminars). B. Goal setting C. Action planning E. Coordinating F. Supporting G. Reviewing (goal were monitored. Not sure if they were reviewed as such or changed to complete circle).*

Details of comparison intervention	no intervention
Details of co-interventions in all groups	*Na*
Delivery of intervention, e.g. stages, timing, frequency, duration, for each intervention included in the study	10 full days over 5 month period
Co-interventions included?	No
Details of providers	Researchers, working with local Centre for Independent Living a non-profit organisation providing community based recreational opportunities for people with disabilities. (From Block 2002 … Shake-a-leg Inc is the local recreation and rehabilitation programme for those with paralysis and PARL Inc. a local resource centre for independent living for those with disabilities p,186)
Duration of intervention	5 months
Intervention quality (if relevant):	Adheres to Wagner's CCM which argues that patients need support in navigating community services. It's grounded in theory. Not much information on how staff and peers were trained. But we do know they're from the 'centre for independent living' (p.742). Not sure how collaborative the goal setting is, "Case-coordinators talk with each participant individually, and ask him or her to express goals they wish to attain."p.189 2002
Follow-up period and rationale for chosen period	baseline, program completion, 6 months and 12 months (p,741)
Fidelity/integrity	*no pressure would be put on individuals to participate in activities that they were unsure of' (p.745) This is quite a vague statement and there is no explanation as to whether participants did adhere to intervention or not. In the ethnographic report, they state that lack of transportation is the most common reason for missing intervention sessions (after health issues) p.13. Missed sessions is not mentioned in the main article. On p.14 it states that personal circumstances and health problems have interfered with staff and participant participation. Suggesting that this intervention may not be practical*
Methods:	
Details of Study	
Aim of intervention	*To increase participants' self-advocacy*

Aim of study	*an intervention for capacity building and health promotion on self-efficacy and the ability to set and achieve goals, to recognize supports, and to navigate barriers to goal attainment for a group of individuals with neurological impairments'(p.743). Self-efficacy as an aspect of empowerment in that it's how people perceive their ability to overcome barriers and achieve goals. Sense of wellbeing can be attained from navigating challenges and attaining personal goals. Link between self-efficacy and QoL. 'this approach acknowledges that concepts such as empowerment, capacity building, to increase independence, and goal attainment will have multiple meanings and pathways depending upon the individuals and communities addressed [...] [To assess the effectiveness of intervention to Project Shake-It-Up was designed to allow individuals to define and achieve individually identified goals in a group setting that provides peer support, essential information , and access to community resources and activities (p. 742-3)*
Study design	*Mixed methods 'quasi experimental repeated-measures quantitative design and triangulation with qualitative data' (From semi-structured interviews, personal activity logs, independent living assessments).*
Details of setting (incl. medical setting of health service - description, age, gender, population served and clinical context of patients)	*Various community based settings, such as parks, libraries, university campuses. (p.744)*
Methods of recruitment of participants	*Announcements in organisational newsletters, mailing from state Department of health & word of mouth from local Centre for independent living and other local disability organisations.*
Inclusion/exclusion criteria for participation in study	*Inclusion: Neurological impairment, living within driving distance of centre. Exclusion: inability to speak English or give informed consent.*
Informed consent obtained?	*Yes (744)*
Ethical approval?	*Unclear*
Funding	*The study was funded by the Department of Education, National Institute on Disability and Rehabilitation Research (Field Initiated Research Award CFDA Program 84.133, PR/Award No: H133G010094), January 2002-December 2005. (taken from website)*
Statistical methods and their appropriateness	*Independent t test change in efficacy scores between groups. General linear model for difference in scores over time. Authors*

	acknowledge the small sample size is not appropriate for GLM.
Consumer involvement	*Yes, participants involved in choosing the topics for info sessions. And they developed a code of conduct for all members of the group.*
Outcomes:	
Principal and secondary outcome measures e.g. desired behaviour change?	*Primary: Self efficacy and goal setting/attainment. Secondary: Increased independence, Community access and participation. (table on p.743) Plus qualitative exploration of barriers and enables to goal attainment.*
Corresponding protocol outcome	*2-3. 2. Self-management: self-efficacy, confidence or perceived competence, ability to access relevant support 1. PAL daily living*
Methods of assessing outcome measures (outcome validated?)	*Unclear*
Methods of follow-up for non-respondents	*None*
Timing of outcome assessment	*Self-efficacy, baseline, after program, 6mos and 12 mos. Goal attainment progress 'was assessed periodically' (p,741)*
Adverse events	*(e.g. Complaints, levels of dissatisfaction, adverse incidents, side effects)*
Assessment of study quality:	
Key features of allocation	*3 groups. 1) Intervention group 2) control waiting list group 3) Control, no intervention group. Non-random assignment. Participants chose whether to receive the intervention or not. Those who chose to receive it went into either the intervention or wait list group. There were allocated with aim for equal distribution of spinal cord injury or MS. With accommodating 2 participants requests to go into wait list group. Those who chose not to receive intervention went into the comparison group (so was self-selected). Some people couldn't take part due to access. Gives example of participant that was house bound (743) (did they go into comparison group??) Also, one participant swapped from intervention group to comparison due to difficulty in accessing the intervention (p.746).*
contemporaneous data collection? (for intervention and control)	*Unsure of dates when the data for the control group were collected. It appears that the 1st intervention group and WL control were collected at same time. Then the second intervention group's data were collected a year later than the first intervention group.*

Interrupted time series - number of data points collected (before and after intervention)	*NA*

Assessment of risk of bias (used to complete the 'Risk of Bias' tables in RevMan 5)		
For details on how to complete this section, you must refer to the Cochrane Handbook, chapter 8, particularly Table 8.5.c. *NOTE: if you are including non-RCTs you may need to tailor the Risk of Bias criteria accordingly.* *For tips on how to enter data into RevMan 5, see "Risk of Bias" tables in the RevMan User Guide.*		
Adapted from Cochrane Handbook Table 8.5a: The Cochrane Collaboration's tool for assessing risk of bias		
Domain	**Review authors' judgement**	**Description**
Random sequence generation: Was the allocation sequence adequately generated?	***No***	*Participants chose whether to receive the intervention or not. Those who chose to receive it went into either the intervention or wait list group. Those who chose not to receive intervention went into the comparison group (so was self-selected). Some people couldn't take part due to access. Gives example of participant that was house bound (743) (did they go into comparison group??) Also, one participant swapped from intervention group to comparison due to difficulty in accessing the intervention (p.746). So not suprising that those in comparison group cannot access services.*
Allocation concealment: Was allocation adequately concealed?	***No***	*Participants chose which arm they wished to be in.*
Blinding: Was knowledge of the allocated intervention adequately prevented during the study?	***No/unclear***	*None*

<u>Attrition bias:</u> Were incomplete outcome data adequately addressed? *Assessments should be made for each main outcome (or class of outcomes).*	***no (evidence from other papers)***	
<u>Reporting bias:</u> Are reports of the study free of suggestion of selective outcome reporting?	***YES***	*State how the possibility of selective outcome reporting was examined by the review authors, and what was found.*
Other sources of bias. *For example (below):*		*In the ethno study they note that the consent process may have been long and patronising which 'served to discourage the participation of people form a disability group at highest risk for alcohol and substance use. P.14 'we ended up with a relatively self-selected population who reported themselves to be teetotallers.'p.14*
- Were the intervention and control groups comparable at baseline?	***No.***	*Age difference.*
- Have measures been taken within the study to protect against contamination?	***no***	*They chose which arm to go on.*
Other quality indicators (ie. potential sources of imprecision). *For example (below):*		
- Were outcome measurement tools validated?	***UNCLEAR***	
- Are the outcome measures reliable?	***UNCLEAR***	
- Did the study obtain ethics approval?	***UNCLEAR***	

Continuous outcomes

Outcome	Timing of outcome assessment (days/months)	Intervention group 1			Intervention group 2			control group			wait list Control group			Notes
		*Mean / Mean change	Standard deviation	N	*Mean / Mean change	Standard deviation	N	*Mean / Mean change	Standard deviation	N	*Mean / Mean change	Standard deviation	N	
self-efficacy	pre intervention	31.9	6.2	13	33.2	4.6	13	32.4	5.3	9	34.7	5.6	9	
self-efficacy	post intervention	33.7	3.6	13	33.4	5.5	13	29.6	5.7	9	33.1	4.7	9	
self-efficacy	6 months post intervention	33.5	5.2	13	31.9	5.1	13	29.6	9.2	9	n/a	n/a		WLC had become the second intervention group by this point.
self-efficacy	12 months post intervention	32.7	6.3	13	31.8.	4.8	13	30.3	7.8	9	n/a	n/a		

I.2. Data extraction of Eyssen 2013

Extraction details:	
Form version/date	*Version 1.2, 16/12/2013*
Review Title	*Personalised care planning for adults with MS*
Study ID	*Eyssen 2013*
Name of trial or acronym	
Other papers reporting on same study	*Eyssen 2014*
Language	*English*
Name of review author completing this form	*AE*
Date form completed	*16/11/2015*
Notes *(Unpublished – for own use)*	*Eg. References to be followed up, source of information (especially if multiple reports of same trial, or unpublished data/personal communication included).*
Participants:	
Geographic location	*Netherlands*
Setting	*outpatient clinics in hospitals and rehabilitation centres (1637)*
Start and finish date of trial	*May 2007 - March 2009 (p.529, 2014)*
Number (number randomised, number completing the study)	*CENTRES: 27 assessed for eligibility, 7 excluded, 6 refused to take part. 14 centres were stratified and randomised (7 hospitals and 7 rehabilitation centres). EXP: 6 centres (4 HP & 2 RC). CON: 7 centres (3 HP & 4 RC). 1 centre withdrew. OTS: Number of OTs not recorded. PATIENTS: n=269, exp n=156 (51/207, 25% excluded), con n=113 (63/176, 36% excluded). Lost to follow up at 4 mos: exp = 6 , con = 4, at 8mos: exp= 2, con=2*
Age	*45.5 (10.9), exp 44.4 (11.4), control 47.1 (9.9)*
Sex	*Female 68% Male 32%, exp: 71% F, con 66% F.*
Ethnicity	*Raised in Netherlands 94%*
Principal health condition/s	*MS*
Other health problem/s (if relevant)	*Absence of depression that would interfere with therapy (assessed by OTs)*
Stage of problem/illness (if relevant)	
Treatment received/receiving	occupational therapy
Literacy level	Participants were fluent in Dutch
Cognitive or communication difficulties	
Other social/demographic details	*(eg. Literacy or reading level)*
Interventions:	

Details of intervention, including theoretical basis, aim, content, format(s), media, source, setting for each arm of the study.	*Intervention based on 'Canadian practice process framework', eight stages: 1. Enter/initiate 'collaborative decision is made to either engage in or terminate the therapy' 2. Set the stage: OT and participant determine how they will work together. They clarify expectations and assumptions and identify priority issues and possible goals. 3. Assessment of evaluation of the personal, environmental and occupational factors underlying participants' issues. 4. Agree on objectives and plan. OT and participant establish goals and agree on objectives and plan of intervention. 5. Implement the plan with power sharing and participant involvement. 6. Monitoring and modifying with ongoing evaluation to assess in strategies are meeting objectives, 7. Examination of the plan and goal completion to decide in they are met or if new goals or plans are needed. 8. Conclude: a collaborative decision as to whether they should pursue other objectives or conclude the relationship. P.1637 OTs in experimental group have 2 days' training on client centred practice based on framework (above), followed by 5 booster sessions during the course of the trial. Training comprised of theory, role-playing and DVDs with examples. p.1638 OTs also had access to client-centred process model and kept a diary with criteria based on the CPPF (above) (p.528 (2014)*
Stages of protocol' s model for personalised care planning cycle completed (elements A to G in Figure 1)	*Difficult to assess which parts of the protocol's model were incorporated in the performance of the intervention. OTs received training on the CPPF which incorporates stages A, B, C, E, F and G and possibly D, however they do not refer to a systematic way of documenting action plan and goals set. It is unclear whether all the stages were performed. In accompanying paper Eyssen 2014 they describe how a much longer time was spent on the diagnostic processes, rather than therapy 'The time spent on these action points depends on the client's and therapist's situation and therapy process' (p. 528 2014) Secondary analysis demonstrated that experimental group took 11.4 days longer to formulate their first goal, compared with control (p.529, 2014)*
Details of comparison intervention	Control group carried out OT as usual with goal setting and providing therapy p.1638 OTs did not have access to the client-centred framework and tools (p.528 (2014)
Details of co-interventions in all groups	*(co-interventions may be separate to the intervention of interest for this review, or they may be other similar elements in a suite of interventions having a common purpose. Record all relevant information).*

Delivery of intervention, e.g. stages, timing, frequency, duration, for each intervention included in the study	Care received from OTs who have attended 2 day course followed by booster sessions during data collection period (unclear when).
Co-interventions included?	
Details of providers	OTs in experimental group were trained two trainers 1) professional from 'Dutch Association for Occupational Therapy' who had experience of training OTs in a client-centred way and 2) coordinating researcher who had experience in client-centred practice and using implementation strategies. p.1638
Duration of intervention	2 days' training followed by 5 booster sessions during the course of the trial, unclear when.
Intervention quality (if relevant):	Training appeared to cover all stages of the PCP model, however it is difficult to assess which parts of the protocol's model were incorporated in the performance of the intervention. Only broad details of the training provided.
Follow-up period and rationale for chosen period	4 months, 8 months, no rationale provided
Fidelity/integrity	*Coordinating researcher ensured OTs were carrying out therapy as agreed by providing support through visits and answering questions via emails, phone calls and post. (p.1639). ECGP assessment of adherence to the CCPF approach was assessed. P.1640 OTs in experimental performed better in client centre, therapeutic, relationship and organisational aspects (table 3)*
Methods:	
Details of Study	
Aim of intervention	*To carry out client centred therapy based on a structured client-centred process framework (CCPF) p.1637*
Aim of study	*[…] to assess the efficacy of client-centred OT as compared to usual care OT in patients with MS' (1637)*
Study design	*Multi-centre cluster randomised controlled trial. Institution as the unit of randomisation*
Details of setting (incl. medical setting of health service - description, age, gender, population served and clinical context of patients)	*Outpatient Hospitals (7) and rehabilitation centres (6). Both had 2 OTs and 10 new MS patients each year*

Methods of recruitment of participants	*Participants were recruited post randomisation. It is unclear how participants were recruited, but appears that they aimed to select consecutive patients, because in limitations they state, 'We cannot completely eliminate the possibility that some physicians or OTs did not strictly select all consecutive eligible patients' p.1644*
Inclusion/exclusion criteria for participation in study	*Adults with MS, did not include those without consent. Eligible centres must have at least 2 OTs regularly providing care for people with MS in outpatient setting and have at least 10 new MS patients each year p. 1637. Patients had to have at least one limitation in daily life activity. Those with a single health question, or depression that interferes with OT (according to physician) were excluded (p.1637) (although it is unclear how this was assessed). Participants were fluent in Dutch. Newly referred and had not received OT in previous 6 months.*
Informed consent obtained?	*Yes, as they did not include those who did not consent*
Ethical approval?	*Yes, Medical Ethics Committee of the VU University Medical Centre in Amsterdam*
Funding	*The Stichting MS research, the Netherlands, provided finanicial support. P.1645*
Statistical methods and their appropriateness	*ITT and PP analyses*
Consumer involvement	*None recorded*
Outcomes:	
Principal and secondary outcome measures e.g. desired behaviour change?	*Primary: disability, participation and autonomy. Secondary: fatigue, generic health-related QoL, quality and evaluation of therapy and therapy compliance and therapy frequency.*
Corresponding protocol outcome	*1. (Self-reported) disability, fatigue, generic health-related QoL. 1. iii Psychosocial health: Participation and autonomy. (Assessment of the process is important but not covered in outcomes). 1. upper extremity function, effects of pain on everyday life, cognitive functioning 2. measured in self-care*

Methods of assessing outcome measures (outcome validated?)	*Disability was measured by the disability and impact profile (DIP) which is 'a 39-item self-administered questionnaire regarding activities that may be restricted by a disabling disease. Two questions about fatigue were added to the DIP.' (DIP domains include symptoms, mobility, self-care, social activities, communication and psychological status). p.1639 Impact on participation and autonomy (IPA) assesses impact of condition on perceived participation and autonomy of outdoor and indoor autonomy, family role, work and social relations. Upper extremity function: nine hole peg test. Fatigue: the modified fatigue impact scale. Pain and disturbing sensations effect on everyday life: The pain effects scale. Cognitive functioning: Perceived Decifits Questionnaire. Health related quality of life: Short-Form Health Survey Questionnaire (SF36). Patient's perception of therapeutic outcome: Canadian Occupational Performance Measure. Process outcomes 'the evaluation of client-centred process (ECGP) and the quality of care through the patient's eyes (Quote -EEE). 'The ECGP is a translation of the Canadian client-centred process evaluation' p.1639*
Methods of follow-up for non-respondents	*ITT analysis*
Timing of outcome assessment	*Baseline (pre-treatment), four months (post-treatment) and 8 months (follow up)*
Adverse events	*None recorded*
Assessment of study quality:	
Key features of allocation	*Cluster randomised trial of rehab centres and hospitals. Random allocation before recruitment of patients. Allocation was concealed from patients and outcome assessors.*
Contemporaneous data collection? (for intervention and control)	*yes in homes*
Interrupted time series - number of datapoints collected (before and after intervention)	*3 datapoints. Baseline, 4 months and 8 months*

Assessment of risk of bias (used to complete the 'Risk of Bias' tables in RevMan 5)

For details on how to complete this section, you must refer to the Cochrane Handbook, chapter 8, particularly Table 8.5.c.

NOTE: if you are including non-RCTs you may need to tailor the Risk of Bias criteria accordingly.

For tips on how to enter data into RevMan 5, see "Risk of Bias" tables in the RevMan User Guide.

Adapted from Cochrane Handbook Table 8.5a: The Cochrane Collaboration's tool for assessing risk of bias

Domain	Review authors' judgement	Description
Random sequence generation: Was the allocation sequence adequately generated?	***Yes***	*Stratified for type of institution. Randomisation procedure based on random number tables and performed using computer generated sequence (with allocation concealment). P.1637*
Allocation concealment: Was allocation adequately concealed?	***Yes***	
Blinding: Was knowledge of the allocated intervention adequately prevented during the study?	***Yes***	*1) The nature of the intervention meant that OTs could not be blinded. But they were instructed not to inform patients about the allocated intervention. 2) the patients and the test assistants who collected the data were blinded.*
Attrition bias: Were incomplete outcome data adequately addressed?	***Yes***	*ITT*
Reporting bias: Are reports of the study free of suggestion of selective outcome reporting?	***No***	*State how the possibility of selective outcome reporting was examined by the review authors, and what was found.*
Other sources of bias. *For example (below):*		*State any important concerns about bias not addressed in the other domains in the tool. If particular questions/entries were pre-specified in the review's protocol, responses should be provided for each question/entry.*

- Were the intervention and control groups comparable at baseline?	***No***	Significant differences between exp and con grps in age (44 vs 47yrs, p=0.05), institution (73% vs 36% hospitals, p=0.00), academic centre (73% vs 17% academic centre, p=0.00), MS type (39% vs 21% RR, 16% vs 14% PP, 12% vs 26% SP, 6% vs 4% PR, p=0.00). Significant differences in basline secondary outcomes: PES total scale (p=0.04, the SF36 bodily pain scale (p=0.01 and the SF36 physical component scale (p=0.05) . P.1640
- Have measures been taken within the study to protect against contamination?	***Yes***	*Cluster method of randomisation ensured trained OT's and patients were separate*
See Cochrane Handbook 8.14.1.6 Further examples of potential biases	***Yes***	In terms of PCP, the exp and the con arms may be too similar. Usual care OT incorporates goal setting and is described as client centred. Study made efforts to demonstrate differences between groups by exp grp completing questions about their adherence and dis-adherence to the CPPF (theoretical basis of the intervention). Both groups also carried out ECGP questionnaire before and after data collection to assess client-centeredness. My understanding is that ECGP is derived from CPPF which the intervention is based on P.1639 ABI: this method of assessment assesses whether the specific intervention worked, and may have overlooked other ways that the interactions may have been client-centred.
Other quality indicators (i.e. potential sources of imprecision). *For example (below):*		*Note: A potential source of bias must be able to change the magnitude of the effect estimate, whereas sources of imprecision affect only the uncertainty in the estimate (i.e. its confidence interval). Potential factors affecting precision of an estimate include technological variability (e.g. measurement error), and observer variability.*
- Were outcome measurement tools validated?	***Yes***	
- Are the outcome measures reliable?	***Yes***	
- Did the study obtain ethics approval?	***Yes***	**Medical Ethics Committee of the VU University Medical Centre in Amsterdam**

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