Evaluation of a self-help intervention for improving well-being and reducing depression in a sample of people with MS

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Executive Summary

Background

Multiple Sclerosis. Multiple Sclerosis (MS) is an incurable progressive disease that develops when the immune system mistakenly attacks the central nervous system. The damage results in wide-ranging symptoms of physical disability and neuropsychiatric symptoms. MS effects one in 1000 people, making it the most common neurological disease in young adults. The progressive and unpredictable course of MS and threat to personal autonomy from fatigue, disability, and functional loss can harm the well-being of those diagnosed. People with MS have lower levels of well-being and higher rates of mental health problems than the general population, making both the treatment of mental health problems and the enhancement of well-being important areas in the care of people with MS.

Enhancing Well-being. Well-being is a multidimensional construct that encompasses various psychological, emotional and social dimensions. Definitions of well-being typically include the presence of high positive affect, low negative affect, high life satisfaction, and the actualisation of human experience and potentials, such as relationships or achievements. Interventions that aim to cultivate well-being, as opposed to treat illness, are known as Positive Psychology Interventions (PPIs). PPIs use techniques designed to enhance key processes such as positive emotions, competence, optimism, self-acceptance and positive relations and have been found to effective. Benefits of the PPI approach are that it may be applied widely in the absence of mental health diagnoses, and that having high levels of well-being is a protective factor against developing future mental health problems. Although reducing negative symptoms is not the focus of PPIs,

research shows that individuals have a significant reduction in depressive and anxiety symptoms after treatment.

Treating mental health problems. Psychological interventions have a large evidence base for treating common mental health problems such as depression and anxiety and can be more effective at preventing relapse than pharmacotherapy. There is evidence that psychological interventions are effective for people with MS, and may be of particular value due to effective medication being poorly tolerated in many people with MS.

Self-Help Interventions. Whilst there is a large evidence base for psychological interventions, there is a limited number of available psychological therapists, and self-help interventions (SHIs) have the potential to fill the shortfall by providing evidence-based therapies with minimal therapist contact time. SHIs may be particularly relevant for people with MS because they foster a sense of autonomy and can be accessed from home, making them accessible for those with limited mobility. Whilst a large evidence base exists for SHIs in different populations, research suggests that the effectiveness of SHIs differs significantly between populations with different health conditions, emphasising the need for their evaluation in people with MS.

Systematic Review

Aim. A systematic review was conducted on 9th December 2018 to evaluate how effective psychological SHIs are for people with MS in reducing distress and improving well-being.

Method. Searches of the electronic databases PubMed and PsycINFO were conducted using search terms related to MS, well-being, mental health,

mental illness and self-help. To include breadth of results, no search terms related to ‘intervention’ were used. Studies were included if they were controlled trials (with or without randomisation) that reported quantitative psychological outcome data following a psychological SHI in people with MS. No exclusion criteria were applied.

Results. The search yielded 165 results, and 7 were eligible for the review. All 7 were RCTs and all SHIs were web-based. Six used a wait-list control, and one an active control, and three included a long-term follow-up (3, 4, and 6 months). Two studies evaluated versions of the same intervention. Three were pure self-help and four were guided self-help using email (n=2), telephone (n=1), and Skype (n=1) support.

The primary focus of SHIs was fatigue (n=3), depression (n=3), and physical activity (n=1), and all of them were primarily based on Cognitive Behavioural Therapy (CBT) principles. In addition, one included PPI strategies, several included mindfulness, and one drew from social cognitive theory.

SHIs designed to treat fatigue all had significant improvement in fatigue relative to controls with medium to large effect sizes and improved QOL, maintained at 3m follow-up, but not necessarily improved anxiety or depression. Of those designed to treat depression, only one SHI reported significant improvement in depression relative to controls, with a medium effect size, which also translated to improved QOL. Improvement in depression was maintained at 6-month follow-up. One did not report statistics due to a small sample, and one had significant improvement in both groups meaning between-group differences were non-significant. The SHI aimed at increasing physical activity saw significant increase in physical activity,

reduction in fatigue and perceived disability, and a trend towards improved anxiety relative to controls.

Conclusion. The systematic review revealed that there is a paucity of studies evaluating SHIs in individuals with MS, and none designed to improve well-being, anxiety, or cognitive impairment. There was heterogeneity within the intervention content and format, and levels of guidance offered. A trend towards higher effect sizes for greater guidance was observed but the number of studies was too small to make any firm conclusions. The promising results found in SHIs for fatigue supports the ongoing development and evaluation of SHIs for people with MS. Further research is required to determine the active ingredients of successful SHIs for people with MS.

Empirical Study

Aim. To evaluate the effectiveness of a pure self-help PPI for increasing well-being and reducing depression for people with MS.

Background of intervention. The Goals and Planning (GAP) intervention is designed to increase well-being by helping individuals identify and work towards positive life goals through teaching goal-setting and planning skills. The techniques taught in GAP relate directly to the knowledge derived from the empirical literature that show the pursuit of chosen goals can increase well-being through providing structure, meaning, and sense of purpose, facilitating positive future thinking, and providing daily experiences of autonomy, competence, and relatedness. Certain skills have been shown to strengthen the positive relationship between goal striving and well-being, and these form the basis of GAP's six modules: Choosing self-concordant goals, imagining achieving those goals, having small, specific and

realistic planned steps towards goals, being able to adapt goal plans, and being able to adapt or abandon unattainable goals. In a self-help format GAP has been shown to increase well-being and reduce depression in community and depressed samples.

This intervention may be beneficial for people with MS for several reasons. For example, people with MS face challenges concerning life goals due to unpredictable disease progression and exacerbation, and goal adjustment capacities are thought to be crucial for coping and adjustment among individuals with stressful life events such as ill health.

Method. An RCT was conducted in which a sample of 58 participants with MS were randomly allocated to receive GAP (n=29) or to a WLC group (n=29). Measures of well-being and depression were taken at three time points; baseline, post-intervention, and at eight-week follow-up. After the follow-up measures, the WLC group were given access to GAP. Changes over time in well-being and depression were compared between the two groups. A 2 (treatment vs control) x 3 (Time 1, Time 2, Time 3) repeated measures ANOVA was employed to look at differences between the groups.

Recruitment. Participants were recruited through asking MS Centres and support groups to circulate study adverts to their members, and three MS organisations sharing it on social media. 122 centres and groups were contacted, and 57 circulated the advert. Inclusion criteria were a diagnosis of MS, fluency in English, and consent to inform GP of participation. No exclusion criteria were applied.

Measures. Sociodemographic information was collected at baseline only, and the following measures at all three points:

Positive and Negative Affect Scale Short Form (I-PANAS-SF) yields two sub-scales of positive affect and negative affect.

The Flourishing Scale (FS) measures the individual's self-perceived success in areas such as relationships, self-esteem, purpose, meaning and optimism.

The Functional Assessment of Multiple Sclerosis (FAMS) is a QOL instrument developed for use with people with MS covering the domains of mobility, symptoms, emotional well-being, general contentment, thinking/fatigue, and family/social well-being.

Patient Health Questionnaire (PHQ9) measures the severity of depressive symptoms.

Results.

Missing Data. 16 (27.6%) participants had missing data from one or more time point, and they were significantly more likely to be in the intervention group rather than WLC (χ^2 (1, N=58) = 8.63, p = .007). Multiple Imputation was used to manage missing data.

Sample. Participants were mainly White (97%), female (74%), middle-aged (mean = 50 years, SD=11.9), with relapsing-remitting MS (53%), and had been diagnosed for 13 years (SD=9.5). The intervention group had had MS for significantly longer than those in the control group (mean difference = 5.2 years, t(55) = 2.13, p = .033).

Treatment effects. The intention to treat (ITT) primary analysis yielded a non-significant result for group x time interactions on all outcomes. There was a significant decrease in depression (F(2,112) = 0.97, p = .005, η_p^2 = 0.02) and negative affect (F(2,112) = 4.34, p = .018, η_p^2= 0.07) for the whole sample over

time. A modified ITT analysis (n= 41) revealed a significant group x time interaction effect for the Family and Social well-being FAMS subscale only, with a large effect size ($F(2, 40) = 4.65$, $p = .012$, $\eta_p^2 = .12$).

Discussion.

Potential explanations for the unexpected significant reduction in depression and NA for the whole sample is a spontaneous recovery of a highly motivated subsample of patients, and anticipation of receiving the intervention at the end of the waiting period creating hope and reduction in symptoms in the WLC.

It was unusual that a significant increase in the Family/Social subscale of the FAMS found in the sensitivity analysis occurred in isolation. Possible explanations are that it is a chance finding from multiple testing or from case-wise deletion creating unequal groups, or that the study lacked power to detect changes in other domains.

Potential explanations for a lack of treatment effect include MS-related variables. For example, cognitive symptoms and fatigue could have reduced the ability for participants to engage fully with the modules, and the additional challenges posed by MS in the pursuit and adaptation of goals could mean they require additional guidance in the process, for example in a guided SHI.

Conclusion. There is no evidence that GAP in a self-help format improves depression for people with MS. It may improve individual's well-being in the social domain for those that adhere to the program. Further research is needed before recommendations could be made to use GAP in the population of people with MS.

Integration, Impact and Dissemination

Integration. The paucity of studies found in the systematic review confirmed the belief that SHIs for people with MS is an area that requires further research and gives value to the empirical study, that appeared to be the first of its kind. The limitations found in the studies in the review helped to inform the empirical study. For example, using a QOL measure that included both positive and negative factors of QOL, using two or more measures to capture different dimensions of well-being, reporting effect sizes for all possible results, and using an ITT analysis.

Difficulties recruiting to the study were evident due to a lower than expected response rate, despite the advert reaching a large demographic, and the study had low power as a result. Rhul ethical approval only allowed for non-clinical site recruitment; using a pre-post RCT design with no follow-up would have relieved time pressure and allowed for NHS ethical approval and new recruitment from clinical settings to achieve a larger sample size.

Impact. The findings overall of this thesis have highlighted a significant need for SHIs, especially those focussing on anxiety, cognitive symptoms, and increasing well-being, to be evaluated in people with MS and the importance of even well-established interventions to be evaluated in this population before being recommended. Understanding the factors associated with effectiveness, so that they can be incorporated into the design of future SHIs, is paramount.

Dissemination. A plain English summary of the overall research findings will be provided to study participants and staff members of all participating MS Centres and support groups, who can become sources of continued dissemination. Reaching a broader audience will be sought through submission to established MS and well-being journals.

Systematic Review: How effective are self-help interventions at improving well-being or reducing distress for people with multiple sclerosis?

Abstract

Multiple sclerosis (MS) is a progressive neurological disease that damages the central nervous system, resulting in wide ranging symptoms and disability that can negatively impact on the mental health and well-being of those diagnosed. Many effective psychological interventions have been transformed into a self-help format (SHIs) which may be particularly relevant for people with MS because they foster a sense of autonomy and are accessible for those with limited mobility, but this is yet to be systematically reviewed. This systematic review aimed to determine how effective psychological SHIs are for people with MS in reducing distress and improving well-being.

Searches of the electronic databases PubMed and PsycINFO were conducted using search terms related to MS, well-being, mental health, and self-help. The search yielded 165 results, and 7 were eligible for the review. All 7 were RCTs and all SHIs were web-based. The primary focus of SHIs was fatigue (n=3), depression (n=3), and physical activity (n=1). SHIs designed to treat fatigue all had positive significant results with medium to large effect sizes and improved QOL, but not necessarily improved anxiety or depression. Only 1 SHI designed to treat depression reported significant improvement relative to controls, with a medium effect size, which translated to improved QOL. The SHI aimed at increasing physical activity saw significant increase in physical activity, reduction in fatigue and perceived disability, and a trend towards improved depression and anxiety.

There is a paucity of studies evaluating SHIs in individuals with MS, and none designed to improve well-being, anxiety, or cognitive impairment. The promising results found in SHIs for fatigue supports the ongoing development and evaluation of SHIs for people with MS. Further research is required to determine the active ingredients of successful SHIs for people with MS.

Background

Multiple Sclerosis

Multiple Sclerosis (MS) is a chronic inflammatory disease of the central nervous system, which is characterised by focal plaques of demyelination and axonal loss in the brain and spinal cord. In addition to focal plaques, there is global neurodegeneration in the brain and spinal cord (Lassmann, 2018), and the changes gradually result in brain tissue loss and atrophy (Mahad, Trapp, & Lassmann, 2015).

It is estimated that more than two million people worldwide have MS, making it the among the most common causes of neurological disability in young adults (World Health Organisation, 2008). Most people diagnosed with MS start with a relapsing-remitting course (RRMS), which, after several years, develops into a secondary progressive phase (SPMS), which progresses without periods of remission. Some people are diagnosed with a primary progressive form of MS (PPMS) and experience uninterrupted disease progression from the onset, missing the relapsing-remitting phase (Lublin & Reingold, 1996).The ultimate cause of MS remains unknown (Mahad et al., 2015).

The neurological damage results in wide-ranging symptoms of physical disability, including sensory and motor loss, and neuropsychiatric symptoms including anxiety, depression and cognitive impairment. Unfortunately, the range of symptoms experienced by people with MS impacts significantly on tasks of daily living and quality of life (QOL), over and above those with other chronic conditions (Mitchell, Benito-León, González, & Rivera-Navarro, 2005).

As well as MS causing neuropsychiatric symptoms such as anxiety and depression through neural damage, the progressive and unpredictable course of MS and threat to personal autonomy from physical disability, can have a particularly negative impact on the mental health and well-being of those diagnosed (McCabe & McKern, 2002; Mullins et al., 2001). Equally, research has repeatedly shown that the presence of anxiety and depression within people with MS is associated with poorer health outcomes (Mohr & Cox, 2001), creating a cyclical pattern. Thus, the relationship between mental health problems and MS is multi-factorial and complex.

Presently, MS is an incurable condition, and although disease-modifying drugs exist, they are only beneficial for those with RRMS and are only moderately effective (Wiendl & Hohlfeld, 2009). The lack of medical interventions available means that non-pharmalogical interventions are especially crucial for improving the mental health and well-being of those diagnosed. Research has shown that over 50% of people with MS report that they have unmet non-pharmalogical needs relating to MS (Lonergan et al., 2015).

Mental Health Problems

Research has consistently shown that people with MS have higher rates of mental health problems than the general population, with a recent study reporting 35% of people with MS having diagnosed anxiety and 31% diagnosed with depression (Marrie et al., 2013). As well as being associated with poorer health outcomes for people with MS, mental health problems severely affect QOL. Interestingly, the negative association between mental health problems and QOL is stronger than that of disease-related disability and QOL for people with MS (Kern et al., 2009),

making them vital areas for intervention. Despite being recognised as an important area in the literature, both the awareness and treatment of mental health problems continue to be an unmet need for people with MS in the current healthcare system (Rieckmann et al., 2018).

Fortunately, psychological interventions exist to treat many of the negative factors associated with lower QOL in people with MS, including depression, anxiety, fatigue, pain and cognitive impairment, although more trials are needed to evaluate their efficacy in people with MS specifically (Fiest et al., 2016). Psychological interventions may be of particular value due to effective medication being poorly tolerated in many people with MS, and due to being more effective at preventing relapse than pharmacotherapy. In addition to treating negative symptoms, interventions designed to improve well-being can help individuals live a valued life in the presence of physical difficulties.

Well-being

Well-being is a multidimensional construct that can be thought of as feeling good and functioning well. Well-being and mental health problems are sometimes thought of as being two ends of one continuum. However, research has shown that despite being moderately interrelated, they are two partially separate constructs (Keyes, 2005). Well-being spans many dimensions, and therefore, measurement usually involves using several measures that cover positive affect, negative affect, satisfaction with life, and positive functioning. QOL instruments are often used to measure one aspect of well-being because they capture an individual's satisfaction with the physical, psychological and social aspects of their life.

Positive psychological interventions (PPIs) focus on cultivating well-being and optimal functioning by using techniques designed to enhance key processes such as positive emotions, competence, optimism, self-acceptance and positive relations (Hone, Jarden, Schofield, & Duncan, 2014). Benefits of the PPI approach are that it may be applied widely for anyone whether or not they are experiencing symptoms of anxiety or depression, and that having high levels of well-being is a protective factor against developing future mental health problems (Keyes, Dhingra, & Simoes, 2010). Although reducing negative symptoms is not the focus of PPIs, research shows that individuals have a significant reduction in depressive and anxiety symptoms after treatment (Chakhssi, Kraiss, Sommers-Spijkerman, & Bohlmeijer, 2018). Both the treatment of mental health problems and the enhancement of well-being are essential in the care of people with MS.

Self-Help Interventions

Whilst there is a substantial evidence base for psychological interventions, the number of available and suitable psychological therapists does not match up to the tremendous burden of common mental health problems both in the general population and for people with MS. Self-help interventions (SHIs) have the potential to fill the shortfall by providing evidence-based therapies with minimal therapist contact time. SHIs have primarily been made up of reading material which guides the user through psychoeducation and exercises or practices to promote change. Traditionally, the material was in printed or book form, and more recently, technology has allowed for internet-based and multimedia interactive SHIs. They are popular because they can be rolled out to large amounts of people with relatively low costs and can be used in a step-cared approach or in addition to other interventions on offer.

Systematic reviews and meta-analyses have shown SHIs to be effective in reducing depression and anxiety symptoms (Cavanagh, Strauss, Forder, & Jones, 2014; Lewis, Pearce, & Bisson, 2012), medically unexplained symptoms (van Gils et al., 2016) and reduce distress in chronic health conditions (Beatty & Lambert, 2013). The majority of studies of SHIs are for those based on a CBT framework, however there is evidence for other models including Acceptance and Commitment Therapy (ACT) (Fledderus, Bohlmeijer, Fox, Schreurs, & Spinhoven, 2013), and PPIs (Schotanus-Dijkstra, Pieterse, Drossaert, Walburg, & Bohlmeijer, 2017; Schueller & Parks, 2012).

Online SHIs may be particularly relevant for people with MS because they foster a sense of autonomy and can be accessed from home, making them accessible for those with limited mobility and positively influencing engagement with treatment (Rieckmann et al., 2018). However, one could also hypothesise that the effects of MS on an individual such as fatigue and cognitive impairment might make accessing and engaging with self-help more difficult than compared to other populations. Previous reviews that have compared the effectiveness of SHIs between populations with different health conditions suggest a significant difference in outcomes (Beatty & Lambert, 2013; Matcham et al., 2014), emphasising the need for an evidence base in individual conditions. It is therefore vital to assess if psychological SHIs are efficacious for people with MS, and if so, what components are associated with better outcomes. As far as the authors are aware, there are no existing systematic reviews that focus on psychological SHIs for people with MS.

The objective of this review was to determine how effective psychological SHIs are for people with MS in reducing distress or improving well-being.

Method

A systematic review of the literature was conducted with evidence sourced up to the date of the search; 9th December 2018. There was no lower date limit enforced. The review process followed the PRISMA guidelines.

Search Strategy

Searches of specialist databases were conducted on 9th December 2018 using the following index/MeSH (Medical Subject Heading) and strings of keyword terms: (Multiple Sclerosis) plus (wellbeing or well-being or "well being" or "quality of life" or "positive affect" or "happiness" or “mental health” or anxiety or depression or distress or “negative affect”) plus (self-help or "self help" or self-management or web-based or "web based" or internet-based or "internet based" or self-directed). To increase breadth of results, and to avoid missing studies that used only the name of a specific intervention rather than generic terms, no search terms related to ‘intervention’ were used. Databases included the Cochrane Database of Systematic Reviews, PubMed and PsycINFO. Search results were exported into Zotero 5.0.60 software and duplicates removed before titles and abstracts were screened in relation to the inclusion/exclusion criteria. The reference lists of all primary studies and review articles were searched for additional references. The search process is shown in Figure 1.

Eligibility criteria and study selection. Search results were excluded or included based on the following pre-defined inclusion criteria:

1. Controlled trials with or without randomisation that reported quantitative outcome data following a self-help intervention in people with MS.
2. Included outcome data on one or more measure of mental health, well-being or QOL
3. Participants with an MS diagnosis of any type (e.g. primary progressive, secondary progressive, relapsing-remitting, progressive relapsing).
4. Published in a peer reviewed publication and available in English
5. Conducted in any country and any setting

To examine the effectiveness of SHIs at improving well-being as broadly as possible, no restrictions were placed on the country or setting of the study, MS related factors such as type or duration, presence of comorbid conditions, symptomatology at baseline, or any individual participant demographic characteristics. Studies were excluded if based on purely educational or exercise interventions. Searches across all databases yielded n = 165 results. After removing duplicates and applying the inclusion/ exclusion criteria to titles and abstracts n = 53 remained. Full text articles were retrieved for closer inspection and after excluding those that did not fulfil the review eligibility, a final total of seven articles were eligible and included in the analysis (see Figure 1).

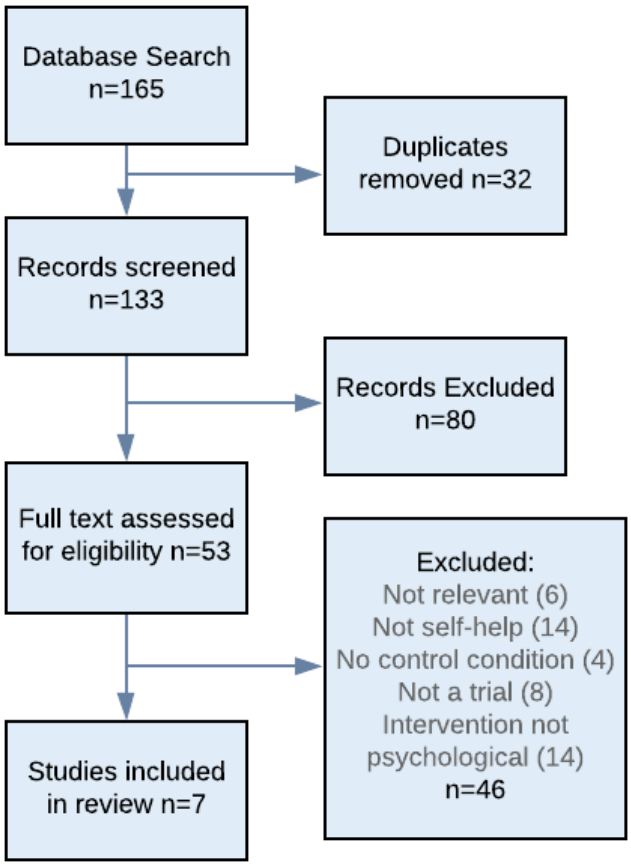

Figure 1. Consort flow diagram of study selection.

Data Extraction Process

Data required for the critical appraisal and of studies was extracted from the final articles by one researcher using a pre-designed data extraction form and entered into a summary table to enable easy comparison. The key areas included: aims, primary/secondary outcomes, sample, intervention content, length of follow-up, analysis methods, results, intervention effectiveness and study limitations.

Strength of Evidence Assessment of Studies

The Cochrane Risk of Bias Tool (Higgins et al., 2011) was used to analyse each study for bias. The risk of bias tool assesses seven domains which are sequence

generation, allocation concealment, blinding of participants and personnel, blinding of outcomes assessed, treatment of incomplete data, selective outcome reporting and other risks of bias. The risk of bias in each subcategory was classified as high, low or unclear. The assessment of bias was conducted by the author (EB) and borderline decisions were discussed with their supervisor (AM) to achieve consensus.

Results

Study Characteristics

Seven RCTs were included in the review (see Table 1). One study was designed to increase levels of physical exercise and was included after discussion due to the intervention being based on psychological theory, involving psychological components, and having psychological outcomes. Of the seven studies, all seven evaluated the impact of the intervention on depression, five on anxiety, four on QOL, and one on Well-being as an outcome. Psychological variables were the primary outcome measure for three studies, and a secondary outcome measure in four studies in which the primary outcome measure was Fatigue (n=3) and physical activity (n=1). Sample sizes varied from 11 to 275, the total and mean numbers of participants were 673 and 96, respectively. The participants' mean ages from the studies ranged between 41- 51 years, with all but one study having a mean age in the 40s. Between 74% and 91% of each sample was female.

Six of the seven studies based their intervention on CBT principles, and one based their behavioural intervention on social cognitive theory. The components of the intervention varied, with three including comprehensive elements of CBT such as psychoeducation, emotions, cognitive restructuring, behavioural activation, self-

monitoring. One of these studies, in addition, included positive psychology strategies to increase well-being. One study focussed mainly on psychoeducation and cognitive restructuring. The intervention in all seven studies was delivered online for participants to access at home. The level of guidance offered during the SHI varied. In three studies, participants received no guidance (pure self-help); in two, weekly email support; in one, up to three telephone support sessions; and one, weekly – monthly Skype support.

Duration of the intervention period ranged between six and twelve weeks for six of the studies, and six months for one study. The total time participants were expected to spend accessing the intervention was specified in four studies and ranged between 4-10 hours; approximately 30 – 60 minutes per week. Six studies used a waitlist control, and one used an active control. Three studies included a long-term follow-up at either three, four, or six months post-intervention.

Table 1

Information for the final studies included in the current review

First Author (year)	Sample Characteristics	Intervention Format and Components	Study Design	Outcomes and Measures	Summary of Findings	Strengths and Limitations	Drop-out Rate
Boeschoten et al., (2017)	N = 171 Mean Age: 48.9 (SD = 10.5) 80% Female Depressed (Moderate and severe) 45% able to walk without aid or rest for 500m	Name: “Minder Zorgen” (“Worry Less”) Duration: 5 modules; 5 hours over 5 – 10 weeks Delivery: Online with weekly email support Components: PST, CBT	RCT Control Condition: WLC Follow-up: 4 months	Primary: Depression; BDI-II Secondary: Anxiety; HADS-A; BAI, Fatigue; FSS, Cognitive Symptoms; MSNQ, Impact of MS; MSIS-29, HR-QOL; EQ-5D Problem-solving; SPSI-R, Mastery; PMS	No treatment effect relative to controls on any measure using an ITT analysis: BDI-II: (*p*=.26, *d*=.23), HADS-A: (*p*=.46, *d*=.11), BAI: (*p*=.10, *d*=.20), FSS: (*p*=.33, *d*=.17), MSNQ: (*p*=.54, *d*=.06), MSIS-29: (*p*=.72, *d*=.03), EQ-5D: (*p*=.35, *d*=.13), SPSI negative: (*p*=.26, *d*=.14), SPSI positive: (*p*=.44, *d*=.10), SPSI avoidant: (*p*=.17, *d*=.16), PMS: (*p*=.50, *d*=.09) Significant within-group reduction in depression (*d*=1.18), maintained at follow-up (*d* = 1.11). Control group also had unexpected significant improvements in depression pre-post (*d* = 0.95) and at follow-up (*d* = 1.12).	- Self-report outcomes -Low adherence rates (67% completers)	11% at T1, 23% at T2.

First Author (year)	Sample Characteristics	Intervention Format and Components	Study Design	Outcomes and Measures	Summary of Findings	Strengths and Limitations	Drop-out Rate
Fischer et al., (2015)	N = 90 Mean Age: 45.28 (SD=11.6) 78% Female Depressed 50% Able to walk without aid or rest for 500m (Measured by HAQUAMS item 15)	Name: “Deprexis” Duration: 10 Modules; 10 hours over 9 weeks Delivery: Online and automated; pure self-help Components: PsyEd, BA, CM, MF, Acc, IPS, Rel, PhysEx, LSMod, PS, expressive writing and forgiveness, PP, EFI	RCT Control Condition: WLC Follow-up: 6 months	Primary: Depression; BDI Secondary: QOL; WHO-QoL BREF HR-QOL; HAQUAMS Fatigue; FSMC	Significant treatment effect relative to controls for depression (p=0·015, d=0·53), Psychological well-being subscale of WHO-QOL-BREF (p=0·04, d=0·44), and motor fatigue subscale of FSMC (p=0·03, d=0·46). Mean depression scores at follow-up were significantly lower than at baseline for the intervention group (p=0.001); statistics relative to controls are not reported. No information regarding secondary outcomes at follow-up is reported**.**	-Excluded suicidal ideation -Self-report outcomes - Lack of reporting of statistics from follow-up	21%
Moss-Morris et al., (2012)	N = 40 Mean Age: 41.0 (SD=14.6)	Name: “MS Invigor8” Duration: 8 Modules; 4-8 hours over 8-10 weeks	RCT	Primary: Fatigue Severity; Fatigue Scale	Significant treatment effect relative to controls on all measures: Fatigue severity (p<.001, d=1.19), fatigue impact (p<.001 d =1.22), Anxiety (p=.001), Depression (p=.001), and HR-QOL (p= .038).	-Low adherence (60.8% completed half)	13%

First Author (year)	Sample Characteristics	Intervention Format and Components	Study Design	Outcomes and Measures	Summary of Findings	Strengths and Limitations	Drop-out Rate
	81.6% Female Significant Fatigue All Participants were ambulatory with or without a stick for at least 100 meters	Delivery: Online; up to 3 telephone support sessions (30-50mins each); automated emails Components: CBT, PsyEd, self-monitoring, CM, BA, EFI, LSMod	Control Condition: WLC No follow-up	Fatigue Impact; MFIS Secondary: Depression; HADS-D Anxiety; HADS-A QOL; EQ-5D		-No Follow-up -Bugs in software	
Motl et al., (2017)	N= 47 Mean Age: 51.9 (SD = 8.6) 76% Female Low activity levels EDSS: median IQR = 3.5 (2.0);	Name: No name given Duration: 4 modules over 6 months. Expected hours not reported. Delivery: Online multimedia website and interactive video	RCT Control Condition: WLC No follow-up	Primary: Physical Activity; GLTEQ and accelerometer data Secondary: Fatigue Severity; FSS	Significant treatment effect relative to controls for physical activity on GLTEQ (P = 0.05, η_p^2 = 0.10), overall fatigue impact on MFIS (P = 0.018, η_p^2 = 0.13), and the Physical subscale of MFIS (P = 0.003, η_p^2 = 0.2), disability on MSWS-12 (P = 0.047, η_p^2 = 0.1) and EDSS (P = 0.03, η_p^2 = 0.11). No significant difference relative to controls on moderate exercise measured	-excluded ages >63 - many excluded at screening -very good adherence (93%)	0%

First Author (year)	Sample Characteristics	Intervention Format and Components	Study Design	Outcomes and Measures	Summary of Findings	Strengths and Limitations	Drop-out Rate
	Most participants were able to walk without aid or rest for at least 500m	courses, with weekly – monthly Skype support. Components: Social Cognitive Theory, self-efficacy, outcome expectations, impediments, goal-setting, self-monitoring		Fatigue Impact; MFIS Pain; SF-MPQ Disability; EDSS; PDDS; MSWS Depression; HADS-D Anxiety; HADS-A	by accelerometer (P = 0.24, η_p^2 = 0.04), fatigue severity (P = 0.10, η_p^2 = 0.06), Cognitive subscale of MFIS (P = 0.2, η_p^2 = 0.04), Psychosocial subscale of MFIS (P = 027, η_p^2 = 0.03), or Pain (P= .70, η_p^2 = .00). No significant difference relative to controls for disability on PDSS (P = 0.1, η_p^2 = 0.07), Depression (P = .10, η_p^2 = .07), or Anxiety (P = .06, η_p^2 = .09).	- very low attrition	
Pöttgen et al., (2018)	N=275 Mean Age: 41.4 (SD; 11.4) 80.5% Female Reporting Fatigue	Name: “ELEVIDA” Duration: Twice a week Over 12 weeks (mean time of modules not specified)	RCT Control Condition: WLC Follow-up: 3 months	Primary: Fatigue; CFS Secondary: Motor and Cognitive Fatigue; FSMC	Significant treatment effect relative to controls for Fatigue on CFS (p=0.0007, d=0.53), FSMC-cognition (p=0.009), FSMC-motor (p=0.006) and Fatigue QOL (p=0.0001); for anxiety (p=0.04), thinking QOL (p=0.045), lower mobility QOL	- 11% participants never accessed programme. - Large variation in adherence	19% at T1 and 21% at T2

First Author (year)	Sample Characteristics	Intervention Format and Components	Study Design	Outcomes and Measures	Summary of Findings	Strengths and Limitations	Drop-out Rate
	PDSS: 64% able to walk without aid or rest for at least 500m	Delivery: Online, pure self-help with Optional text reminders Components: CBT using 'simulated Dialogue', Psych-Ed, Mindfulness, acceptance, CM, imagery, self-monitoring, improving sleep, BA, social support		Anxiety; HADS-A Depression; HADS-D HR-QOL; HAQUAMS Cognitive Symptoms; MSNQ Impact on daily Living; FAI	(p=0.039), and impact on daily living (p=0.005). Treatment effects relative to controls maintained at follow-up for Fatigue on CFS (p=0.008), FSMC-cognition (p=0.004), FSMC-motor (p=0.02) and Fatigue QOL (p=0.01); thinking QOL (p=0.049), and impact on daily living (p=0.02); but not for Anxiety (p=0.052), or lower mobility QOL (p=0.38). No treatment effect relative to controls for depression (p=0.196), cognitive symptoms (p=0.089), upper mobility QOL (p=0.22), mood QOL (p=0.184), or communication QOL (p=0.107).	-Effect sizes not reported for secondary outcomes or at Follow-up - Large sample size - Numerous sensitivity analyses conducted to confirm treatment effects	

First Author (year)	Sample Characteristics	Intervention Format and Components	Study Design	Outcomes and Measures	Summary of Findings	Strengths and Limitations	Drop-out Rate
Tietjen et al., (2018)	N=11 Mean Age: 45.0 (10.0) 90.9% Female Depressed (Moderate - Severe)	Name: ‘‘Think Clearly About Depression’’ Duration: 8 weeks (time duration not specified) Delivery: Online, pure self-help Components: PsyEd, CM, EFI, hopeful thinking	Secondary analyses of a pilot RCT, using sub-sample of people with MS Control Condition: WLC No long-term follow-up	Primary: Depression; PHQ-8 Secondary: Health-related Emotional distress; HDS Self-perceived health status; SRHS Self-efficacy; CDSES	Only descriptive statistics reported due to small sample. Clinically significant improvement in depression for 60% of the treatment group (0% in control). Trends in improvements for health distress, and self-efficacy in relation to managing disease, doing chores, social activities, managing symptoms, and managing depression.	-Small MS sample -descriptive statistics only -No Follow-up -Self-report	13% (of whole sample, not reported for MS sub-sample specifically)
Van Kessel et al., (2016)	N= 39 Mean Age: 45 (SD=8.1)	Name: “MS Invigor8-Plus”	RCT Control Condition: Active control	Primary: Fatigue severity; CFSM;	Significant treatment effect relative to controls for Fatigue on CFSM ($p < 0.01$, $d = 0.99$) and FIS ($p < 0.02$, $d = 0.81$).	-Small sample -Outcome measures completed	21% for Plus Group 55% for Controls

First Author (year)	Sample Characteristics	Intervention Format and Components	Study Design	Outcomes and Measures	Summary of Findings	Strengths and Limitations	Drop-out Rate
	74% Female Estimated EDSS: 46.5% able to walk without aid or rest for at least 500m	Duration: 8 Modules; 4-8 hours over 8-10 weeks Delivery: Online, with weekly email support (10mins per week) Components: CBT, PsyEd, self-monitoring, CM, BA, EFI, LSMod	of MSInvigor8-Only, with no support No follow-up	Fatigue Impact; FIS Secondary: Anxiety; HADS-A Depression; HADS-D	No significant treatment effect relative to controls for Anxiety (p = 0.83) or Depression (p = 0.21). Effect sizes for within-group effects: Fatigue Severity; MSInvigor8-Plus Pre-Post (d=1.35); MSInvigor8-Only Pre-Post (d=0.54) Fatigue Impact: MSInvigor8-Plus Pre-Post (d=1.08); MSInvigor8-Only Pre-Post (d=0.54)	when most Participants had not completed the modules. -No long-term Follow-up	

Outcomes Key: BAI = Beck Anxiety Inventory; CDSES = Chronic Disease Self-Efficacy Scales; CFS = Chalder Fatigue Scale; EQ5D = EuroQol quality of life measure; FSMC = Fatigue Scale For Motor And Cognitive Function; FIS = Fatigue Impact Scale; FSS = Fatigue Severity Scale; GLTEQ = Godin leisure time exercise questionnaire; HADS = Hospital Anxiety and Depression Scale; HAQUAMS = Hamburg Quality of Life Questionnaire in Multiple Sclerosis; HDS = Health Distress Scale; MSNQ = Multiple Sclerosis Neuropsychological Questionnaire; SRHS = Self-Rated Health Scale; MSIS = Multiple Sclerosis Impact Scale; PHQ-8 = Personal Health Questionnaire Depression Scale (Risk question excluded); PMS = Pearlin Mastery Scale; SPSI-R = Social Problem Solving Inventory-Revised; WHO-QoL BREF = WHO Quality of Life scale

Intervention Components Key: Acc = Acceptance; BA = Behavioural Activation; CM = Cognitive Modification; EFI = emotion-focus interventions; IPS = interpersonal skills; LSMod = lifestyle modification; MF = Mindfulness; PhysEx = physical exercise; PP = Positive Psychology; PS = problem solving; PsyEd = psychoeducation; Re = relaxation

Risk of Bias

Studies were assessed for risk of bias using the Cochrane collaboration assessment tool, illustrated in Table 2. Six of the seven papers reported an adequate method of sequence generation and six reported adequate concealment of intervention allocation. None had blinding of participants and study personnel. All seven reported blind outcome assessment; six collected online, and one used assessors blind to group. Five studies reported sufficient information to indicate an adequate assessment of incomplete outcome data, and five indicated a lack of bias due to selective outcome reporting. The overall assessment of the risk of bias was low (n=5), low-moderate (n=1) and high (N=1).

Table 2

Cochrane risk of bias tool assessment for included studies

First Author and year of study	Random sequence generation	Allocation concealment	Blinding of participants and personnel	Blinding of outcome assessment	Missing data addressed	Selective outcome reporting	Other bias	Decision
Boeschoten et al., (2017)	LOW	LOW	HIGH	LOW	LOW	LOW	MODERATE	LOW
Fischer et al., (2015)	LOW	LOW	HIGH	LOW	LOW	MODERATE	LOW	LOW
Moss-Morris et al., (2012)	LOW	LOW	HIGH	LOW	LOW	LOW	MODERATE	LOW
Motl et al., (2017)	LOW	UNCLEAR	HIGH	LOW	UNCLEAR	LOW	MODERATE	MODERATE
Pöttgen et al., (2018)	LOW	LOW	HIGH	LOW	LOW	LOW	MODERATE	LOW
Tietjen et al., (2018)	LOW	UNCLEAR	HIGH	LOW	HIGH	HIGH	UNCLEAR	HIGH
Van Kessel et al., (2016)	LOW	LOW	HIGH	LOW	LOW	LOW	MODERATE	LOW

Well-being

Only one study attempted to measure psychological well-being as an outcome variable in addition to health-related QOL (HR-QOL), using the Psychological well-being subscale of the WHO-QOL-BREF. Fischer and colleagues (2015) saw significant improvement compared to controls in the Psychological well-being subscale only of the WHO-QOL with a small to medium effect size. The intervention ("Deprexis") that focusses on depression is also the only intervention that included PPI strategies.

Anxiety

Five studies examined the impact of an SHI on anxiety outcomes. All used the Hospital Anxiety and Depression Scale (HADS) as the outcome measure, and one additionally used the Beck Anxiety Inventory (BAI). The HADS Anxiety subscale (HADS-A) includes seven items that measure the frequency of anxiety symptoms in the past week. The possible range of scores is 0-21, higher scores indicate more frequent symptoms of anxiety (<8 subclinical; 8-10 mild; 11-14 moderate; 15-21 severe). Baseline HADS-A scores for intervention groups ranged between 5.2 and 10.4, indicating mostly subclinical to mild anxiety. Four studies used a wait-list control, and one used an active control (MSInvigor8-Only vs MSInvigor8-Plus) for comparison. Two had a long-term follow-up.

All five interventions saw a reduction in scores on the HADS-A pre-post. Four reported that the intervention group means at all time points and reduction ranged from -1.1 to -1.8 points. This reduction was significant compared to the control group in only two of the studies (Moss-Morris et al., 2012; Pöttgen et al., 2018). Both interventions, 'MSInvigor8' and 'ELEVIDA', are designed to reduce the severity and impact of fatigue and only included participants with high reported

fatigue. ‘MSInvigor8’ saw the most substantial significant mean reduction of -1.8 points on the HADS-A pre-post compared to the waitlist control group who had an increase of 2.1 points. Pöttgen and colleagues (2018) did not report mean group scores but saw a significant mean difference between intervention and waitlist control groups pre-post of -0.64. The mean difference was maintained at three months follow-up but was no longer significant.

Interestingly the study by Van Kessel and colleagues (2016) of MSInvigor8-Plus, which is the MSInvigor8 intervention with weekly email support from a clinical psychologist instead of telephone support, did not find a significant reduction in anxiety compared to their active control group (Invigorate-Only, no telephone or email support). The Invigor8-Plus group saw a -1.1 reduction pre-post compared to Invigor8-Only who saw a mean reduction of –0.86. The results suggest that some therapist support may be an important component of this intervention for anxiety, and telephone support results in more beneficial results than email support. This study had a higher dropout rate than Moss-Morris and colleagues’ study which may also account for some of the difference in results because an intention to treat analysis was used (13% vs 38%).

The Boeschoten and colleagues (2017) intervention study had the highest baseline Anxiety scores (HADS-A = 10.4), but despite the intervention group achieving a -1.4 change in anxiety levels post-intervention that was maintained at follow-up (-1.6), they did not see a significant mean difference compared to the waitlist control who had similar and unexpected reductions in anxiety. The behavioural intervention (Motl et al., 2017) also saw a trend towards a significant reduction of -1.4 pre-post, which equated to a mean difference of -1.3 compared to controls, with a medium to large effect size; however, this was non-significant. The mean

difference in this study compares favourably to that of ELEVIDA that did achieve significance, and this may be due to the difference in sample size and therefore, power (n = 47 vs n=275). Unfortunately, Pöttgen and colleagues did not report effect sizes for their secondary outcomes for comparison.

Depression

All seven studies examined the impact of a SHI on depression outcomes. Four studies used the depression subscale of the HADS (HADS-D) as an outcome measure, two studies used the BDI-2, and one used the PHQ8. HADS-D scoring is identical to the HADS-A, described previously. The BDI-2 ranges from 0-63: 0-13 subclinical; 14-19 = Mild; 20-28=Mod; 29 = < = Severe. The PHQ8 scores range from 0-24: 5-9 = Mild, 10-14 = Moderate, 15-19 Moderately Severe, and 20+ = Severe. Baseline mean depression scores were in the subclinical-mild range in the four studies for which depression was not a primary outcome, and Mild-Moderate (n = 1) or Moderate-Severe (n = 2) in the studies for which depression was the primary outcome.

A significant treatment effect, relative to controls, was found in two studies (Fischer et al., 2015; Moss-Morris et al., 2012). Baseline mean depression scores in Moss-Morris and colleagues' study (2012) were within the subclinical to mild range (HADS-D = 7.96 SD = 3.64), and they found a significant mean difference between groups pre-post of -4.76 on HADS-D. Effect size was not reported. In Fischer and colleagues (2015) study, mean baseline depression was in the mild to moderate range (BDI-2 = 19.44, SD = 9.02), and they found a significant mean difference between groups pre-post of -4.02 on BDI-2 with a medium effect size, maintained at six months follow-up.

The Boeschoten and colleagues (2017) study included participants with moderate to severe depression at baseline and found a significant within-group reduction in depression score of -7.9 on BDI-2 pre-post with a large effect size, which was maintained at four months follow-up. However, this was not significantly different to the control group who also had unexpected improvements in depression pre-post and at follow-up. Tietjen and colleagues (2018), looking at an MS-only subsample of a larger trial of the 'Think Clearly About Depression' intervention, also designed to treat depression, found a mean difference between groups of -8.43 pre-post on the PHQ8, however due to a small sample size (n = 11) they did not report significance statistics (Tietjen, Wilson, Amiri, & Dietz, 2018). Motl and colleagues' (2017) study of a Behavioural Intervention which found a significant increase in the primary outcome of physical activity saw a mean difference between groups pre-post of -1.3 on HADS-D, which although had a medium effect size, was non-significant. Pöttgen and colleagues (2018), in their intervention for fatigue, saw no significant difference between groups on HADS-D pre-post or at three months follow-up despite seeing a significant reduction in the primary outcome, and to a lesser extent, anxiety. MSInvigor8-Plus, in a sample reporting subclinical to mild levels of baseline depression, saw a mean difference between groups of -1.32 on HADS-D pre-post that was non-significant.

Quality of Life

Only one study (Fischer et al., 2015) included a QOL measure, the WHO-QOl-BREF, as opposed to a HR-QOL measure. HR-QOL focuses on an individual's symptoms and impairments, and how the effects of illness and treatment impact on QOL. The WHO-QOL-BREF covers four domains of Physical, Psychological, Social Relationships and Environment, and scores are converted into a percentile

score (0 -100). Four of the seven studies included a measure of HR-QOL, either the EQ-5D (n = 2) or the HAQUAMS (n = 2). The EQ-5D includes five items, covering five domains: mobility, self-care, usual activities, pain/discomfort and depression/anxiety. Individuals get an index score between 0 and 1, with higher indicating better perceived functioning. In line with previous research, baseline EQ-5D scores for MS samples in this review were lower than population norms for the perspective country and age group (0.47 vs. 0.87 in Netherlands; 0.56 vs. 0.90 in the UK). The HAQUAMS is a MS-specific HR-QOL measure and covers the six domains of Fatigue, thinking, mobility in upper extremities, mobility in lower extremities, mood and communication.

Three of the four studies saw a significant increase in QOL or HR-QOL on at least one subscale pre-post, relative to controls. Effect size was only reported for one study and it was small to medium; The Fischer and colleagues (2015) study saw a significant improvement relative to controls in the Psychological well-being subscale only of the WHO-QOL but saw no difference in the HAQUAMS subscales.

The Moss-Morris and colleagues' study, focussing on fatigue, found a significant improvement in HRQOL on the EQ5D compared to controls of 0.15. Pöttgen and colleagues (2018) found HR-QOL was significantly increased relative to controls for the fatigue, thinking and mobility lower extremities subscales of the HAQUAMS. The improvement was maintained at 3-month follow-up for fatigue and thinking subscales.

Discussion

To the author's knowledge, this is the first review that has specifically focused on examining the effectiveness of SHIs at reducing distress and improving well-being

in people with MS. This review highlights the paucity of high-quality controlled trials available and identifies a significant gap in the research literature. The paucity of studies in this area is in line with a 2014 systematic review and meta-analysis of written SHIs for people with a physical health condition, in which none of the 29 RCTs identified was with an MS population (Matcham et al., 2014). All seven of the studies identified in this review apart from one were published after 2014, which hopefully indicates a recent trend towards an increased interest in the area.

Treatment Effects

Most studies (n = 5) achieved a significant result in their primary outcome compared to controls, suggesting that self-help formats may be suitable for people with MS. Effect sizes in relation to control group in primary outcome were reported in four studies and varied between medium (d = 0.53) and large (d = 1.22). The effect sizes are comparable to those of meta-analyses of SHIs in other populations that generally find a medium effect size (Farrand & Woodford, 2013), or a large effect size for computerised CBT for depression or anxiety (ES = 0.88, NNT 2.15) (Andrews, Cuijpers, Craske, McEvoy, & Titov, 2010), suggesting that some SHIs can be just as effective for people with MS. Of the four studies that reported effect size at post-treatment, two had a long term follow and although treatment gains were still significant, neither reported effect size at this time, so it is unclear to what extent treatment gains are maintained.

Fatigue. SHIs based on a CBT framework aimed at reducing fatigue were successful in all four studies with moderate to large effect size, suggesting that SHIs of this kind can be efficacious in people with MS. Participants also saw an improvement in HR-QOL in the two studies that measured it, which is in line with literature that shows MS-related fatigue significantly impacts HR-QOL (Bol, Duits,

Hupperts, Vlaeyen, & Verhey, 2009). Fatigue is identified by people with MS as one of the most common and troubling problems and the results from this review suggest that SHIs targeting fatigue may be a promising treatment to help alleviate the severity and impact on QOL. Indeed, if as many as 92% of people with MS complain of fatigue (Brañas, Jordan, Fry-Smith, Burls, & Hyde, 2000), and in the absence of any reported negative consequences from fatigue SHIs, then an SHI could justifiably be offered to all those diagnosed. The relatively low baseline levels of depression in this sample of people with MS with high fatigue is surprising considering the strong positive association between fatigue and depression in people with MS (Bol et al., 2009).

Another surprising finding considering this association, is that of the four successful Fatigue SHIs, only one saw significant reduction in depression (MSInvigor8 with telephone support; Moss-Morris et al., 2012). Quality of sleep is one variable that has been shown to account for a high variance in the relationship that exists between MS fatigue and depression (Strober & Arnett, 2005); MS symptoms cause disturbed sleep resulting in both fatigue and low mood, and disturbed sleep as a symptom of depression causes fatigue. Both MSInvogr8 and ELEVIDA contain an 'Improving Sleep' module but ELEVIDA did not see an improvement in depression symptoms, perhaps due to the low baseline levels already mentioned. Another pathway that MSInvigor8 could improve depression and anxiety symptoms is through teaching coping strategies which could be generalised, as coping style (e.g. avoidant versus active) is consistently associated with depression and anxiety (Arnett, Barwick, & Beeney, 2008; Feinstein, Magalhaes, Richard, Audet, & Moore, 2014). The one study that had physical activity as a primary outcome saw significantly increased physical activity and improved motor fatigue, providing

further evidence that people with MS can utilise SHIs to make significant behavioural changes.

Depression. For the two studies that found significant treatment effects on depression relative to controls, only one reported an effect size and had a follow-up (Fischer et al., 2015). The effect size for pre-post was medium to large, and although depression was significantly lower at follow-up compared to baseline, results are not reported relative to controls, and nor was an effect size. Fischer and colleagues (2015) study assessing 'Deprexis' was of high quality and at a low risk of bias, making their findings worthy of future research. 'Deprexis' is fully automated requiring no clinician contact and achieved significant results despite a moderate sample size (*n* = 90) and 21% attrition. The results are in line with previous literature that has identified depression SHIs in particular to be efficacious in a pure-self-help format.

The other study that focussed on depression (Boeschoten, Dekker, et al., 2017), saw a surprising significant improvement in the WLC group as well as the intervention group, which resulted in a non-significant difference between the groups. The results of this review suggest that depression can be improved using SHIs in people with MS, but it may need to be targeted directly; participants who had significantly reduced fatigue or increased physical activity did not necessarily experience a reduction in depression or anxiety symptoms. Baseline mean levels of anxiety and depression in these studies were all in the subclinical to mild range and so a ceiling effect may have occurred where participants did not have much room to improve.

Quality of Life. Three of the four studies included in this review that used a QOL or HR-QOL outcome measure saw an improvement on at least one subscale,

suggesting that improvements in depression and fatigue from SHIs can be translated into an improved quality of life for people with MS. However, effect size was only reported for one study, which was small to medium.

Only one study included a QOL measure, with most choosing to use a health-related QOL measure only. HR-QOL measures have been criticised for having an exclusive focus on symptomology, with little emphasis on positive aspects of QOL. This limits the understanding they provide, as research shows that treating depression in people with MS influences both the negative and positive aspects of QOL (Hart, Fonareva, Merluzzi, & Mohr, 2005).

The 'Deprexis' study (Fischer et al., 2015), that saw a significant improvement in their primary outcome of depression and secondary outcome of Motor Fatigue, found a significant improvement in the psychological well-being subscale of the WHOQOL-BREF (ES =0.44). This intervention was the only one included in the review which utilised PPI modules that included strategies such as expressive writing and forgiveness, suggesting that SHIs could be used to increase the well-being of people with MS. This may be an important and clearly under-researched area in MS.

Interestingly, the changes observed did not translate to an improvement in HR-QOL as measured by the HAQUAMS, including the Mood subscale (ES=0.00). This finding is in line with research suggesting that psychological well-being is a separate construct to HR-QOL. Unlike a lot of HR-QOL measures, the HAQUAMS does include four positive items within the Mood domain regarding contentment, enjoyment, sense of purpose and energy. However, for the people with MS in this particular sample, the questions captured something different to the psychological well-being domain in WHO-QOL-BREF, which includes six questions related to

bodily image, negative feelings, positive feelings, self-esteem, spirituality, thinking, learning and concentration. The results of this study seem at odds with the findings that depression is a strong predicter of reduced HR-QOL, which would suggest an improvement in depression would result in an improvement in HR-QOL. These findings underscore the importance of examining well-being with two or more complementary approaches.

Intervention characteristics

Only three of the interventions were designed to target mental health specifically, and these all focussed on depression. No studies meeting the review criteria were identified that assessed an intervention targeting any type of anxiety, nor any designed specifically to improve well-being in the form of a Positive Psychology Intervention (PPI). The lack of an anxiety focus is surprising considering that a third of people with MS will be diagnosed with an anxiety disorder (Marrie et al., 2013), and that many SHIs exist for anxiety disorders in the general population which have been shown to be efficacious with a large effect size (Lewis et al., 2012). In terms of neuropsychiatric symptoms, fatigue received the most interest (*n* = 3) whilst no studies used an intervention targeting cognitive impairment. This may be due to the medical profession only relatively recently acknowledging the strong impact cognitive impairment has on QOL (Mitchell et al., 2005), and a lack of high quality studies showing efficacy for cognitive rehabilitation (Chiaravalloti & DeLuca, 2008).

All seven of the studies included in this review used an internet-based delivery method. This may reflect a trend towards computer-based programs for self-help as opposed to traditional written material or audio. In a 2010 meta-analysis of

studies comparing guided SHIs to face-to-face therapy, only seven of the 24 guided SHI conditions used a computer based delivery method (Cuijpers, Donker, Straten, Li, & Andersson, 2010). The majority of SHIs were based on a CBT framework, with one also incorporating PPI modules, and one was a behavioural intervention. Therapeutic frameworks which have an evidence base in the general population and in physical health such as ACT and PPI appear to be underrepresented in research in people with MS. A recent systematic review of ACT interventions in long-term conditions, which have considerable evidence base in the domain of physical health, found only two studies with people with MS (Graham, Gouick, Krahé, & Gillanders, 2016), and none were in a self-help format.

There was significant homogeneity in terms of the theoretical framework the interventions were based on, with six out of seven using CBT. Despite this, there was heterogeneity in terms of the intervention content, as well as baseline symptomology and amount of therapist guidance between in the interventions. Due to the small amount of studies, and the varied amount of information regarding intervention content available, it is not possible to ascertain which factors determine the effectiveness of a self-help programme for people with MS. Previous literature suggest that the most effective interventions tend to be based on a therapeutic model and involve relaxation therapy, goal setting and cognitive restructuring (Matcham et al., 2014), which applies to most of the included interventions.

Although there have been studies concluding that increased support or guidance results in better outcomes in SHIs, this notion has been challenged recently by several meta-analyses. For example, a meta-analyses that only included studies at low risk of bias, found no significant difference in outcome between pure-self-help,

minimal contact, or guided SFIs (Farrand & Woodford, 2013); another found no significant difference in outcome between face-to-face CBT and computerised CBT (Andrews et al., 2010); and another found no significant difference between pure SHIs and those with minimal therapist contact (Hirai & Clum, 2006).

There are not enough studies available in this current review to ascertain what level of guidance is optimal for people with MS, which may differ to that of other populations. There does however appear to be a trend towards improved outcomes with increased guidance. For example, the MSInvigor8 intervention saw the largest effect size when delivered face-to-face; effect sizes reduced slightly in an SHI format when accompanied by telephone support, reduced further with email support, and were smallest with no support (Moss-Morris et al., 2012; van Kessel, Wouldes, & Moss-Morris, 2016). Also, within Fischer and colleagues (2015) study, they found higher effect sizes in a subgroup of participants that received one face-to-face assessment appointment in addition to the pure SHI, compared to the whole sample.

Quality of the Studies

The quality of the studies included in this review was variable, but the majority were at a low risk of bias. Only two studies used independent researchers, and five had the programme developers themselves evaluating the intervention, which could introduce bias.

Only four studies included in the review clearly used the gold-standard technique of intention to treat for all primary analysis. One of the key benefits of an RCT is the randomisation of participants into groups, which creates comparable groups whilst avoiding selection bias. Changing these groups by removing incomplete data or excluding dropouts reduces this comparability (Newell, 1992), and is likely to lead

to biased results. Only three of the studies included in this review explicitly reported using a power calculation to ensure the sample would have sufficient power to find significant effects.

Only one study used an active control and the remaining ones used a wait-list control. Whilst this is deemed an appropriate control when assessing a novel behavioural intervention (Mohr et al., 2009), it is noted that WLC patients may decrease their help-seeking behaviours in anticipation of professional help in the future, which may lead to an overestimation of intervention effects (Cuijpers et al., 2010).

Finally, many studies failed to report effect sizes for secondary outcomes and for follow-up outcomes. Failure to report effect sizes compromises comparison of treatment effects across studies, and failure to report over periods of follow-up limits conclusions that can be reached regarding the efficacy of SHIs. This is because inclusion in a study may cause short-term gains that may not be maintained, due to motivating participants through the creation of deadlines and assessments (Lewis et al., 2012).

Attrition and Adherence

Attrition varied between 0 – 55% in intervention groups. Previous research also reports large variance in attrition between computerised SHIs in LTCs of 4 – 62% (Beatty & Lambert, 2013), with no clear variables identified that directly effecting proportions dropping out. In this review there appeared to be a trend in which attrition went down as the amount of therapist guidance went up. This was seen in the pure SHIs having the largest attrition (n = 3; 19-55%); those that offered email support slightly lower (n = 2; 11-23%), and those that provided telephone or Skype guidance the least (n = 2; 0-13%), however the number of studies was too small to

draw any firm conclusions from this. The high levels of attrition rates in the pure self-help studies in this review are similar to those reported by a study of PPI strategies in a pure self-help format which had attrition rates of around 50% (Schueller & Parks, 2012). The relationship between the amount of therapist contact and therapeutic outcome of SHIs is still something that is unclear with conflicting results throughout the literature (Beatty & Lambert, 2013).

Adherence to the intervention varied largely both within and between studies. The average amount of participants completing over half of the modules appeared to be around 60% in those that reported it, which is lower than median adherence for computerised CBT in other populations, which a recent meta-analysis reported is 80% (Andrews et al., 2010). This may suggest that people with MS find it more difficult than other populations to adhere to the full course of a SHI. One included study found that the intervention group continued to use the SHI after the predicted finishing point and post measures, suggesting that people with MS may need to more time allocated to complete the intervention. The exception in this review was a behavioural intervention which retained an impressive 100% of participants in the intervention group, with an adherence of 97% (Motl et al., 2017). Motl and colleagues used Skype guidance which may suggest that more personalised guidance helps adherence to the intervention as well as commitment to the study in people with MS.

Although attrition rates varied and the adherence rates seemed to be below that of SHIs in other populations, participants in the included studies reported similar high satisfaction rates for the SHIs to other populations (Andrews et al., 2010).

Implications for Practice and Policy

Results initially appear encouraging and are in line with existing work that suggests SHIs can be effective in reducing distress, and furthermore give evidence for use in people with MS explicitly. There is however insufficient evidence to determine the exact extent to which SHIs led to improvements in anxiety, depression and QOL in people with MS due to the small number of studies available and the variable quality observed.

The self-help approach has received growing recognition in a climate of limited resources and pressure on psychological services, and reviews have confirmed their potential for effectively treating mental health problems and increasing well-being. This review highlights that there is a paucity of studies evaluating such interventions in individuals with MS. The promising results found in the limited amount of high-quality studies supports the ongoing development of SHIs for people with MS. A recent systematic review that searched Google and App stores in addition to academic online databases for pure online self-help tools (free, publicly available, and not requiring a facilitator) for people with MS and depression found only two relevant self-management programs (Lukmanji et al., 2017). Depression and anxiety did not necessarily improve with improved physical activity and fatigue and vice versa. This finding suggests that the primary cause of an individual's distress should be identified and targeted first, and comorbid symptoms should be monitored and targeted directly if persisting.

Directions for Future Research

The results of this review have highlighted SHIs as a promising area for future research for reducing distress and improving well-being in people with MS. Ensuring sample sizes are large enough to meet power requirements would substantially improve the quality of evidence for future research, although it is

noted that this is a relatively new area of research and a specific population making large samples challenging to ensure.

Further research is required to determine the active ingredients of successful interventions. There is evidence in the broader literature to suggest that some therapist guidance results in better outcomes than pure self-help, however, the evidence is unclear and may be different for people with MS, and different target domains. Determining the level of guidance at which cost-effectiveness is optimal should be an avenue of future research. Furthermore, identifying individual predictors of treatment response will be valuable. For example, there may be differences between people with low versus high levels of disability, subthreshold symptoms versus diagnosed mental health problems, or male versus females. Such investigations were not possible in the included studies due to small size and homogeneity of the sample demographics, which tended to be majority aged in 40s, female and with low disability.

Personal preference of the individual seeking help also needs to be taken into consideration; one could hypothesise that those participating in the included trials found the concept of an SHI acceptable and were willing to engage, and this is unlikely to be the case for all individuals with MS. This variable could be taken into account by collecting data from those who decline to participate in future SHI trials.

Researchers should consider broadening their conceptualization and measurement of QOL by using measures that capture both the positive and negative aspects to QOL, which may provide useful information in the subsequent development of interventions for people with MS.

Existing SHIs based on therapeutic frameworks others than CBT such as ACT-based (Brown, Glendenning, Hoon, & John, 2016) or PPI-based approaches, as

well as those designed to improve anxiety, pain and cognitive symptoms should be assessed with people with MS to see if they would be of benefit. Any existing SHIs should be tailored to people with MS before being trialled to increase potential acceptability (Hind et al., 2010).

Evaluation of a self-help intervention for improving well-being and reducing depression in a sample of people with MS

Abstract

Well-being is an essential aspect of mental health and treatment outcome. 'GAP' is an intervention that teaches goal-setting and planning skills, and in a self-help format has been shown to increase well-being and reduce depression in community and depressed samples. People with MS have lowered well-being and face challenges concerning adjusting important life goals due to unpredictable disease progression and exacerbation. The current study aimed to evaluate the effectiveness of GAP in a pure self-help (SHI) format for increasing well-being and reducing depression for a sample of people with MS.

A Randomised controlled trial was employed. 58 participants with MS were randomly allocated to receive GAP (n=29) or to wait-list control (n=29). Measures of well-being (I-PANAS-SF; Flourishing Scale; FAMS) and depression (PHQ9) were taken at baseline, post-intervention, and eight weeks follow-up. A 2 (treatment vs control) x 3 (Time 1, Time 2, Time 3) repeated measures ANOVA was employed to look at differences between the groups.

The intention to treat (ITT) primary analysis yielded a non-significant result for group x time interactions on all outcomes measures. There was a significant decrease in depression ($F(2,112) = 0.97$, $p = .005$, $\eta_p^2 = 0.02$) and negative affect ($F(2,112) = 4.34$, $p = .018$, $\eta_p^2 = 0.07$) for the whole sample over time. A modified ITT analysis revealed a significant group x time interaction effect for the Family and

Social well-being FAMS subscale only, with a large effect size ($F(2, 40) = 4.65$, $p = .012$, $\eta_p^2 = .12$).

Potential explanations for the results are discussed, including methodological issues and MS-related factors resulting in GAP being ineffective. People with MS may require therapist guidance due to the increased challenges posed by MS requiring more adapting of goals. Further research is needed before recommendations could be made to use GAP in a MS population. Directions for future research are discussed.

Background

Defining Well-being

Well-being is a multidimensional construct that encompasses various psychological, emotional and social dimensions and is an essential aspect of mental health and treatment outcome. Definitions of well-being are typically divided into either hedonic or eudaimonic approaches. Hedonic approaches equate well-being with high positive affect, low negative affect, and high life satisfaction; eudaimonic approaches equate well-being with the actualisation of human experience and potentials, such as relationships or achievements (MacLeod, 2015). The concept of flourishing combines hedonic and eudaimonic components. An individual who is 'flourishing' experiences hedonic well-being, including happiness, calmness and peacefulness, but also eudaimonic well-being, through engagement with life, positive relationships with others and a sense that life is worthwhile (Seligman, 2011). Due to well-being spanning many dimensions, measurement usually involves using several measures that cover positive affect, negative affect, satisfaction with life, and positive functioning.

The concept of cultivating well-being, as opposed to treating illness within clinical psychology, was popularised by the positive psychology movement (Seligman, 2002). As discussed in the previous chapter, interventions that aim to bring about positive thoughts, feelings or behaviours are known as Positive Psychology Interventions (PPIs) and have been found to effective. For example, Sin and Lyubomirsky (2009) carried out a meta-analysis of 51 well-being interventions which revealed that PPIs significantly enhanced well-being (mean r = 0.29) and decreased depressive symptoms (mean r = 0.31). PPIs differ between one another in their approach to cultivating well-being and optimal functioning in people. One

area that is fundamentally linked to well-being and has been utilised for intervention is that of personal goals.

Goals and well-being

Goals and related constructs are pervasive in psychological research and span the history of psychology. Personal goals are important determinants of quality of life; they provide purpose for living, direct individual behaviour, and contribute to long-term patterns of successful development. Goals have been defined as internal representations of meaningful outcomes that people want to achieve (Austin & Vancouver, 1996).

Goals are embedded within the concept of well-being in several ways. An individual's perception of their ability to fulfil goals or aspirations has been emphasised as a fundamental component of well-being theories (Felce & Perry, 1995); Indeed, some theories propose that the definition of well-being itself is to be engaged in pursuing goals that have personal value (Schmuck & Sheldon, 2001). Extensive research has confirmed the relationship between goals and well-being. For example, goal setting and planning skills and working towards intrinsic valued goals are associated with high levels of well-being (MacLeod & Conway, 2005; Sheldon & Kasser, 2001). The pursuit of chosen goals can increase well-being, the type of goals one pursues and even the type of goals one's partner pursues, significantly effect well-being (Headey, Muffels, & Wagner, 2013). In a recent meta-analysis of 85 studies, the authors found the overall effect of successful goal pursuit on well-being yielded a medium to large average effect size of $\rho = .43$. The findings show that goal striving is one of the most important correlates of well-

being, followed by health (ρ = .32), socioeconomic status (ρ = .20), and personality (ρ = .19) (Klug & Maier, 2015).

Similarly, inadequate skills in goal-setting and planning are associated with psychological distress. For example, through the pursuit of maladaptive goals or too many goals, or through perceived failure to achieve goals. Such events are associated with depression, negative affect and can expend an individual's coping resources resulting in perceived stress (Wrosch, Scheier, & Miller, 2013). The specific ways in which personal goals can influence well-being will now be explored further.

Setting goals: Research has shown that having goals in of itself can increase well-being by providing structure and meaning to one's life; Indeed, having goals is a potent predictor of life satisfaction (Brunstein, Schultheiss, & Maier, 1999; Emmons & Kaiser, 1996). In addition, the goal content and appraisal are influential. The relationship between having goals and well-being is strengthened when the goals are self-concordant, i.e., where the goals are those that people either enjoy or value. (Sheldon & Elliot, 1999). Similarly, goals that are about moving towards a desirable outcome, known as 'approach' goals, are associated with higher levels of well-being than those which are about preventing undesirable outcomes, known as 'avoidance' goals (Elliot & Church, 1997).

Imagining achieving those goals: Born from the model of fantasy realisation (Oettingen, Hönig, & Gollwitzer, 2000) the use of imagery has been shown to create strong goal commitments (Oettingen, Mayer, & Thorpe, 2010). Specifically, imagining what it would be like to achieve that goal, then identifying what is

currently preventing progress towards goal achievement, has been shown to increase goal motivation and facilitate action initiation. Research has shown that adults who were more skilled at envisaging positive future opportunities experienced a greater sense of purpose and well-being (Schmitt, Zacher, & Lange, 2013).

Planning and taking actions towards them. Research stresses that the experienced progress towards life goals influences subjective well-being. In a study of neurological patients, researchers found that highly important life goals only increase subjective well-being through perceived progress (Conrad, Doering, Rief, & Exner, 2010). Similarly, in a sample of people with enduring mental illness, increases in well-being constructs were reliant on perceived progression towards goal attainment (Clarke, Oades, Crowe, Caputi, & Deane, 2009). Planning and taking action towards goals is thought to increase well-being through thinking positively about the future and daily activity-based experiences of autonomy, competence, and relatedness that accumulate during the period of striving. This theory was supported by MacLeod and Conway (2005) who found that the number of planned steps taken to achieve goals was linked to thinking positively about the future, which in turn was associated with high positive affect and life satisfaction. Skills in planning such as identifying achievable planned steps are therefore crucial for people to feel as though their goals are attainable and that they are making progress.

Overcoming obstacles to progress. Although making progress towards goals is strongly related to well-being in and of itself, the actual attainment of goals is also related to higher well-being (Klug & Maier, 2015). In their dual process model,

Brandtstädter and Rothermund (2002) propose that individuals take two approaches to life goals that are difficult to obtain. Assimilation mode refers to making active corrective interventions to plans or one's behaviour to make achieving the goal more likely. At times, however, it is impossible for a person to make further progress toward a goal because it is not attainable due to a lack of necessary skills or opportunities, or changes in circumstance that deplete resources or opportunities such as ill health. In the dual process model, the process of adjusting or replacing goals that have become unattainable is named 'accommodation'.

Accommodation has been found to play a meaningful role in the well-being of individuals who experience unattainable goals (Brandtstädter & Rothermund, 2002; Wrosch et al., 2013) through facilitating the abandonment of futile endeavours (goal-disengagement), and through promoting the pursuit of new meaningful activities (goal-reengagement). Specifically, goal-disengagement can prevent/reduce the experience of repeated failure and associated negative aspects of well-being such as negative affect, and goal-reengagement promotes positive aspects of well-being such as positive affect and a sense of purpose. Goal-setting and planning are cognitively based processes that can be targeted in interventions and interventions that aim to enhance these skills have been shown to increase well-being and reduce depression and anxiety symptoms (Cheavens, Feldman, Gum, Michael, & Snyder, 2006).

Goal-setting and Planning (GAP) Intervention

One such intervention devised by MacLeod, Coates, and Hetherton (2008), is the 'Goal-setting and Planning' (GAP) intervention. GAP is designed to increase well-being by helping individuals identify and work towards positive life goals through teaching goal-setting and planning skills. The techniques taught in GAP relate directly to the knowledge derived from the empirical literature described in the previous section. GAP has six modules that cover making goals and selecting self-concordant approach goals to work towards, daily imagery, developing specific and realistic planned steps to achieve chosen goals, identifying and overcoming obstacles, accommodation, and maintaining progress. Throughout the modules, psychoeducation is given about the link between goals, planning and well-being.

GAP was initially devised as a group intervention and was successful in this format in increasing well-being in general community samples (MacLeod, Coates, & Hetherton, 2008), those with long term mental health problems (Farquharson & MacLeod, 2014), and those in a forensic setting (Ferguson, Conway, Endersby, & MacLeod, 2009). GAP has also been evaluated as a self-help intervention. MacLeod et al. (2008) recruited participants from the general population to work individually through the GAP manual over five weeks, with the support of four brief telephone calls. Compared with controls, GAP participants showed a significant increase in positive affect and life satisfaction and a significant decrease in negative affect. A similar result was found with a sample of people with moderate-severe depressive symptoms, with the support of only one telephone call. Within a cross-over design, the whole sample that received GAP saw significant increases in positive affect and life satisfaction and significant decreases in negative affect and depression pre-post intervention, that was maintained at five-week follow-up (Coote & MacLeod, 2012). Most recently, GAP was trialled in a self-help format

with non-clinical sample of working adults and found significant increases in PA and flourishing compared to controls (Oliver & MacLeod, 2018). The results from the three studies just described suggest that the GAP intervention is an effective self-help intervention to increase well-being for community samples and those with depression. Its effectiveness is yet to be tested in those with a long-term physical health condition.

GAP for people with MS

Multiple Sclerosis (MS) is a chronic inflammatory disease of the central nervous system, which is characterised by focal plaques of demyelination and axonal loss in the brain and spinal cord. In addition to focal plaques, there is global neurodegeneration in the brain and spinal cord (Lassmann, 2018), and the changes gradually result in brain tissue loss and atrophy (Mahad et al., 2015). The damage results in wide-ranging symptoms of physical disability and neuropsychiatric symptoms.

As discussed in the previous chapter, the progressive and unpredictable course of MS and threat to personal autonomy from fatigue, disability, and functional loss can harm the well-being of those diagnosed (McCabe & McKern, 2002; Mullins et al., 2001). MS symptomatology has been hypothesised to impact on well-being via two mechanisms, related to 'illness intrusiveness': firstly, by reducing the availability of positive experiences through decreased involvements in valued activities, and secondly, through the reduction of personal control over important domains of life experience (Devins, Seland, Klein, Edworthy, & Saary, 1993).

For those diagnosed, some premorbid important life goals may be no longer attainable or only within limited constraints, and current goals may get disrupted due to unpredictable disease progression and episodes of exacerbation. In this

way, people with MS are at particular risk of experiencing psychological distress through a perceived failure of achieving goals, and this can undermine the individual's optimism or enthusiasm for long-range planning of valued activities. In a recent study, 60% of people with MS reported that having MS interfered with their plans for the future (Lonergan et al., 2015). Goal adjustment capacities are thought to be especially crucial for coping and adjustment among individuals who are confronted with stressful life events such as ill health. For example, a study with female breast cancer survivors showed that goal disengagement capacities were associated with lower levels of daily negative affect and goal reengagement capacities with high levels of positive affect (Wrosch & Sabiston, 2013). Overall, skills in goal setting and planning appear to be very relevant for people with MS, and they may be a group who would particularly benefit from an intervention such as GAP.

Concerning depressive symptoms specifically, results from the systematic review in the previous chapter suggested that there may be limitations of using SHIs for reducing depression for people with MS when it is not directly targeted; only two of the seven studies had significant decreases in depression compared to controls. A proposed explanation is that MS-related depression itself is a more complex persistent condition, meaning it may be unrealistic to expect recovery from a single intervention (Feinstein et al., 2014). However, there is an inverse relationship between successful goal pursuit and depression, and GAP in a self-help format has reduced depression in other populations and may, therefore, have the potential to reduce depressive symptoms for people with MS.

There has been a call for treatment to focus on increased well-being for people with MS (Mitchell, Benito-León, González, & Rivera-Navarro, 2005). Self-help

interventions may be particularly relevant for this group because they foster autonomy, can be easily accessed by those with limited mobility and require minimal support. However, as discussed in the previous chapter, studies testing this theory are limited, and further research is needed to determine the usefulness of SHIs for people with MS. As far as the authors are aware, no intervention designed to increase well-being focussing on goal and planning skills has been evaluated in this population.

Outline of the current study

The aim is to establish whether an online self-help intervention that teaches goal setting and planning skills can improve well-being and reduce depression in a sample of people with MS and whether any improvement is maintained over time. A sample of participants with MS were randomly allocated to receive GAP or to a WLC group. Measures of well-being and depression were taken at three time points; at baseline, post-intervention, and at eight weeks follow-up. After the follow-up measures, the WLC group were given access to GAP. Changes over time in well-being and depression were compared between the two groups.

The hypotheses predict that GAP participants, relative to controls, will show:
1) Increases in positive affect and decreases in negative affect on the I-PANAS -SF
2) Increases in flourishing on The Flourishing Scale
3) Increases in QOL on the Emotional Well-being, Family/Social well-being and General Contentment subscales of the FAMS
4) Decreases in depressive symptoms on the PHQ9

It is also predicted that Intervention effects will be maintained over the 8-week follow-up

Method

Research Approval

Ethical approval was obtained from the research ethics committee at Royal Holloway, University of London. The protocol did not involve recruitment from NHS sites and therefore ethical approval was not sought from the NHS Health Research Authority.

Design

A randomised controlled design was employed. Once recruitment saturation had occurred, the final sample of participants who had completed the online consent form were randomly assigned to the intervention group or the wait-list control (WLC) group using a random number generator function in Excel.

Participants

Between June 2018 and September 2018, a total of 61 people with MS were recruited; three dropped out before baseline measures and therefore a final sample of 58 participants entered the study. Twenty-nine were randomised to the intervention group (5 males, 24 females, mean age 52 *SD*=11.9) and 29 were randomised to the WLC group (10 males, 19 females, mean age = 48 *SD*=11.7). Further demographic and Baseline characteristics of the sample can be found in Table 2, and the number and reason for dropouts are described in Figure 3, Results section.

Recruitment

Individual MS centres that were members of 'Multiple Sclerosis National Therapy Centre (MSNTC)' in the United Kingdom (UK) were identified from the MS National Therapy Centres website and contacted via telephone to ask if they would be interested in circulating information about the study to their members. MSNTC is a membership organisation, created by the member centres and is a registered charity in England & Wales and Scotland. Individual MS support groups, identified through the MS Society website, were also contacted via telephone. One hundred and twenty two MS Centres and MS support groups were contacted in total; of these 22 did not answer and did not respond to an initial answer machine message; 43 were initially contactable but either declined or contact was subsequently lost; and 57 agreed to circulate the study advert either in a newsletter, on social media or by putting up a poster. In addition to contacting MS Centres and support groups, the study advert was shared on social media by the organisations MS Trust, MS UK, and MS Research. Recruitment efforts ended on 31st August 2018 after no further MS Centres or support groups could be identified to contact.

The study advert (Appendix A) included a brief description of the study and a link to an online participant information sheet (PIS; Appendix B). The PIS included a detailed description of the study, the researchers contact details for further information, and a link to the online consent form (Appendix C) for people to opt into the study. Once they had signed the consent form, the participant was sent an email thanking them for signing up to the study and advising that they will be contacted at the start of the study to let them know what group they had been randomised to.

Inclusion and Exclusion Criteria

Inclusion criteria and exclusion criteria were deliberately kept minimal to not restrict a specific population which may have resulted in a sample size that was too small, and to increase external validity. The inclusion criteria applied were a) a diagnosis of MS as identified by the participant, b) fluency in English, and c) consent to inform their GP of their participation in the study. There were no exclusion criteria applied.

Power Analysis

A previous study that evaluated the effectiveness of the GAP intervention within a population of people with depression was used to estimate effect sizes for the current study. The researchers reported medium to large effect sizes (partial n^2 0.058 to partial $n^2 = 0.106$; (Coote & MacLeod, 2012), and on this basis, a medium to large effect size was estimated for the current study. With power at .80, alpha of 0.05 (Cohen, 1992), and predicting a medium to large effect size using a three (time: baseline, post intervention, six-week follow up) x two (Group: Intervention or WLC) mixed ANOVA design, a total sample of 52 participants was required to ensure the study was sufficiently powered to detect significant change within and between conditions. The actual sample obtained (i.e. that completed measures at all three timepoints) was 42.

Measures and materials

A total of four measures were used in the study (see Appendices D - G). A description of each of these is presented below. The measures were chosen to capture depression, different aspects of well-being including positive affect, negative affect, quality of life, and positive functioning. Socio-demographic information was collected for each participant at baseline only. This included age, gender, ethnicity, MS Type, disease duration and GP.

The *Positive and Negative Affect Scale Short Form* (I-PANAS-SF; Thompson, 2007).

The I-PANAS-SF measures the affective component of well-being consisting of 10 affect adjectives for which the participant indicates the extent to which they "feel this way generally" from 1 = "very slightly or not at all" to 5 "extremely". The scale yields two sub-scales of–positive affect and negative affect. Correlations between the original scales and their short counterparts were r=0.92 and r=0.95 for positive and for negative affectivity respectively, indicating they preserve the validity and reliability of the original measure (Mackinnon et al., 1999; Thompson, 2007). Internal consistency is good for the positive scale (a = .73-.79) and acceptable for the negative scale (a = .65-.67), and reliability is adequate with Cronbach alpha values of the short negative and positive affectivity scales of .80 (Gyollai, Simor, Koteles, & Demetrovics, 2011).

The Flourishing Scale (Diener et al., 2010).

The Flourishing Scale measures the cognitive component of well-being, providing a general overview of the individual's self-perceived success in areas such as relationships, self-esteem, purpose, meaning and optimism. It consists of 8 statements for which the participant indicates their agreement on a Likert scale from 1 strongly disagree to 7 strongly agree. All items are phrased in a positive direction, for example "I lead a purposeful and meaningful life". Scores can range from 8 (Strong Disagreement with all items) to 56 (Strong Agreement with all items). High scores signify that respondents view themselves in positive terms in important areas of functioning. The measure has good psychometric properties; when validated in a non-clinical sample, the Flourishing Scale demonstrated good internal reliability, with alpha coefficients ranging from .85 to .86 across the items. It

was also shown to have high convergence with other psychological well-being scales; the Ryff Scales of Psychological Well-being (a = .78) and Deci and Ryan's Basic Need Satisfaction in General scale and (a = .73) (Diener et al., 2010).

Patient Health Questionnaire (Kroenke, Spitzer, & Williams, 2001).

The PHQ-9 is a nine item self-report measure in which respondents indicate how often in the past two weeks they have been bothered by certain depressive symptoms using a Likert Scale from 0 (Not at all) to 3 (Nearly every day), for example "feeling down, depressed or hopeless". The scores range from 0-27, with higher scores indicating greater severity. It has been shown to have excellent Internal reliability (a = 0.86 – 0.89) and test re-test reliability (correlation = 0.84). In terms of validity, sensitivity has been shown to be between 68% and 95% and specificity between 84% and 95%, and scores show significant correlation with the mental health subscale of the SF20 QOL scale (Kroenke et al., 2001).

The Functional Assessment of Multiple Sclerosis (Cella et al., 1996).

The FAMS is a QoL instrument developed for use with people with MS. It has 44 items across six subscales: mobility, symptoms, emotional well-being, general contentment, thinking/fatigue, and family/social well-being. Participants are asked to indicate the extent to which statements applies to them in the past 7 days on a Likert scale from 0 'Not at all' to 4 'Very Much'. Example statements from each respective subscale are as follows: I have trouble walking; I have pain; I feel trapped by my condition; I have accepted my illness; I have a lack of energy; I feel close to my friends. It has shown to have high internal consistency and test-retest reliability ranging from 0.85 to 0.91; concurrent and construct validity; and psychometrically and conceptually distinct subscales (Cella et al., 1996). The

Mobility subscale was shown to have high correlation in a clinical sample with the physical component summary (PCS) score of the SF-36 QOL scale (r = 0.62) and the EDSS (r = 0.68); the Symptoms subscale had a moderate correlation with the Multiscale depression inventory (MDI) Vegetative subscale (r = 0.62) and PCS score of the SF-36 QOL scale (r = 0.56); the Emotional Well-being subscale correlated with the HADS depression score (r = -0.58), as did the General Contentment subscale (r = -0.71); the Thinking/Fatigue subscale correlated with MDI Vegetative subscale (r = 0.66); and the Family/social well-being subscale correlated with HADS Depression score (r = -0.52) and MDI Evaluative score (r = -0.59).

The Intervention

The GAP intervention was branded “Valuing Goals” for purposes of the study. The Valuing Goals programme was in a pure self-help internet-based format and was made available online through a website created in WordPress content management system (Appendix H). WordPress is a widely used online, open source, website creation tool written in PHP, and has been used as a platform for the programme in previous studies. The programme consists of six modules, with downloadable worksheets to back up its structured exercises and tasks. The six modules are as follows:

Module 1: Concepts of well-being, goals and plans and selecting self-concordant goals.

Module 2: Imagining achieving goals over several days.

Module 3: Developing plans for how to achieve one’s goals.

Module 4: Identifying and overcoming obstacles, pros and cons of one's planned steps.

Module 5: Putting into practice what's been learnt in the preceding modules over one week.

Module 6: Review progress and key points on how to maintain progress.

With guidance from an individual with MS minor adjustments were made to the wording and examples used in the programme to make it more acceptable and relevant for people with MS. Feedback for acceptability and ease of use of the final website was sought from the same individual with MS and a physiotherapist for people with MS. Feedback for the website content was positive and no further changes were sought, and for the website functionality one issue was highlighted and subsequently changed which was for the questionnaires to open in a new tab instead of navigating away from the Valuing Goals website.

Procedure

Participants GPs were notified by the researcher of their patient's involvement in the study by email or fax. Each participant was allocated an anonymised participant identification (ID) number which was used to amalgamate their demographic, baseline and follow-up data. The name and ID number pairings were kept with contact details in separate encrypted and password-protected spreadsheets. Participants were also given an individual username and password to log into the Valuing Goals website, where they could access the programme material and measures.

All participants were asked to complete the outcomes measures (I-PANAS-SF; FS; PHQ9; FAMS) at three time points: before the intervention group started the programme (Baseline), when the intervention group ended the programme (Time 1), and at 8-week follow-up (Time 2). The measures were identical at all time points. All data were collected through Google Forms, a password protected online survey system. After Time 2, the WLC group were given access to the programme without any further involvement with the researcher. The study timeline for the two groups is depicted in Figure 2.

	Week 1	Weeks 2-6	Week 6	Weeks 7-14	Week 14	Week 15
Intervention Group	Baseline Measures	Valuing Goals programme	Time 1 Measures	8-week Follow-up period	Time 2 Measures	End of Study
WLC Group	Baseline Measures		Time 1 Measures		Time 2 Measures	Given access to GAP

Figure 2. *Study Timeline*

If a participant indicated suicidal thoughts by scoring on the PHQ9 risk question at any time point they were called to ask permission to send their PHQ9 results to their GP, and then the results were emailed or faxed to their GP to follow up.

The researcher called all participants 1-2 weeks after they were emailed their website logins to answer any questions and resolve any technical issues. The researcher sent email prompts to participants when they were due to complete the measures at all time points. A written prompt within the last programme module reminded participants in the intervention group to complete their Time 1 measures after completing the programme, the timing of which could differ between

individuals. If, however, they had not completed the measures after 8 weeks, an email reminder was sent by the researcher. In previous studies using the programme, adherence data revealed that there was a drop off in adherence after Module 3 and therefore in this study a 'keeping up momentum' email was sent to those in the intervention group at the estimated mid-way point with the aim of encouraging continued participation in the programme (Appendix I).

At completion, all were thanked for their participation and those in the WLC group were given access to the Valuing Goals programme. All participants were entered into a prize draw to win one of two £50 Amazon vouchers.

The use of a computerised random generator for randomisation ensured that the of bias being introduced in the process was low (random sequence generation and allocation concealment). Blinding to group was not possible due to the nature of the intervention. All data were collected online and so no bias could be introduced during data collection. Incomplete outcome data was addressed using an intention to treat principle and all outcomes were reported.

Results

Overview

All statistical analyses were conducted using the Statistical Package for Social Sciences version 21.0 (SPSS; version 21.0). Descriptive statistics are reported to one decimal place, statistical results are reported to two decimal places, and exact p- values are given. The threshold for statistical significance was set at $p < .05$, and hypothesis testing was two-tailed. Effect sizes were calculated for all main analysis results; Partial eta-squared (η_p^2) = 0.01 was considered small, η_p^2 = 0.06 medium, and η_p^2 = 0.14 large. Where assumptions for parametric tests were met then they

were used, but where they were not then non-parametric tests were used. Continuous clinical characteristics and baseline scores are summarised by means and *SD*. Categorical clinical characteristics are described by frequencies and percentages. The screening of the data including normality of distributions and the management of outliers and missing data are described. Treatment effects were examined using a two (treatment condition) x three (Time) analysis of variance (ANOVA), and results are reported for each outcome variable. In line with an intention to treat (ITT) analysis all primary analysis was completed using data from the full sample (*N*=58).

Missing Data

Data were missing at time points due to participants not responding to prompts (dropouts) or withdrawing from the study. There were no individual missing data values within completed measures. A consort diagram, Figure 3, below, illustrates the flow of participants through the study including dropouts and missing data. Forty-two (72.4%) participants provided complete data, and 16 (27.6%) participants had missing data from one or more time point. Overall, 14.1% of data was missing from the dataset, whilst 20.7% of data was missing at Time 1 and Time 2 follow-up.

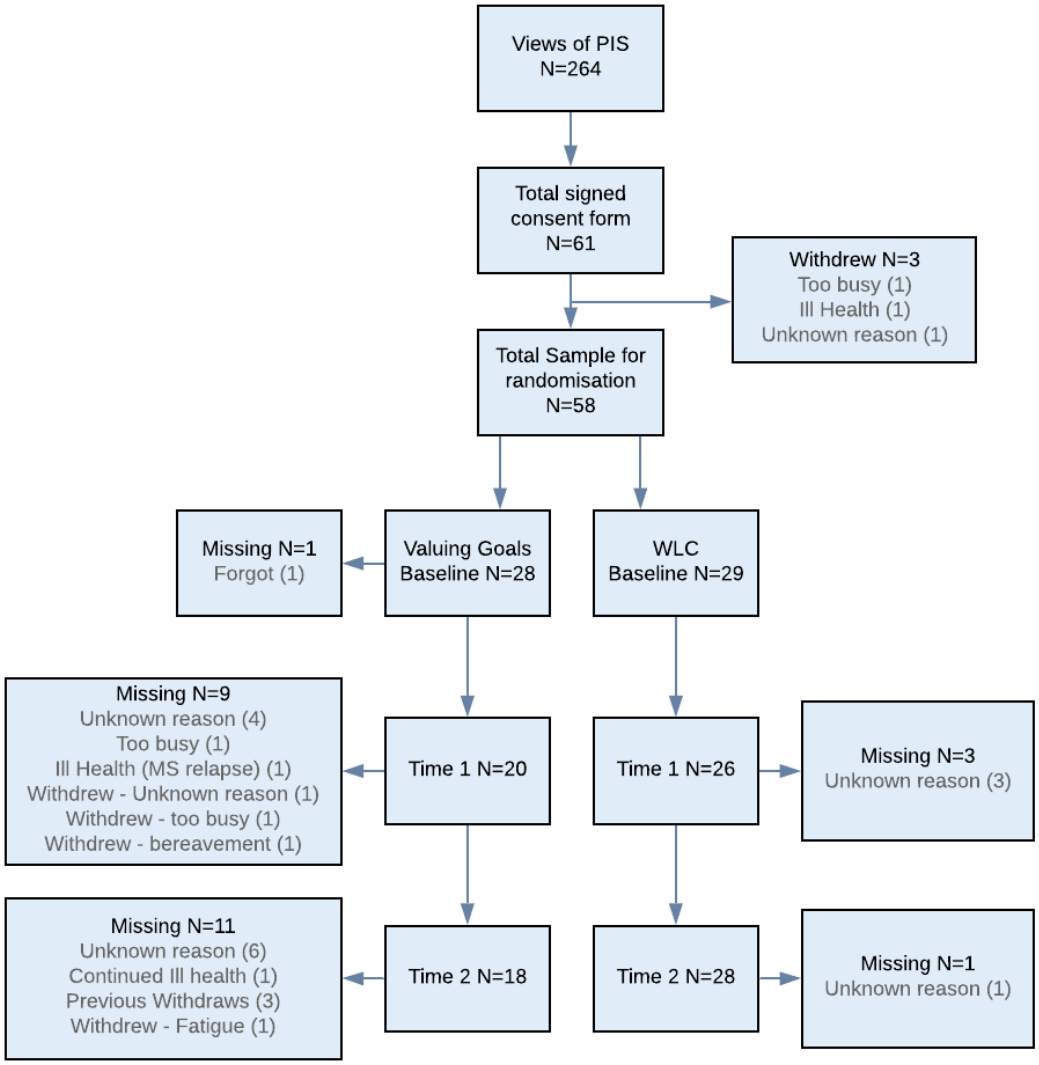

Figure 3. *Consort diagram of participant flow*

An overview of the descriptive statistics for demographic information at baseline for participants who completed and those with missing data are presented in Table 3. Chi-Square revealed that those with missing data were significantly more likely to be in the intervention group rather than WLC (χ^2 (1, N=58) = 8.63, p = .007), but they did not significantly differ from those with complete data on any other baseline demographic or clinical variables. A Missing at Random (MAR) assumption was

made, and the possibility of an interaction effect involving randomised group was accounted for in the MI method employed, described below.

Multiple Imputation (MI) method (Rubin, 2004) was used to manage missing data to allow for an ITT approach for primary analysis. MI was favoured over other methods as it can use information from partially observed cases to better predict imputations in analysis of datasets with several follow-ups (Bell & Fairclough, 2014). Imputed missing scores were conducted for Intervention group and WLC group separately which produces unbiased results when there is a possible interaction effect involving the randomised groups, and has been shown to be more robust than conducting MI for an overall sample in RCTs (Sullivan, White, Salter, Ryan, & Lee, 2018).

All demographic, baseline and outcome variables were included as predictors in the single imputation model for each group, so that any possible associations between related outcomes could aid imputation. As such, predictors in the model were: age, sex, MS type, disease duration; baseline, Time 1 and Time 2 total scores for the I-PANAS-SF positive, I-PANAS-SF negative, PHQ9, FS, FAMS Total and FAMS subscales (mobility; symptoms; emotional well-being; general contentment; thinking/fatigue; family/social well-being). Minimum and maximum values were entered based on the possible minimum and maximums for each variable. In SPSS the random number generator function was used to set the random seed; Mersenne Twister was selected, and the starting point was set at the default fixed value of 2000000. The number of imputations was set to 15 based on recent guidelines suggesting the number of imputations should be equal or greater to the overall percentage of missing data in the dataset. Maximum case draws

were set to 100 and maximum parameter draws set to 50. All primary analysis results are reported using pooled statistics from the 15 MI datasets.

Outliers

Frequencies and boxplots of all variables were examined to identify any extreme outliers, defined as values that were more than three standard deviations away from the variable mean for each group (Field, 2005). Three data points were identified that belonged to the same participant for Baseline PANAS Negative, Time 1 FAMS Symptoms, and Time 1 FAMS Total score. Two-sided winsorizing set at 99.7% was used to change the extreme outliers (*n*=3), in which they were transformed to the value of the next score plus one unit of measurement.

Distribution

The z-scores were calculated and examined for each variable to ascertain any skewness and kurtosis. A distribution was considered normal if a z-score was less than 2.58 and considered slightly skewed or kurtosed but still acceptable for parametric tests if less than 3.29. The PANAS Negative at Time 2 for the control group was positively skewed (Z-score = 3.34). All other variables were found to have acceptable levels of skew and kurtosis. As skewness for The PANAS Negative was only 0.05 over the threshold it was decided to consider all variables as meeting the assumptions for using parametric statistical analysis.

Socio-demographic and baseline characteristics of sample

Table 1 provides the socio-demographic information for randomised groups and total sample and Table 2 provides the baseline characteristics. Independent t-tests revealed that those in the intervention group had had MS for significantly longer than those in the control group (mean difference = 5.2 years, $t(55) = 2.13$, p =

.033). There were no other significant differences between groups. Participants with complete data were also compared with those who dropped out on socio-demographic and baseline characteristics, and no significant differences were found on any of the variables measured.

Table 3

Demographic information; comparing intervention versus control and completers versus dropouts

	Intervention Group (n=29)	Control Group (n=29)	Total Sample (n=58)	Difference (p value)	Completers (n=42)	Dropouts (n=16)	Difference (p value)
Sex (Female)	24 (83)	19 (66)	43 (74)	0.23	30 (71.4)	13 (81.3)	0.343
Age (years)	52 (11.9)	48.2 (11.7)	50.1 (11.9)	0.221	49.2 (11.9)	52.5 (11.8)	0.357
MS Type				0.315			0.58
Primary Progressive	4 (14)	5 (17)	9 (15.5)		7 (16.7)	2 (13.3)	
Relapsing-Remitting	12 (43)	18 (62)	30 (53)		23 (54.8)	7 (46.7)	
Secondary Progressive	11 (39)	5 (17)	16 (28)		10 (23.8)	6 (40)	
Unsure	1 (4)	1 (4)	2 (4)		2 (4.8)	0 (0)	
MS Duration	15.6 (9.8)	10.4 (8.6)	13.0 (9.5)	**0.033***	12.0 (9.3)	15.8 (9.8)	0.18
Ethnicity				0.368			0.263
White	27 (93)	29 (100)	56 (97)		42 (100)	14 (87.5)	
Other	1 (3)	0 (0)	1 (2)		0 (0)	1 (6.3)	
Missing	1 (3)	0 (0)	1 (2)		0 (0)	1 (6.3)	

Note. Bold* = significant at the <.05 level

Table 4

Baseline measures; comparing intervention versus control groups and completers versus dropout

	Intervention Group (n=29)	Control Group (n=29)	Total Sample (n=58)	Difference (p value)	Completers (n=42)	Dropouts (n=16)	Difference (p value)
PHQ9 score	8.9 (5.5)	8.5 (5.5)	8.7 (5.4)	0.745	8.7 (5.6)	8.7 (5.0)	.968
Depressed (PHQ9=>10)	12 (41.4)	10 (34.5)	22 (37.9)	0.787	15 (35.7)	6 (40)	.766
FAMS Total	94.5 (31.9)	102.5 (30.4)	98.5 (31.1)	0.320	99.9 (31.9)	95.3 (29.6)	.631
Mobility	9.8 (6.8)	13.2 (7.9)	11.5 (7.5)	0.077	11.9 (7.9)	10.5 (6.2)	.542
Symptoms	20.7 (5.7)	21.2 (4.4)	20.9 (5.0)	0.689	21.6 (4.8)	19.1 (5.5)	.098
Emotional Wellbeing	18.2 (6.8)	18.2 (6.9)	18.2 (6.8)	0.975	18.1 (7.3)	18.5 (5.6)	.876
Contentment	14.5 (6.3)	14.2 (6.6)	14.4 (6.4)	0.859	14.3 (6.6)	14.7 (5.9)	.837
Thinking and Fatigue	14.9 (9.7)	16.9 (7.9)	15.9 (8.8)	0.374	16.1 (8.7)	15.4 (9.2)	.802
Family and Social	16.5 (7.6)	18.8 (6.6)	17.7 (7.1)	0.231	17.8 (7.2)	17.1 (7.0)	.747
PANAS Positive	15.0 (3.8)	14.1 (4.0)	14.6 (3.9)	0.345	14.1 (3.9)	15.6 (3.6)	.216
PANAS Negative	9.4 (3.9)	9.8 (4.0)	9.6 (4.0)	0.668	9.5 (3.8)	10.0 (3.0)	.630
Flourishing Scale	38.9 (9.8)	38.3 (11.7)	38.6 (10.7)	0.816	38.5 (10.7)	38.8 (11.3)	.938

Well-being outcomes

A 2 (treatment vs control) x 3 (Time 1, Time 2, Time 3) repeated measures ANOVA was employed to look at differences between the groups over the three time periods on the I-PANAS-SF, Flourishing Scale, PHQ9, and the FAMS. An overview of descriptive statistics for clinical variables at baseline and follow-up is presented in Table 5. For the Symptoms subscale of the FAMS Mauchly's test indicated violation of sphericity ($\chi^2(2, N=58) = 9.32$, $p = 0.016$) and as Green-Geisser Epsilon was >0.75 ($p = 0.87$), Huynh-Feldt results are reported. No violation of sphericity was indicated for any other variables.

The intention to treat primary analysis yielded a non-significant result for group x time interactions on all outcomes measures supporting the null hypothesis of no treatment effect. There was a trend towards a treatment effect favouring Valuing Goals on the family/social well-being subscale of FAMS with a small to medium effect size but this was non-significant ($F(2,112) = 2.60$, $p = .088$, $\eta_p^2 = 0.04$). All other measures yielded a small effect size for time x group interactions (η_p^2 range = .01 - .03), except for PANAS Negative which had very small effect size ($\eta_p^2 = .001$). There was a trend towards a main effect of time on the Flourishing Scale but this was also non-significant ($F(2,112) = 2.96$, $p = .063$). There was a significant main effect of Time on PANAS Negative with a medium effect size ($F(2,112) = 4.34$, $p = .018$, $\eta_p^2 = 0.07$) and PHQ9 with a small effect size ($F(2,112) = 0.97$, $p = .005$, $\eta_p^2 = 0.02$).

Paired t-tests revealed that PANAS Negative scores were significantly lower at Time 2 compared to baseline (mean difference = -1.29, $t(57) = -2.76$, $p = .008$), but not did not differ significantly between Baseline and Time 1 (mean difference = -

0.85, $t(57) = -1.83$ $p = 0.08$), or Time 1 and Time 2 (mean difference = -0.44, $t(57) = -1.09$, $p = .30$). Paired t-tests revealed that PHQ9 scores were significantly lower at Time 1 compared to Baseline (mean difference = -1.65, $t(57) = -2.46$, $p = .018$), and at Time 2 compared to baseline (mean difference = -1.98, $t(57) = -2.89$, $p = .004$). They did not differ significantly between Time 1 and Time 2 (mean difference = -.33, $t(57) = -.59$, $p = .56$).

Table 5

Results of ITT 2x3 repeated measures ANOVA

	Intervention (*N*=29)			Controls (*N*=29)			*P* Value		
	Baseline	Time 1	Time 2	Baseline	Time 1	Time 2	Time x Group	Time	Group
FAMS Total	94.7 (31.3)	101.2 (29.2)	100.1 (27.8)	102.5 (30.4)	103.6 (20.0)	105.9 (26.1)	.561	.180	.445
Mobility	9.7 (6.7)	10.4 (6.5)	10.5 (5.8)	13.2 (7.9)	12.6 (6.6)	13.4 (6.9)	.243	.592	.110
Symptoms	20.8 (5.6)	21.6 (5.0)	21.1 (4.5)	21.2 (4.4)	21.3 (3.2)	20.6 (4.1)	.543	.382	.905
Emotional Wellbeing	18.2 (6.7)	19.3 (6.0)	18.1 (6.5)	18.2 (6.9)	18.7 (5.3)	19.4 (5.4)	.380	.470	.837
Content-ment	14.7 (6.2)	15.1 (5.4)	14.7 (5.7)	14.2 (6.6)	14.6 (5.3)	15.7 (5.9)	.339	.419	.944
Thinking/ Fatigue	14.8 (9.5)	16.4 (8.2)	17.0 (8.4)	16.9 (7.8)	16.5 (6.8)	18.3 (7.1)	.578	.177	.517
Family/ Social	16.5 (7.5)	18.3 (5.3)	18.7 (5.5)	18.8 (6.6)	18.3 (5.7)	18.6 (6.7)	.088	.255	.635
PANAS Positive	15.0 (3.8)	15.8 (2.9)	14.6 (3.3)	14.0 (4.0)	14.7 (4.0)	14.9 (4.7)	.392	.268	.462
PANAS Negative	9.5 (3.7)	8.8 (3.3)	8.2 (2.7)	9.7 (3.5)	8.8 (3.0)	8.4 (2.9)	.895	**.018***	.839
PHQ9	8.9 (5.4)	6.5 (4.2)	6.5 (4.1)	8.5 (5.5)	7.6 (4.4)	7.0 (3.6)	.392	**.005***	.674
Flourishing Scale	39.0 (9.7)	41.2 (7.4)	42.2 (7.8)	38.3 (11.7)	39.3 (10.5)	39.9 (10.5)	.681	.063	.471

Note. Bold* = significant at the <.05 level

Modified ITT Analysis

A modified ITT analysis was conducted in which only completers were included (those who completed measures at all three time points), and those who reported they completed two or fewer modules were excluded (N = 1). Results are reported in Table 4. Mauchly's test indicated violation of sphericity For the Emotional Well-being subscale of the FAMS ($\chi^2(2, N = 41) = 7.91$, $p = .019$) and the PANAS Positive ($\chi^2(2, N = 41) = 9.7$, $p = .008$) and as Greenhouse-Geisser Epsilon was >0.75, Huynh-Feldt results are reported for both. No violation of sphericity was indicated for any of the other variables.

There was a significant effect of Time on FAMS Total score with a medium effect size ($F(2, 40) = 3.35$, $p = .04$, $\eta_p^2 = .08$), Thinking and Fatigue subscale with a medium effect size ($F(2, 40) = 3.41$, $p = .038$, $\eta_p^2 = .08$), and PHQ9 with a medium to large effect size ($F(2, 40) = 3.89$, $p = .024$, $\eta_p^2 = .09$).

Paired t-tests revealed that for the whole sample PHQ9 scores at Time 2 were significantly lower than at baseline (mean difference = -1.88, $t(40) = -2.3$, $p = .027$). The differences were non-significant between baseline and Time 1 (mean difference = -1.54, $t(40) = -1.85$, $p = .072$) and between Time 1 and Time 2 (mean difference = -.34, $t(40) = -.49$, $p = .63$). Paired t-tests revealed that for the whole sample FAMS Total scores did not significantly differ between any time points (Baseline to Time 1: mean difference = 3.79, $t(40) = 1.4$, $p = .16$; Time 1 to Time 2: mean difference = 0.57, $t(40) = .26$, $p = .795$; Baseline to Time 2: mean difference = 4.4, $t(40) = 1.6$, $p = .112$). Similarly, no significant differences between time points were found for the Thinking and Fatigue subscale (Baseline to Time 1: mean

difference = 0.58, $t(40) = .57$, $p = .566$; Time 1 to Time 2: mean difference = 1.2, $t(40) = 1.4$, $p = .162$; Baseline to Time 2: mean difference = 1.8, $t(40) = 1.69$, $p = .092$).

There was a significant group x time interaction effect for the Family and Social well-being subscale with a large effect size ($F(2, 40) = 4.65$, $p = .012$, $\eta_p^2 = .12$). One-way repeated measures ANOVA were conducted for each treatment group, which indicated significant differences over time for the intervention group with a very large effect size ($F(2, 14) = 4.56$, $p = .02$, $\eta_p^2 = .25$) but not the control group ($F(2, 25) = .46$, $p = .64$, $\eta_p^2 = .02$). Paired t-tests revealed the intervention group scores significantly increased from baseline to Time 1 (mean difference = 2.73, $t(14) = 2.43$, $p = .029$) and the difference was maintained at time 2 (mean difference = 2.73, $t(14) = 2.25$, $p = .041$).

Table 6

Results of Modified ITT analysis 2x3 repeated measures ANOVA

	Intervention (*n*=15)			Controls (*n*=26)			*P* Value		
	Baseline	Time 1	Time 2	Baseline	Time 1	Time 2	Time x Group	Time	Group
FAMS Total	96.5 (36.8)	103.9 (35.9)	106.0 (35.4)	102.6 (29.6)	103.6 (21.1)	105.9 (24.4)	.346	**.040***	.837
Mobility	10.9 (8.2)	11.6 (8.2)	11.6 (7.5)	12.6 (7.9)	12.6 (7.0)	13.1 (6.4)	.787	.550	.559
Symptoms	22.2 (5.8)	21.8 (6.0)	21.7 (5.8)	21.4 (4.2)	21.4 (3.3)	20.5 (4.2)	.700	.282	.578
Emotional Wellbeing	17.7 (7.9)	19.7 (7.0)	19.5 (7.6)	18.3 (7.1)	18.7 (5.6)	19.4 (5.4)	.548	.098	.922
Content-ment	14.8 (6.7)	15.2 (6.6)	15.8 (7.2)	14.3 (6.7)	14.6 (5.6)	15.6 (5.8)	.938	.136	.829
Thinking and Fatigue	15.4 (9.5)	17.4 (9.2)	19.2 (9.8)	17.1 (7.9)	16.5 (7.1)	18.5 (6.6)	.402	**.038***	.983
Family/ Social	15.5 (8.5)	18.2 (6.5)	18.2 (6.9)	19.0 (6.2)	18.4 (6.0)	18.8 (6.6)	**.012***	.093	.494
PANAS Positive	14.4 (4.0)	15.8 (3.2)	14.8 (4.2)	14.0 (4.0)	14.7 (4.1)	14.8 (4.2)	.568	.158	.683
PANAS Negative	9.6 (4.38)	8.6 (3.68)	8.33 (3.47)	9.5 (3.5)	8.8 (3.2)	8.4 (3.0)	.890	.110	.942
PHQ9	9.3 (5.9)	6.2 (5.3)	6.7 (5.4)	8.3 (5.7)	7.7 (4.6)	6.9 (3.6)	.307	**.024***	.865
Flourishing Scale	39.5 (9.7)	40.5 (7.9)	41.9 (10.6)	38.4 (11.4)	39.2 (11.1)	40.2 (9.7)	.945	.143	.658

Note. **Bold*** = significant at the <.05 level

Website analytics of participant adherence

Analytics data from the website were analysed. As illustrated in Figure X below, completion rate of the modules decreased in succession from 1 to 6; 100% (*N* = 29) of participants accessed Module 1, falling to 24.2% that accessed Module 6 (*N* = 7).

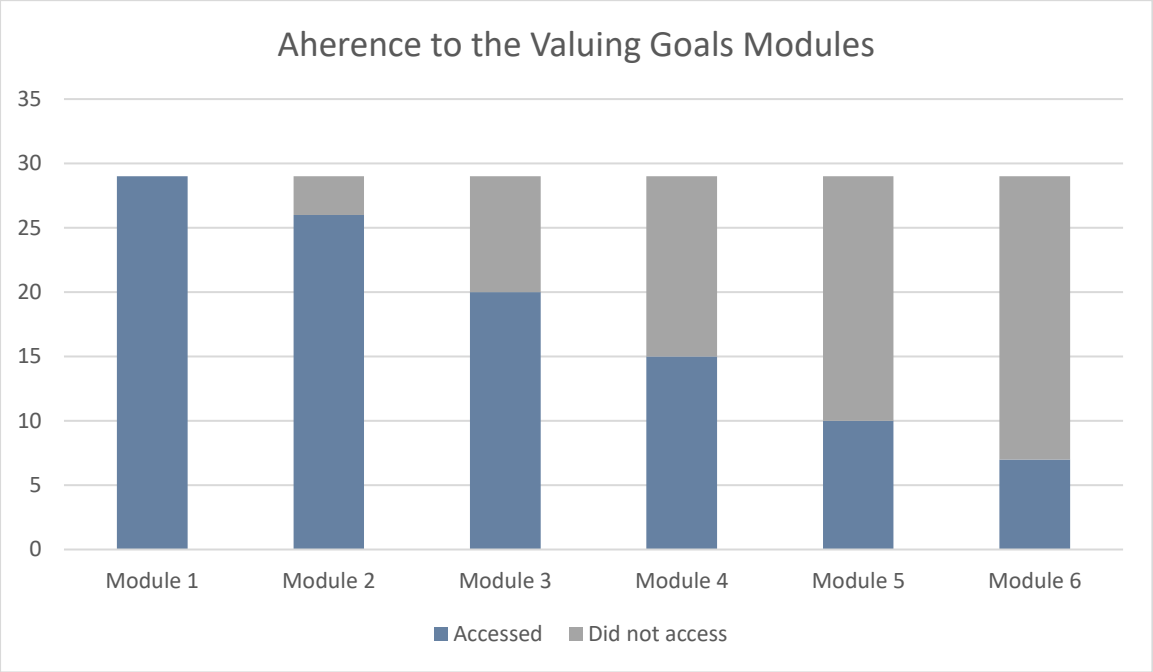

Figure 4. *Adherence to intervention modules*

Discussion

Summary of the Results

The present study aimed to determine whether an online goal-setting and planning (GAP) self-help intervention (MacLeod et al., 2008) could improve the well-being of people with MS in a randomised controlled trial. The study built on evidence from previous trials that showed the GAP intervention to be effective in guided self-help format for people with depression and in an online self-help format for non-clinical

working adults. A pure self-help format was employed in which participants received no intervention guidance, and contact was limited to one telephone call to address practical issues, and a ‘keeping up momentum’ email midway through the program. Sixty-two participants consented to take part in the study, of whom 58 were randomised. MI was used to allow an ITT primary analysis, and a secondary modified ITT analysis was conducted with completers only.

The first predicted outcome of the study, Hypothesis 1, was that, relative to controls, GAP participants would show significantly improved well-being post-intervention. An improvement in well-being was defined as an increase in positive affect, a decrease in negative affect, an increase in flourishing, and an increase in quality of life related to emotional well-being, contentment, and family/social well-being domains. This hypothesis was primarily not supported. GAP participants did not show a significant improvement relative to controls on any measure using an ITT analysis. In a modified ITT analysis, GAP participants showed significant improvement relative to controls in QOL in the family/social domain only.

Hypothesis 2 was that GAP participants, relative to controls, would experience a significant decrease in depressive symptoms post-intervention. This hypothesis was not supported; both GAP and controls experienced a decrease in depressive symptoms.

The final predicted outcome, Hypothesis 3, was that predicted intervention effects as previously outlined would be maintained at eight-week follow-up. This hypothesis was largely redundant due to the previous hypothesis being unsupported. However, in the modified ITT analysis, the increase in family/social QOL found was maintained at eight-week follow-up, partially supporting this hypothesis.

Unexpected results were significant decreases in depressive symptoms between baseline and Time 1 that were maintained at follow-up, and significant decreases in NA between baseline and follow-up for the whole sample.

Baseline Characteristics

Demographic characteristics of the current sample compared to those found in the literature as seen in the previous chapter. Compared to general population samples, baseline measures indicate that participants in the current study were experiencing similar levels of PA and NA, but in line with the literature experienced lower cognitive aspects of well-being as measured by the FS, and higher levels of depressive symptoms (Barak & Achiron, 2011; Bassi et al., 2014). In terms of depressive symptoms, although there was a wide range of scores in our sample, 38% scored 10 or above on PHQ9 compared to around 6% in general populations (Kocalevent, Hinz, & Brähler, 2013; Martin, Rief, Klaiberg, & Braehler, 2006).

Compared to other samples of people with MS, our sample had similar levels of PA and NA (Bassi et al., 2014), and depressive symptoms (Boeschoten, Braamse, et al., 2017). Quality of life as measured by the FAMS was slightly lower in our sample than that reported in other studies of people with MS specifically on subscales of mobility, general contentment, thinking and fatigue, and family/social well-being (Cella et al., 1996; Chang et al., 2002), which may be explained by the current sample having a longer disease duration. In keeping with this, our sample had similar mean flourishing levels as people with MS who self-reported low QOL in a study of newly diagnosed individuals (Strober, 2018).

Consideration of Key Findings

The Comparable reduction in NA and depressive symptoms over time in the WLC group, and the intervention group, was unexpected; the reduction was considerable with medium and medium to large effect sizes respectively for the whole sample. Significant improvement in WLC groups is not observed in most of the literature of SHIs with people with MS, as seen in the previous chapter. Our findings do, however, resemble those of one study by Boeschoten and colleagues (2017), that found significant large decreases in depression in both groups, also maintained at follow-up. The authors ruled out several possible explanations through sensitivity analysis, including those related to study design, depression severity and measurement, and additional mental healthcare outside of the trial. One plausible explanation offered was that improvements were due to regression to the mean as high scores are more likely to decrease, and the sample consisted of people with moderate to severe depressive symptoms. This explanation is unlikely to fit for the current study because mean baseline levels of depression and NA were low. Another explanation discussed which could explain the current findings is that the WLC consisted of highly motivated individuals and enrolment in the trial acted as a trigger to address their issues by other means. Additional possible factors are that anticipation of receiving the intervention at the end of the waiting period produced hope and a reduction in depressive symptoms or reduction represents the natural course of depressive symptoms. In the general population, half of depressed patients recover within three months (Spijker et al., 2002).

The significant increase in the Family/Social subscale of the FAMS found in the sensitivity analysis was unusual in the fact that it occurred in the absence of any observable differences compared to WLC on other outcome measures. This subscale comprises of seven items relating to feeling close to and supported by

others and feeling that others understand and accept one's illness. One mechanism by which improvement could have occurred is through the process of participants talking to family/friends about the GAP modules themselves and this conversation being a platform for open discussion of MS, leading to feeling more understood and supported. Another mechanism could be through the participant's actions taken in stepped plans towards their goals, which may have involved initiating contact with friends/family leading to increased social contact. Research shows that both people in the general population and those with neurological disorders rate the most important life goals as those involving intimacy, suggesting that a large proportion of chosen goals in this study could well be relational (Conrad, Doering, Rief, & Exner, 2010).

The question remains why, if GAP led to significant improvements in family/social QOL, did this not result in improvements on other measures. Based on the literature outlined in the introduction to this chapter, one would expect that engaging in steps towards valued goals to lead to an increase in positive emotions, and improvements in other areas of well-being such as feeling purposeful, engaged, self-efficacious, and optimistic. An explanation that could account for this discrepancy is that the increase in family/social well-being was a chance finding and not a result of the intervention. Such a chance finding could have resulted from multiple testing or from the fact that in the second analysis those remaining in the treatment group were likely to be the more motivated people, due to most case-wise deletions being from the treatment group, and therefore it was not a fair comparison.

The only study identified in the previous chapter that used PPI methods, incorporated into a CBT intervention targeting depression, also had interesting

results concerning different domains and measures of well-being. They saw an improvement in the psychological well-being domain in WhoQoL-BREF which consists of six questions related to body image, negative feelings, positive feelings, self-esteem, spirituality, thinking, learning and concentration, but there was no improvement in the mood subscale of the HAQUAMS which includes four questions related to well-being of contentment, enjoyment, a sense of purpose and energy (Fischer et al., 2015). The results suggest that improvement in one area of well-being does not necessarily result in improvement in others, but the authors do not discuss possible mechanisms or reasons for this.

It is also possible that the study lacked the power to detect significant differences between groups in other domains. In the primary ITT analysis, using MI, we had a large enough sample to detect medium to large effect sizes only, as indicated by our prospective power analysis. The modified ITT analysis was underpowered to detect medium to large treatment effects; differences in the family/social QOL subscale reached significance because it had a large to very-large effect size. There were small effect sizes for time x group interactions in both analyses for all other measures, except for NA; it is possible that with a larger sample the differences between groups indicated by the small effect sizes would be significant.

Another possibility is that there indeed was no significant difference between groups. Previous studies of the GAP program have found it to improve well-being and reduce depression, raising the question of what factors could explain a no treatment effect in this particular group. Potential explanations are discussed below, including ceiling effects, difficulty accessing the intervention due to cognitive difficulties, and other MS-related factors resulting in the intervention being ineffective.

Non-MS-related Factors. One factor that could have influenced efficacy is that our sample was too heterogenous. Stringent inclusion/exclusion criteria are often employed in research to reduce the influence of any confounding variables and target those most likely to benefit from the intervention. Although a decision was made not to have inclusion/exclusion criteria to be as inclusive as possible, some of our participants were unlikely to improve due to experiencing very high levels of well-being and minimal depressive symptoms at baseline. Indeed, the optimal score possible was observed at baseline for at least one participant in four of the seven variables producing a floor effect in the case of NA and PHQ9, and a ceiling effect in the case of well-being QOL and Family/Social QOL.

In terms of depression, two previous studies have shown a significant decrease in depression pre-post GAP. One of these saw a treatment effect relative to a WLC (Coote & MacLeod, 2012), meaning treatment effects were over and above any effect of spontaneous recovery over time, regression to the mean, or anticipatory effect of being in a WLC as mentioned previously as an explanation of the current results. However, the study only recruited those reporting clinical levels of depressive symptoms at baseline, reducing the likelihood of ceiling effects occurring, and potentially reducing the likelihood that those in the WLC could address their difficulties by other means. The other GAP study recruited inmates with a stable major psychotic illness in secure forensic units where GAP was delivered in a facilitated group format, rather than self-help. Mean baseline levels of depression were in the mild range, but there was no control group. It is possible that a control group would have also seen an improvement for the reasons mentioned above, but equally a control group in a forensic setting may not have the same levels of motivation as in the current study as they were not required to

volunteer in the same way, and therefore may not have shown significant improvements.

In regards to well-being however, in the two studies of GAP that used non-clinical populations, (MacLeod et al., 2008; Oliver & MacLeod, 2018), no inclusion/exclusion criteria were applied and compared to controls there was still a significant increase pre-post in NA, PA and Satisfaction with Life, and in PA and Flourishing (but not NA or life satisfaction), respectively. These results suggest that a floor/ceiling effect alone is unlikely to account for lack of an effect in terms of well-being measures in our sample, although it may have reduced power to detect differences.

One factor of interest is the level of guidance provided to participants. In the first GAP study that used a non-clinical sample, participants received four supportive, structured telephone calls while working through the manual, compared to no guidance in the current study. Oliver & MacLeod (2018), however, had a similar pure self-help format to the current study, with only 12 of the 170 intervention participants opting to receive the telephone guidance that was offered. The significant increases in PA and Flourishing observed with small to medium effect sizes demonstrating that, with a group of non-clinical working adults, treatment effects can be observed without any therapist guidance.

The issue of how much guidance influences outcomes of SHIs is still debated. There have been many studies concluding that increased support or guidance results in better outcomes, however, this notion has been challenged by a meta-analysis that did not show a significant difference in outcome between pure-self-help, minimal contact, or guided SFIs (Farrand & Woodford, 2013). For people with MS, although the number of studies is small, there does appear to be a trend

towards improved outcomes with increased guidance, as discussed in the previous chapter. For example, the Invigor8 intervention saw the largest effect size when delivered face-to-face; effect sizes reduced slightly in an SHI format when accompanied by telephone support, reduced further with email support, and were smallest with no support (Moss-Morris et al., 2012; van Kessel et al., 2016). Also, an automated CBT SHI found higher effect sizes in a subgroup of participants that received one face-to-face assessment appointment in addition to the SFI, compared to the whole sample (Fischer et al., 2015).

Although GAP has maintained effectiveness in a pure SHI format with working adults, it is possible that this format falls short of being effective for people with MS. The absence of contact with a 'therapist' may have contributed to a lack of commitment and motivation, seen by reduced viewings of the modules as time went on, or to a reduced efficacy due to individuals not following the content as intended. Indeed, past studies of GAP showing the largest treatment effect sizes were the ones with the most guidance.

The absence of therapist contact may also decrease efficacy by a lack of feedback, as goal-setting theory proposes that people need feedback to track their goal progress (Locke & Latham, 1990). Although the program aims to teach the user in setting manageable self-concordant approach goals, it is possible that further guidance through this process was needed at the beginning of the program, as in previous studies, as goals chosen in modules 1 and 2 are used throughout the remainder of the program. If goals were too broad or unattainable participants would be less likely to experience an increase in well-being through anticipatory affect or goal progress, and if chosen goals were extrinsically motivated they would be unlikely to lead to an increase in well-being, and could even lead to decreases

in well-being (Brunstein, Schultheiss, & Grässman, 1998). The association between approach goals, avoidance goals and well-being, is complicated because all goals sit within a hierarchy of motivations and other goals, and for this reason guidance for some people at the goal setting stage may be required (MacLeod, 2013). As eluded to earlier, the need for guidance may be further indicated in the presence of MS-related symptoms discussed below.

MS-related factors. Baseline scores on the thinking and fatigue subscale of the FAMS indicated that some of our sample had problems with cognitive functioning. The nine items on this subscale refer to the impact of decreased cognitive functioning and fatigue such as “I have trouble starting things because I am tired”, “I have trouble concentrating”, “I have trouble learning new tasks or directions”. Every participant indorsed at least four statements, and 15 (26%) reported experiencing all nine statements ‘quite a bit’ or ‘very much’ in the past seven days. As mentioned earlier, the mean scores on this subscale indicate greater impact than those reported in other samples of people with MS (Cella et al., 1996). It is possible that the impact of cognitive symptoms and fatigue reduced the ability for participants to engage fully with the modules and practice during the week.

Another factor to consider is that people with MS face considerable challenges in day to day functioning that could interfere with goal pursuit; this was one reason that such an intervention was thought to be of potential benefit to people with MS. Dodge and colleagues define well-being as the balance point between an individual’s resource pool and the challenges faced (Dodge, Daly, Huyton, & Sanders, 2012). GAP aims to increase individuals’ internal resources concerning goal pursuit and planning; however, the increase may not be enough to increase well-being when the challenges posed by MS are significant. Specifically, GAP

aims to support individuals in experiencing progress in subjectively important goals using planning skills and guidance, and also to abandon, adapt or substitute unachievable goals. Both these processes of assimilating and accommodating are crucial in the link between having important goals and increased well-being. While the guidance in GAP modules may be sufficient for community samples to do this, people with MS may require additional guidance in this process due to the increased restrictions and challenges posed by MS and requiring more adapting and substituting of goals.

Attrition and Adherence

The low interest and uptake of the study in this population are notable. It is hard to know precisely how many individuals saw information about the study, but the number is likely to be over one thousand based on the membership numbers of MS Centres that circulated the study advert via email and the number of followers of the MS Charities that shared it on social media. Only 264 of those who saw the advert clicked on the link to view the PIS, and of those only 62 consented; this is a considerably lower response rate than seen in other populations for the same intervention (e.g. 31% for those with depression (Coote & MacLeod, 2012)). It may suggest that people with MS feel that it is not going to be helpful or perhaps feel they do not have the energy needed to commit to a longitudinal study.

The 20% attrition seen at both Time 1 and Time 2 is comparable to other studies of SHIs of people with MS, which is lower than that of other populations using SHIs, as described in the previous chapter**.** For example, the studies of pure self-help interventions identified in the systematic review had attrition rates of between 19% and 55% (Fischer et al., 2015; Pöttgen et al., 2018; van Kessel et al., 2016).

In regards specifically to PPI self-help interventions, none were identified for people with MS in the systematic review. However, a study of PPI strategies in a pure online self-help format in a non-clinical population found attrition rates of around 50% (Schueller & Parks, 2012). In the current study, there was greater attrition in the intervention group than the control group. This could be due to the nature of the intervention, potentially being perceived as too great a commitment, unmanageable in the context of living with MS, or perhaps, unhelpful. Another contributing factor could have been differences between groups, unaccounted for by randomisation. The intervention group had a significantly longer disease duration than the control group, and in line with a longer disease duration, had a trend towards older mean age and greater proportion of secondary-progressive subtype than relapsing-remitting subtype. Progressive subtypes of MS are associated with greater symptoms, including cognitive impairment (Huijbregts, Kalkers, de Sonneville, de Groot, & Polman, 2006), so it is possible that participants in the intervention group were affected by MS-related symptoms and these symptoms increased the likelihood of dropping out. A larger sample size would be less likely to have any significant differences between groups after randomisation.

In terms of adherence, sixty-eight percent of participants in the current study accessed half the modules; similar to other SHI studies using MS samples as identified in the systematic review, and similar to working adults using pure self-help GAP (Oliver & MacLeod, 2018). This adherence figure is substantially lower than the average for SHIs. For example, a meta-analysis of computerised CBT found that on average, 80% of people completed all stages (Andrews et al., 2010). The discrepancy in both attrition and adherence to SHIs suggests that people with MS find it more difficult than other populations to adhere to the full course of an

SHI, and that GAP may need changes to improve adherence. A weakness in our website analytic data is that it only tells us the numbers that accessed the modules and not how many completed it; so although 68% accessed Module 3, the proportion that completed it could be substantially less.

There have been some studies evaluating SHIs for people with MS, however, that have found high adherence rates. For example, Motl and colleagues reported 97% adherence to a computerised guided SHI (Motl et al., 2017). The intervention included interactive video courses which could have increased engagement in the content, and participants received guidance through skype support, which is likely to have had an impact on commitment. 85% of their sample had RRMS, compared to 53% if this sample, which could have meant that they were less impacted by symptoms and more able to engage in an intervention. Also, over 80% of people who contacted the study were excluded due to not meeting inclusion/exclusion criteria such as being relapse free in the past 30 days and being non-active during the past six months, meaning only those most likely to benefit from the intervention were included.

Limitations

In spite of its original contributions, there were several limitations to this study. Firstly, the small sample size. Only medium to large effect sizes could be detected in the ITT analysis, and due to missing data, only large to very-large effect sizes could be detected in the modified ITT analysis; this means possible small to medium treatment effects may have been unidentified. Recruitment was sought only through charitable organisations and not clinical settings, which could have resulted in a sample that is not wholly representative of the population of people with MS. The study sample was comparable to reported national epidemiological

estimates in terms of age, gender distribution and MS subtypes distribution (National Multiple Sclerosis Society, n.d.), but the current sample may have differed on variables that were not measured, such as level of disability, income or employment.

The study relied on self-report of a MS diagnosis rather than medical records, which means that some participants may have been mistaken either in the diagnosis of MS itself, or in the subtype indicated. Due to broad inclusion criteria, some participants may have been unable to improve on specific measures, and some may have been limited in their capacity to follow the intervention. In particular, severe cognitive dysfunction may have presented a barrier to adherence and comprehension.

The website analytics data was not detailed enough to identify which ID numbers accessed all of the modules, and so the modified ITT used those that completed all outcome measures as a proxy for adherence to the intervention, which could be flawed as some people may have completed measures but not the intervention and vice versa, skewing results.

Although the chosen measures in this study have been used in published studies with samples of people with MS previously, the I-PANAS and the Flourishing Scale have not been validated specifically for use in people with MS. It is possible, therefore, that certain variables related to MS could affect how accurately these questionnaires measure state affect and flourishing, respectively.

Implications

Our results suggest that GAP intervention could be used to improve the social aspect of well-being for those that adhere to the program. There is no evidence

that other aspects of well-being such as affect, flourishing, and satisfaction in life are improved in this population. The lack of clear benefits in other areas of well-being raises the question of how clinically meaningful improvement in social well-being is for people with MS in isolation. Research shows that the life goal of intimacy is rated as 'extremely' important, is the most important goal for people with neurological disorders, and that they rate it as significantly more important than healthy controls do (Conrad, Doering, Rief, & Exner, 2010). Intimacy is also seen as the most attainable and favourable life goal. The importance placed on social well-being does suggest that an intervention that could help improve the perceived quality of one's relationships would be worthwhile for people with MS. Although worth investigating further, further research is needed before recommendations could be made to use GAP in the population of people with MS due to factors outlined in the limitations section.

Future research

Future research could involve qualitative investigation to find a) how GAP could be made more appealing to people with MS to increase uptake and recruitment, and b) what changes could be made to make GAP more helpful for people with MS. Further improvements could be made to aid adherence and commitment by using multimedia, providing regular reminders by email, and providing telephone guidance at critical points such as choosing a goal to work on during the intervention and making adaptations or replacing goals that turn out to be unattainable. Measures that identify levels of cognitive impairment could be used so that any moderating effects on treatment effects could be seen, while still being inclusive. Finally, it would be helpful for future studies to set up analytics in such a

way that individual user adherence could be tracked to aid sensitivity analysis.

Conclusion

This study aimed to determine whether an online goal-setting and action-planning self-help intervention could improve well-being and reduce depression in people with MS. The intervention was delivered in a pure self-help format in the hope that, if it proved effective, it could be made available to more adults with MS as a low-cost, accessible well-being initiative.

The study did not support the hypothesis that GAP can reduce depression in people with MS relative to controls because although depressive symptoms improved over time for the intervention group, similar improvement was seen in the WLC group. An explanation could be sought in spontaneous recovery of a highly motivated subsample of patients. Concerning well-being, results suggest that for those that adhere to at least three modules, this intervention can significantly improve individual's well-being in the domain of family and social relationships relative to controls and that this improvement is maintained over time. The finding of no treatment effects relative to controls on other measures of well-being could be attributable to low power because small effect sizes were found in favour of the intervention group for all measures except NA. Alternatively, the intervention may not be effective enough in a pure self-help form to increase well-being in other domains for people with MS; the additional challenges posed by MS in the pursuit and adaptation of goals could mean they require additional guidance in the process

Integration, impact, and dissemination

Integration

The overall aim of the current research was to investigate if SHIs could be used to improve well-being for people with MS, and specifically, whether an SHI that teaches goal-setting and planning skills could improve well-being in people with MS. To gain an understanding of what kind of SHIs have already been evaluated in populations of people with MS and how effective they are, a systematic review was conducted. Following this, an RCT was conducted to evaluate the efficacy of an established SHI that teaches goal-setting and planning skills at improving well-being using a sample of people with MS.

The systematic review explored the effectiveness of SHIs for increasing well-being and reducing distress for people with MS. Although SHIs are well evidenced in many populations, results from the systematic review highlighted the paucity of research in the MS populations. Results were mixed in terms of efficacy and suggested that they could be beneficial, but it was difficult to determine what features were related to greater treatment effect. The review revealed a trend towards difficulty in supporting adherence to self-help interventions, especially if therapist guidance was minimal. The paucity of studies found in the systematic review confirmed the belief that SHIs for people with MS is an area that requires further research and gives value to the empirical study. It was surprising that the systematic review did not result in a single study investigating an SHI aimed to improve well-being, considering that this was the primary interest. Again, this gave value to the empirical study in that it appeared to be the first of its kind. Finding no studies of PPI SHIs in the systematic review, however, posed a problem that the results of the review did not provide a full context for the results of the empirical

paper to be formulated within. Although the results of the empirical study could be compared to the results of those included in the review in terms of intervention methodology such as participants characteristics, format and level of guidance given, a comparison could not be made in terms of the content of the intervention. The combined results highlighted the need for future research to address the extent to which the different SHI approaches and techniques are responsible for improvements in outcomes and establishing who responds best to the interventions.

In addition, the limitations found in the studies in the review helped to inform the empirical study. For example, most studies used a health-related QOL measure which have an exclusive focus on symptomatology, which limits the understanding they provide, as research shows that treating depression in people with MS influences both the negative and positive aspects of QOL. For the empirical study, an MS-specific QOL measure was selected that included positive aspects of quality of life such as feeling supported, accepting one's illness and enjoying activities, as well as the negative impact of physical health symptomatology. Results also underscored the importance of examining well-being with two or more complementary approaches, which was achieved in the empirical study by including a measure of flourishing as well as the more traditional well-being measures of satisfaction in life, measured by the FAMS, and affect, measured by the PANAS-SF. Another issue that was encountered was that some studies did not report effect sizes, which made it difficult to make comparisons between studies with different sample sizes. In the empirical study, effect sizes were reported for all possible results so that comparisons could more easily be made. For some studies identified in the systematic review, a risk of bias was introduced through using

case-wise deletion instead of ITT analysis, and so procedures were used to allow a primary ITT analysis in the empirical study.

Securing sample sizes large enough to meet power requirements was a point identified that would substantially improve the quality of evidence for future research. Unfortunately, despite the considerable effort, I was unable to attain a large enough sample size to allow for dropouts. It was also beyond the scope of this thesis to collect data from those who declined to participate to see how these individuals differ to those that participated and to gather information to improve the desirability of the GAP intervention. Difficulties concerning recruitment are further discussed in the following section.

Research Challenges and their Implications for the Project

Recruitment. Recruitment and retention of participants in research are serious methodological concerns because sufficient sample size is required to ensure adequate statistical power (High 2001) and for the avoidance of Type II error (Drew et al. 2002) and high attrition can influence the validity of the research findings. Despite the widely recognised importance, only 31% of RCTs reach their recruitment target (McDonald et al., 2006). The aim for the empirical study was to recruit a minimum of 62 participants, as identified by the a priori power calculation of 52 and accounting for an expected 15% attrition and was hoping to recruit substantially more. The final sample was 58, and more than expected participants dropped-out, in the intervention group particularly, meaning overall, only 46 participants had complete cases at all three time points.

Recruitment for the empirical study started early, and active recruitment ceased not because of time restraint as often occurs but because of using up all available avenues. I now wonder if a larger sample would have been possible to acquire if I

had applied for NHS ethical approval. A deliberate decision to only seek rhul ethical approval and not NHS was made for two reasons. Firstly, the RCT and longitudinal design of my study required a substantial study period, and therefore recruitment had to start and finish promptly; waiting for NHS ethics approval could have delayed the study start and risked running out of time to write up. Secondly, I believed that, as there were many non-NHS organisations involved with supporting people with MS, there would be a sufficient pool of people to recruit from. However, I overestimated what the response rate would be. Despite 57 MS Centres and support groups circulating the study advert to their members, many of which had 100+ members, and three major MS charities publishing the study advert on their social media (who have over 60,000 combined followers), only 264 people viewed the online PIS. Although it is impossible to say precisely how many people saw and read the study advert and therefore what proportion responded, it is almost certainly substantially less than 10% and fair to say reflects a low level of interest. Furthermore, only 61, 23% of those who viewed the PIS, then decided to consent and take part. The low response rate was disappointing, especially after the successful recruitment of so many MS Centres.

The GAP intervention has struggled to gauge interest in previous studies, but to a lesser extent. For example, in Oliver and MacLeod's recent study the advert was circulated to 4000 working adults of whom 335 viewed the PIS (8.4%), and 330 of those then consented (98%). It is maybe not surprising that a lower response rate would occur in the current population considering that physical limitations such as tiredness and ill health are one of the most frequently cited reasons for refusal to participate and withdrawal from a study (Gul & Ali, 2010). In other studies that have recruited MS samples for SHIs, many do not report the recruitment process in

enough detail to ascertain response rate, a factor that could be improved in future reporting of studies. In those that have, the response rate has varied. Two studies used similar recruitment strategies as the current study. Moss-Morris and colleagues advertised on MS Society and MS Trust websites and a press release in the UK over 61 days had 112 people respond. Boeschoton and colleagues who invited people through MS centres and websites in the Netherlands took four years to reach 495 respondents. Others used a combination of clinical and non-clinical recruitment methods. Fischer and colleagues mailed 2,904 people from an MS outpatient clinic database in Germany in addition to online forums, and 241 responded (8.3%), and Pöttgen advertised on MS organisation websites in Germany and via newsletter and leaflet distribution at an outpatient clinic over 140 days and received 531 responses.

With retrospection, it may have been better to have used a pre-post RCT design with no follow-up, which would have relieved time pressure and allowed for NHS ethical approval and new recruitment from clinical settings. The difficulty in recruiting participants does also raise questions about the usefulness of GAP for people with MS, although it may be less daunting and more desirable as an SHI outside of a longitudinal study.

Attrition and missing data. Some attrition is bound to occur in applied research, especially when a follow-up period is involved. The 20.7% observed in the current study at post-intervention and follow-up is within the normal reported range for RCTs, but bias is still expected in the results when the attrition rate exceeds 20% (Dumville, Torgerson, & Hewitt, 2006). Bias is introduced because attrition causes alterations in the composition of experimental and control groups and consequently affects the internal validity of the study. Furthermore, because we have significantly

more participants leaving the intervention group than the WLC group, the likelihood that participants in one group are not balanced with similar participants in the other trial arm is increased.

I attempted to address attrition and the subsequent missing data in several ways. Firstly, I included a table of comparisons of participants with complete data and those with missing data on all baseline variables, as recommended in the literature (Dumville et al., 2006). Comparison through statistical analysis revealed that there were no significant differences between the two groups, which gave some confidence that the risk of bias was not high. However, Altman and colleagues warn that an imbalance of a predictor variable may still bias the study results, even if the imbalance does not reach conventional levels of significance (Altman, 1985). I noted that those with missing data on average were older, had a longer disease duration, had secondary-progressive type of MS which is associated with more disability than relapsing-remitting type, and had lower QOL in the domains of mobility and symptoms than those who had completed measures at every time point.

Secondly, I used MI in order to allow for an ITT approach for primary analysis, and I imputed missing scores separately for Intervention group and WLC group, which produces unbiased results when there is a possible interaction effect involving the randomised groups. Although this is a method used to reduce bias introduced through missing data, it is likely that some bias remained. Despite some of the limitations described, the results from the thesis have advanced the understanding of SHIs for people with MS and have implications for future research.

Impact

The findings overall of this thesis have highlighted a significant need for well-being interventions to be evaluated in people with MS. The systematic review revealed that there were very few SHIs evaluated in people with MS, and none that were designed to increase well-being or to reduce anxiety. The mixed findings from the studies in the systematic review and the empirical study show that although some SHIs can be useful for people with MS, effort needs to be put into understanding the factors associated with effectiveness so that they can be incorporated into the design of future SHIs. The use of online formats has opened up a host of different content options in interventions such as videos, audio, skype therapist support, automated feedback, automated reminders, and interactive interface in which the module content is adapted based on the users' feedback to questions. The studies identified in the systematic review used different combinations of these techniques, making it difficult to compare the impact of any one technique directly. There are also techniques that have not yet been utilised in SHIs for people with MS, for example encouraging social contact between users to discuss progress in online learning groups could help increase motivation and provide peer support which is positively associated with perceived physical and mental health status in MS patients (Krokavcova et al., 2008).

The empirical study evaluated a self-help format of a well-established well-being intervention which enabled comparison of results to those of previously published studies of the same intervention with different populations. The demonstration of efficacy in other populations allowed some confidence that the lack of treatment effect in the primary analyses, if not a Type II error, was due to factors associated with MS and not that the intervention is ineffective in isolation. The fact that an intervention that has been shown to increase well-being in various populations was

less effective in a sample of people with MS highlights the different needs people with MS have and the importance of even well-established interventions to be evaluated in this population before being recommended. It also gives support to the idea of interventions being designed collaboratively with people with MS from the early stages, rather than adapting interventions that have already been designed for use in other populations. Collaboration between people with MS and professionals is an area that has only recently been championed, with initiatives such as the Multiple Sclerosis in the 21st-century group bringing the groups together to form shared understandings and shared decisions for future research to improve patient outcome (Rieckmann et al., 2018).

The current project will raise awareness of mental health and well-being for people with MS in the community, an area that people with MS identify as an unmet need in current society (Rieckmann et al., 2018). The study supported the knowledge in the literature that many people with MS report clinically relevant depressive symptoms and lower levels of well-being than the general population, and this is pertinent to professionals working with people with MS. Professionals should be asking about mental health and can use depression screening questionnaires and well-being questionnaires to open up conversations.

The points raised from this thesis are relevant to the research community, who can take the lead on building our currently limited knowledge of how to help increase well-being and reduce distress for people with MS. To further knowledge in the field, it will require: collaboration and input from people with MS; the evaluation and subsequent adaptation of interventions known to be effective in other populations; the incorporation of qualitative information into study designs; and the use of experimental methods to identify optimal factors associated with effectiveness.

Dissemination

One aim of making the results of this widely available is to encourage engagement in and promote the need for research into self-help interventions for people with MS. The first stage of the dissemination strategy involves providing a plain English summary of the research findings to those who participated in the study, as interaction with the end-user is key to the successful dissemination of findings (Wilson, Petticrew, Calnan, & Nazareth, 2010). The PIS and email correspondence stated that participants would receive a summary of the findings via email unless they would prefer not to, and as no one opted out, all participants will be sent one. The fact that no participants opted out, and indeed several people responded saying that they would appreciate knowing the results, is a testament to the value of this part of the strategy. The plain English summary will be written by me with collaboration with my supervisor, Professor Andy MacLeod, and agreed upon before sharing with participants. As the results of the study may be disappointing for participants to hear, the wording is especially important so as not to have any negative impact.

A version of the plain English summary will also be given to the contact staff member of all participating MS Centres and MS support groups. Dissemination to staff members will be an essential step as staff members at centres can then become sources of continued dissemination themselves, which may have a more significant impact than when coming from the researchers (Petty, Gleicher, & Jarvis, 1993).

The main message that I will be aiming to communicate is that efforts should be made to open up conversations about mental health and well-being for people with

MS and that the research community need to collaborate with people with MS to create and evaluate psychological interventions for people with MS. A focus on self-help interventions that can be accessed remotely and require minimal therapist contact time, thereby making them accessible for large amounts of people, is especially important.

Channels of communication will include direct emails to participants, emails to contact staff members at MS Centres and support groups, pieces in individual MS Centre newsletters and social media, and a piece in the MS National Therapy Centres (MSNTC) charity newsletter and social media. Beyond the immediate participants and recruitment sites, dissemination to a broader audience involves traditional publication routes through submission of both the systematic review and empirical project to established MS and well-being journals.

References

Altman, D. G. (1985). Comparability of Randomised Groups. *Journal of the Royal Statistical Society: Series D (The Statistician)*, *34*(1), 125–136. https://doi.org/10.2307/2987510

Andrews, G., Cuijpers, P., Craske, M. G., McEvoy, P., & Titov, N. (2010). Computer Therapy for the Anxiety and Depressive Disorders Is Effective, Acceptable and Practical Health Care: A Meta-Analysis. *PLOS ONE*, *5*(10), e13196. https://doi.org/10.1371/journal.pone.0013196

Arnett, P. A., Barwick, F. H., & Beeney, J. E. (2008). Depression in multiple sclerosis: Review and theoretical proposal. *Journal of the International Neuropsychological Society*, *14*(5), 691–724. https://doi.org/10.1017/S1355617708081174

Austin, J. T., & Vancouver, J. B. (1996). Goal constructs in psychology: Structure, process, and content. *Psychological Bulletin*, *120*(3), 338–375. https://doi.org/10.1037/0033-2909.120.3.338

Barak, Y., & Achiron, A. (2011). Happiness and Personal Growth are Attainable in Interferon-Beta-1a Treated Multiple Sclerosis Patients. *Journal of Happiness Studies*, *12*(5), 887–895. https://doi.org/10.1007/s10902-010-9234-6

Bassi, M., Falautano, M., Cilia, S., Goretti, B., Grobberio, M., Pattini, M., … Delle Fave, A. (2014). The coexistence of well- and ill-being in persons with multiple sclerosis, their caregivers and health professionals. *Journal of the Neurological Sciences*, *337*(1), 67–73. https://doi.org/10.1016/j.jns.2013.11.018

Beatty, L., & Lambert, S. (2013). A systematic review of internet-based self-help therapeutic interventions to improve distress and disease-control among adults with chronic health conditions. *Clinical Psychology Review*, *33*(4), 609–622. https://doi.org/10.1016/j.cpr.2013.03.004

Bell, M. L., & Fairclough, D. L. (2014). Practical and statistical issues in missing data for longitudinal patient-reported outcomes. *Statistical Methods in Medical Research*, *23*(5), 440–459. https://doi.org/10.1177/0962280213476378

Boeschoten, R. E., Braamse, A. M. J., Beekman, A. T. F., Cuijpers, P., van Oppen, P., Dekker, J., & Uitdehaag, B. M. J. (2017). Prevalence of depression and anxiety in Multiple Sclerosis: A systematic review and meta-analysis. *Journal of the Neurological Sciences*, *372*, 331–341. https://doi.org/10.1016/j.jns.2016.11.067

Boeschoten, R. E., Dekker, J., Uitdehaag, B. M., Beekman, A. T., Hoogendoorn, A. W., Collette, E. H., … van Oppen, P. (2017). Internet-based treatment for depression in multiple sclerosis: A randomized controlled trial. *Multiple Sclerosis (Houndmills, Basingstoke, England)*, *23*(8), 1112–1122. https://doi.org/10.1177/1352458516671820

Bol, Y., Duits, A. A., Hupperts, R. M. M., Vlaeyen, J. W. S., & Verhey, F. R. J. (2009). The psychology of fatigue in patients with multiple sclerosis: A review. *Journal of Psychosomatic Research*, *66*(1), 3–11. https://doi.org/10.1016/j.jpsychores.2008.05.003

Brañas, P., Jordan, R., Fry-Smith, A., Burls, A., & Hyde, C. (2000). Treatments for fatigue in multiple sclerosis: A rapid and systematic review. *Health Technology Assessment (Winchester, England)*, *4*(27), 1–61.

Brandtstädter, J., & Rothermund, K. (2002). The Life-Course Dynamics of Goal Pursuit and Goal Adjustment: A Two-Process Framework. *Developmental Review*, *22*(1), 117–150. https://doi.org/10.1006/drev.2001.0539

Brown, M., Glendenning, A., Hoon, A. E., & John, A. (2016). Effectiveness of Web-Delivered Acceptance and Commitment Therapy in Relation to Mental Health and Well-Being: A Systematic Review and Meta-Analysis. *Journal of Medical Internet Research*, *18*(8). https://doi.org/10.2196/jmir.6200

Brunstein, J. C., Schultheiss, O. C., & Grässman, R. (1998). Personal goals and emotional well-being: The moderating role of motive dispositions. *Journal of Personality and Social Psychology*, *75*(2), 494–508. https://doi.org/10.1037/0022-3514.75.2.494

Brunstein, J. C., Schultheiss, O. C., & Maier, G. W. (1999). The pursuit of personal goals. *Action & Self-Development*, 169–196.

Cavanagh, K., Strauss, C., Forder, L., & Jones, F. (2014). Can mindfulness and acceptance be learnt by self-help?: A systematic review and meta-analysis of mindfulness and acceptance-based self-help interventions. *Clinical Psychology Review*, *34*(2), 118–129. https://doi.org/10.1016/j.cpr.2014.01.001

Cella, D. F., Dineen, K., Arnason, B., Reder, A., Webster, K. A., Karabatsos, G., … Stefoski, D. (1996). Validation of the Functional Assessment of Multiple Sclerosis quality of life instrument. *Neurology*, *47*(1), 129–139. https://doi.org/10.1212/WNL.47.1.129

Chakhssi, F., Kraiss, J. T., Sommers-Spijkerman, M., & Bohlmeijer, E. T. (2018). The effect of positive psychology interventions on well-being and distress in clinical samples with psychiatric or somatic disorders: A systematic review and meta-analysis. *BMC Psychiatry*, *18*(1), 211. https://doi.org/10.1186/s12888-018-1739-2

Chang, C.-H., Cella, D., Fernández, O., Luque, G., de Castro, P., de Andrés, C., … de Ramón, E. (2002). Quality of life in multiple sclerosis patients in Spain. *Multiple Sclerosis Journal*, *8*(6), 527–531. https://doi.org/10.1191/1352458502ms851oa

Cheavens, J. S., Feldman, D. B., Gum, A., Michael, S. T., & Snyder, C. R. (2006). Hope Therapy in a Community Sample: A Pilot Investigation. *Social Indicators Research*, *77*(1), 61–78. https://doi.org/10.1007/s11205-005-5553-0

Chiaravalloti, N. D., & DeLuca, J. (2008). Cognitive impairment in multiple sclerosis. *The Lancet Neurology*, *7*(12), 1139–1151. https://doi.org/10.1016/S1474-4422(08)70259-X

Clarke, S. P., Oades, L. G., Crowe, T. P., Caputi, P., & Deane, F. P. (2009). The role of symptom distress and goal attainment in promoting aspects of psychological recovery for consumers with enduring mental illness. *Journal of Mental Health*, *18*(5), 389–397. https://doi.org/10.3109/09638230902968290

Conrad, N., Doering, B. K., Rief, W., & Exner, C. (2010). Looking beyond the importance of life goals. The personal goal model of subjective well-being in neuropsychological rehabilitation. *Clinical Rehabilitation*, *24*(5), 431–443. https://doi.org/10.1177/0269215509358930

Coote, H. M. J., & MacLeod, A. K. (2012). A Self-help, Positive Goal-focused Intervention to Increase Well-being in People with Depression. *Clinical Psychology & Psychotherapy*, *19*(4), 305–315. https://doi.org/10.1002/cpp.1797

Cuijpers, P., Donker, T., Straten, A. van, Li, J., & Andersson, G. (2010). Is guided self-help as effective as face-to-face psychotherapy for depression and anxiety disorders? A systematic review and meta-analysis of comparative outcome studies. *Psychological Medicine*, *40*(12), 1943–1957. https://doi.org/10.1017/S0033291710000772

Devins, G. M., Seland, T. P., Klein, G., Edworthy, S. M., & Saary, M. J. (1993). Stability and determinants of psychosocial well-being in multiple sclerosis. *Rehabilitation Psychology*, *38*(1), 11–26. https://doi.org/10.1037/h0080288

Diener, E., Wirtz, D., Tov, W., Kim-Prieto, C., Choi, D., Oishi, S., & Biswas-Diener, R. (2010). New Well-being Measures: Short Scales to Assess Flourishing and Positive and Negative Feelings. *Social Indicators Research*, *97*(2), 143–156. https://doi.org/10.1007/s11205-009-9493-y

Dodge, R., Daly, A. P., Huyton, J., & Sanders, L. D. (2012). The challenge of defining wellbeing. *International Journal of Wellbeing*, *2*(3). Retrieved from https://internationaljournalofwellbeing.org/ijow/index.php/ijow/article/view/89

Dumville, J. C., Torgerson, D. J., & Hewitt, C. E. (2006). Reporting attrition in randomised controlled trials. *BMJ*, *332*(7547), 969–971. https://doi.org/10.1136/bmj.332.7547.969

Elliot, A., & Church, M. (1997). A Hierarchical Model of Approach and Avoidance Achievement Motivation. *Journal of Personality and Social Psychology*, *72*(1), 218–232.

Emmons, R. A., & Kaiser, H. A. (1996). Goal orientation and emotional well-being: Linking goals and affect through the self. *Striving and Feeling: Interactions among Goals, Affect, and Self-Regulation*, 79–98.

Farquharson, L., & MacLeod, A. K. (2014). A brief goal-setting and planning intervention to improve well-being for people with psychiatric disorders. *Psychotherapy and Psychosomatics*, *83*(2), 122–124. https://doi.org/10.1159/000356332

Farrand, P., & Woodford, J. (2013). Impact of support on the effectiveness of written cognitive behavioural self-help: A systematic review and meta-analysis of randomised controlled trials. *Clinical Psychology Review*, *33*(1), 182–195. https://doi.org/10.1016/j.cpr.2012.11.001

Feinstein, A., Magalhaes, S., Richard, J.-F., Audet, B., & Moore, C. (2014). The link between multiple sclerosis and depression. *Nature Reviews Neurology*, *10*(9), 507–517. https://doi.org/10.1038/nrneurol.2014.139

Felce, D., & Perry, J. (1995). Quality of life: Its definition and measurement. *Research in Developmental Disabilities*, *16*(1), 51–74. https://doi.org/10.1016/0891-4222(94)00028-8

Ferguson, G., Conway, C., Endersby, L., & MacLeod, A. (2009). Increasing subjective well-being in long-term forensic rehabilitation: Evaluation of well-being therapy. *The Journal of Forensic Psychiatry & Psychology*, *20*(6), 906–918. https://doi.org/10.1080/14789940903174121

Field, A. (2005). *Discovering Statistics Using SPSS*. SAGE Publications Ltd.

Fiest, K. M., Walker, J. R., Bernstein, C. N., Graff, L. A., Zarychanski, R., Abou-Setta, A. M., … Marrie, R. A. (2016). Systematic review and meta-analysis of interventions for depression and anxiety in persons with multiple sclerosis. *Multiple Sclerosis and Related Disorders*, *5*, 12–26. https://doi.org/10.1016/j.msard.2015.10.004

Fischer, A., Schröder, J., Vettorazzi, E., Wolf, O. T., Pöttgen, J., Lau, S., … Gold, S. M. (2015). An online programme to reduce depression in patients with multiple sclerosis: A randomised controlled trial. *The Lancet. Psychiatry*, *2*(3), 217–223. https://doi.org/10.1016/S2215-0366(14)00049-2

Fledderus, M., Bohlmeijer, E. T., Fox, J.-P., Schreurs, K. M. G., & Spinhoven, P. (2013). The role of psychological flexibility in a self-help acceptance and commitment therapy intervention for psychological distress in a randomized controlled trial. *Behaviour Research and Therapy*, *51*(3), 142–151. https://doi.org/10.1016/j.brat.2012.11.007

Graham, C. D., Gouick, J., Krahé, C., & Gillanders, D. (2016). A systematic review of the use of Acceptance and Commitment Therapy (ACT) in chronic disease and long-term conditions. *Clinical Psychology Review*, *46*, 46–58. https://doi.org/10.1016/j.cpr.2016.04.009

Gul, R. B., & Ali, P. A. (2010). Clinical trials: The challenge of recruitment and retention of participants. *Journal of Clinical Nursing*, *19*(1–2), 227–233. https://doi.org/10.1111/j.1365-2702.2009.03041.x

Gyollai, A., Simor, P., Koteles, F., & Demetrovics, Z. (2011). Psychometric properties of the Hungarian version of the original and the short form of the Positive and Negative Affect Schedule (PANAS). *Neuropsychopharmacologia Hungarica: A Magyar Pszichofarmakologiai Egyesulet Lapja= Official Journal of the Hungarian Association of Psychopharmacology*, *13*(2), 73.

Hart, S., Fonareva, I., Merluzzi, N., & Mohr, D. C. (2005). Treatment for depression and its relationship to improvement in quality of life and psychological well-being in multiple sclerosis patients. *Quality of Life Research*, *14*(3), 695–703. https://doi.org/10.1007/s11136-004-1364-z

Headey, B., Muffels, R., & Wagner, G. G. (2013). Choices Which Change Life Satisfaction: Similar Results for Australia, Britain and Germany. *Social Indicators Research*, *112*(3), 725–748. https://doi.org/10.1007/s11205-012-0079-8

Higgins, J. P. T., Altman, D. G., Gøtzsche, P. C., Jüni, P., Moher, D., Oxman, A. D., … Sterne, J. A. C. (2011). The Cochrane Collaboration's tool for assessing risk of bias in randomised trials. *BMJ*, *343*, d5928. https://doi.org/10.1136/bmj.d5928

Hind, D., O'Cathain, A., Cooper, C. L., Parry, G. D., Isaac, C. L., Rose, A., … Sharrack, B. (2010). The acceptability of computerised cognitive behavioural therapy for the treatment of depression in people with chronic physical disease: A qualitative study of people with multiple sclerosis. *Psychology & Health*, *25*(6), 699–712. https://doi.org/10.1080/08870440902842739

Hirai, M., & Clum, G. A. (2006). A Meta-Analytic Study of Self-Help Interventions for Anxiety Problems. *Behavior Therapy*, *37*(2), 99–111. https://doi.org/10.1016/j.beth.2005.05.002

Hone, L. C., Jarden, A., Schofield, G. M., & Duncan, S. (2014). Measuring flourishing: The impact of operational definitions on the prevalence of high levels of wellbeing. *International Journal of Wellbeing*, *4*(1). Retrieved from https://www.internationaljournalofwellbeing.org/ijow/index.php/ijow/article/view/286

Huijbregts, S. C. J., Kalkers, N. F., de Sonneville, L. M. J., de Groot, V., & Polman, C. H. (2006). Cognitive impairment and decline in different MS subtypes. *Journal of the Neurological Sciences*, *245*(1–2), 187–194. https://doi.org/10.1016/j.jns.2005.07.018

Kern, S., Schrempf, W., Schneider, H., Schultheiß, T., Reichmann, H., & Ziemssen, T. (2009). Neurological disability, psychological distress, and health-related quality of life in MS patients within the first three years after diagnosis. *Multiple Sclerosis Journal*, *15*(6), 752–758. https://doi.org/10.1177/1352458509103300

Keyes, C. L. M. (2005). Mental Illness and/or Mental Health? Investigating Axioms of the Complete State Model of Health. *Journal of Consulting and Clinical Psychology*, *73*(3), 539–548. https://doi.org/10.1037/0022-006X.73.3.539

Keyes, C. L. M., Dhingra, S. S., & Simoes, E. J. (2010). Change in Level of Positive Mental Health as a Predictor of Future Risk of Mental Illness. *American Journal of Public Health*, *100*(12), 2366–2371. https://doi.org/10.2105/AJPH.2010.192245

Klug, H. J. P., & Maier, G. W. (2015). Linking Goal Progress and Subjective Well-Being: A Meta-analysis. *Journal of Happiness Studies*, *16*(1), 37–65. https://doi.org/10.1007/s10902-013-9493-0

Kocalevent, R.-D., Hinz, A., & Brähler, E. (2013). Standardization of the depression screener Patient Health Questionnaire (PHQ-9) in the general population. *General Hospital Psychiatry*, *35*(5), 551–555. https://doi.org/10.1016/j.genhosppsych.2013.04.006

Kroenke, K., Spitzer, R. L., & Williams, J. B. W. (2001). The PHQ-9. *Journal of General Internal Medicine*, *16*(9), 606–613. https://doi.org/10.1046/j.1525-1497.2001.016009606.x

Krokavcova, M., van Dijk, J. P., Nagyova, I., Rosenberger, J., Gavelova, M., Middel, B., … Groothoff, J. W. (2008). Social support as a predictor of perceived health status in patients with multiple sclerosis. *Patient Education and Counseling*, *73*(1), 159–165. https://doi.org/10.1016/j.pec.2008.03.019

Lassmann, H. (2018). Multiple Sclerosis Pathology. *Cold Spring Harbor Perspectives in Medicine*, *8*(3), a028936. https://doi.org/10.1101/cshperspect.a028936

Lewis, C., Pearce, J., & Bisson, J. I. (2012). Efficacy, cost-effectiveness and acceptability of self-help interventions for anxiety disorders: Systematic review. *The British Journal of Psychiatry*, *200*(1), 15–21. https://doi.org/10.1192/bjp.bp.110.084756

Locke, E. A., & Latham, G. P. (1990). *A theory of goal setting & task performance.* Prentice-Hall, Inc.

Lonergan, R., Kinsella, K., Fitzpatrick, P., Duggan, M., Jordan, S., Bradley, D., … Tubridy, N. (2015). Unmet needs of multiple sclerosis patients in the community. *Multiple Sclerosis and Related Disorders*, *4*(2), 144–150. https://doi.org/10.1016/j.msard.2015.01.003

Lublin, F. D., & Reingold, S. C. (1996). Defining the clinical course of multiple sclerosis. *Neurology*, *46*(4), 907. https://doi.org/10.1212/WNL.46.4.907

Lukmanji, S., Pham, T., Blaikie, L., Clark, C., Jetté, N., Wiebe, S., … Patten, S. B. (2017). Online tools for individuals with depression and neurologic conditions: A scoping review. *Neurology. Clinical Practice*, *7*(4), 344–353. https://doi.org/10.1212/CPJ.0000000000000365

Mackinnon, A., Jorm, A. F., Christensen, H., Korten, A. E., Jacomb, P. A., & Rodgers, B. (1999). A short form of the Positive and Negative Affect Schedule: Evaluation of factorial validity and invariance across demographic variables in a community sample. *Personality and Individual Differences*, *27*(3), 405–416. https://doi.org/10.1016/S0191-8869(98)00251-7

MacLeod. (2015). Well-Being: Objectivism, Subjectivism or Sobjectivism? *Journal of Happiness Studies*, *16*(4), 1073–1089. https://doi.org/10.1007/s10902-014-9561-0

MacLeod, Coates, & Hetherton, J. (2008). Increasing well-being through teaching goal-setting and planning skills: Results of a brief intervention. *Journal of Happiness Studies*, *9*(2), 185–196. https://doi.org/10.1007/s10902-007-9057-2

MacLeod, & Conway. (2005). Well-being and the anticipation of future positive experiences: The role of income, social networks, and planning ability. *Cognition & Emotion*, *19*(3), 357–374. https://doi.org/10.1080/02699930441000247

Mahad, D. H., Trapp, B. D., & Lassmann, H. (2015). Pathological mechanisms in progressive multiple sclerosis. *The Lancet Neurology*, *14*(2), 183–193. https://doi.org/10.1016/S1474-4422(14)70256-X

Marrie, R. A., Fisk, J. D., Yu, B. N., Leung, S., Elliott, L., Caetano, P., … for the CIHR Team in the Epidemiology and Impact of Comorbidity on Multiple Sclerosis. (2013). Mental comorbidity and multiple sclerosis: Validating administrative data to support population-based surveillance. *BMC Neurology*, *13*(1), 16. https://doi.org/10.1186/1471-2377-13-16

Martin, A., Rief, W., Klaiberg, A., & Braehler, E. (2006). Validity of the Brief Patient Health Questionnaire Mood Scale (PHQ-9) in the general population. *General Hospital Psychiatry*, *28*(1), 71–77. https://doi.org/10.1016/j.genhosppsych.2005.07.003

Matcham, F., Rayner, L., Hutton, J., Monk, A., Steel, C., & Hotopf, M. (2014). Self-help interventions for symptoms of depression, anxiety and psychological distress in patients with physical illnesses: A systematic review and meta-analysis. *Clinical Psychology Review*, *34*(2), 141–157. https://doi.org/10.1016/j.cpr.2014.01.005

McCabe, M. P., & McKern, S. (2002). Quality of Life and Multiple Sclerosis: Comparison Between People with Multiple Sclerosis and People from the General Population. *Journal of Clinical Psychology in Medical Settings*, *9*(4), 287–295. https://doi.org/10.1023/A:1020734901150

McDonald, A. M., Knight, R. C., Campbell, M. K., Entwistle, V. A., Grant, A. M., Cook, J. A., … Snowdon, C. (2006). What influences recruitment to randomised controlled trials? A review of trials funded by two UK funding agencies. *Trials*, *7*(1), 9. https://doi.org/10.1186/1745-6215-7-9

Mitchell, A. J., Benito-León, J., González, J.-M. M., & Rivera-Navarro, J. (2005). Quality of life and its assessment in multiple sclerosis: Integrating physical and psychological components of wellbeing. *The Lancet Neurology*, *4*(9), 556–566. https://doi.org/10.1016/S1474-4422(05)70166-6

Mohr, D. C., & Cox, D. (2001). Multiple sclerosis: Empirical literature for the clinical health psychologist. *Journal of Clinical Psychology*, *57*(4), 479–499.

Mohr, Spring, B., Freedland, K. E., Beckner, V., Arean, P., Hollon, S. D., … Kaplan, R. (2009). The Selection and Design of Control Conditions for Randomized Controlled Trials of Psychological Interventions. *Psychotherapy and Psychosomatics*, *78*(5), 275–284. https://doi.org/10.1159/000228248

Moss-Morris, R., McCrone, P., Yardley, L., van Kessel, K., Wills, G., & Dennison, L. (2012). A pilot randomised controlled trial of an Internet-based cognitive behavioural therapy self-management programme (MS Invigor8) for multiple sclerosis fatigue. *Behaviour Research and Therapy*, *50*(6), 415–421. https://doi.org/10.1016/j.brat.2012.03.001

Motl, R. W., Hubbard, E. A., Bollaert, R. E., Adamson, B. C., Kinnett-Hopkins, D., Balto, J. M., … McAuley, E. (2017). Randomized controlled trial of an e-learning designed behavioral intervention for increasing physical activity behavior in multiple sclerosis. *Multiple Sclerosis Journal - Experimental, Translational and Clinical*, *3*(4), 2055217317734886. https://doi.org/10.1177/2055217317734886

Mullins, L. L., Cote, M. P., Fuemmeler, B. F., Jean, V. M., Beatty, W. W., & Paul, R. H. (2001). Illness Intrusiveness, Uncertainty, and Distress in Individuals With Multiple Sclerosis. *Rehabilitation Psychology*, *46*(2), 139–153.

National Multiple Sclerosis Society. (n.d.). Who Gets MS? Retrieved 2 August 2019, from National Multiple Sclerosis Society website: http://www.nationalmssociety.org/What-is-MS/Who-Gets-MS

Oettingen, G., Hönig, G., & Gollwitzer, P. M. (2000). Effective self-regulation of goal attainment. *International Journal of Educational Research*, *33*(7), 705–732. https://doi.org/10.1016/S0883-0355(00)00046-X

Oettingen, G., Mayer, D., & Thorpe, J. (2010). Self-regulation of commitment to reduce cigarette consumption: Mental contrasting of future with reality. *Psychology & Health*, *25*(8), 961–977. https://doi.org/10.1080/08870440903079448

Oliver, J. J., & MacLeod, A. K. (2018). Working adults' well-being: An online self-help goal-based intervention. *Journal of Occupational and Organizational Psychology*, *91*(3), 665–680. https://doi.org/10.1111/joop.12212

Petty, R. E., Gleicher, F., & Jarvis, W. B. G. (1993). Persuasion theory and AIDS prevention. In *The social psychology of HIV infection* (pp. 155–182). Hillsdale, NJ, US: Lawrence Erlbaum Associates, Inc.

Pöttgen, J., Moss-Morris, R., Wendebourg, J.-M., Feddersen, L., Lau, S., Köpke, S., … Gold, S. M. (2018). Randomised controlled trial of a self-guided online fatigue intervention in multiple sclerosis. *Journal of Neurology, Neurosurgery, and Psychiatry*, *89*(9), 970–976. https://doi.org/10.1136/jnnp-2017-317463

Rieckmann, P., Centonze, D., Elovaara, I., Giovannoni, G., Havrdová, E., Kesselring, J., … Ben-Amor, A.-F. (2018). Unmet needs, burden of treatment, and patient engagement in multiple sclerosis: A combined perspective from the MS in the 21st Century Steering

Group. *Multiple Sclerosis and Related Disorders*, *19*, 153–160. https://doi.org/10.1016/j.msard.2017.11.013

Rubin, D. (2004). *Multiple Imputation for Nonresponse in Surveys (Wiley Classics Library)*. Retrieved from http://www.amazon.ca/exec/obidos/redirect?tag=citeulike09-20&path=ASIN/0471655740

Schmitt, A., Zacher, H., & Lange, A. H. de. (2013). Focus on opportunities as a boundary condition of the relationship between job control and work engagement: A multi-sample, multi-method study. *European Journal of Work and Organizational Psychology*, *22*(5), 505–519. https://doi.org/10.1080/1359432X.2012.698055

Schmuck, P., & Sheldon, K. M. (2001). Life goals and well-being: To the frontiers of life goal research. *Life Goals and Well-Being: Towards a Positive Psychology of Human Striving*, 1–17.

Schotanus-Dijkstra, M., Pieterse, M. E., Drossaert, C. H. C., Walburg, J. A., & Bohlmeijer, E. T. (2017). Possible mechanisms in a multicomponent email guided positive psychology intervention to improve mental well-being, anxiety and depression: A multiple mediation model. *The Journal of Positive Psychology*, 1–15. https://doi.org/10.1080/17439760.2017.1388430

Schueller, S. M., & Parks, A. C. (2012). Disseminating Self-Help: Positive Psychology Exercises in an Online Trial. *Journal of Medical Internet Research*, *14*(3). https://doi.org/10.2196/jmir.1850

Seligman, M. E. (2002). Positive psychology, positive prevention, and positive therapy. In *Handbook of positive psychology* (Vol. 2, pp. 3–12).

Seligman, M. E. (2011). Flourish: A visionary new understanding of happiness and well-being. *Policy*, *27*(3), 60–61.

Sheldon, & Elliot. (1999). Goal striving, need satisfaction, and longitudinal well-being: The self-concordance model. *Journal of Personality and Social Psychology*, *76*(3), 482–497.

Sheldon, & Kasser. (2001). Goals, Congruence, and Positive Well-Being: New Empirical Support for Humanistic Theories. *Journal of Humanistic Psychology*, *41*(1), 30–50. https://doi.org/10.1177/0022167801411004

Spijker, J. A. N., De Graaf, R., Bijl, R. V., Beekman, A. T., Ormel, J., & Nolen, W. A. (2002). Duration of major depressive episodes in the general population: Results from The Netherlands Mental Health Survey and Incidence Study (NEMESIS). *The British Journal of Psychiatry*, *181*(3), 208–213.

Strober. (2018). Quality of life and psychological well-being in the early stages of multiple sclerosis (MS): Importance of adopting a biopsychosocial model. *Disability and Health Journal*, *11*(4), 555–561. https://doi.org/10.1016/j.dhjo.2018.05.003

Strober, & Arnett. (2005). An examination of four models predicting fatigue in multiple sclerosis. *Archives of Clinical Neuropsychology*, *20*(5), 631–646. https://doi.org/10.1016/j.acn.2005.04.002

Sullivan, T. R., White, I. R., Salter, A. B., Ryan, P., & Lee, K. J. (2018). Should multiple imputation be the method of choice for handling missing data in randomized trials? *Statistical Methods in Medical Research*, *27*(9), 2610–2626. https://doi.org/10.1177/0962280216683570

Thompson, E. R. (2007). Development and validation of an internationally reliable short-form of the positive and negative affect schedule (PANAS). *Journal of Cross-Cultural Psychology*, *38*(2), 227–242.

Tietjen, K., Wilson, M., Amiri, S., & Dietz, J. (2018). Online Depressive Symptom Self-Management: Comparing Program Outcomes for Adults With Multiple Sclerosis Versus Those With Other Chronic Diseases. *The Journal of Neuroscience Nursing: Journal of*

the American Association of Neuroscience Nurses, *50*(1), 13–19. https://doi.org/10.1097/JNN.0000000000000328

van Gils, A., Schoevers, R. A., Bonvanie, I. J., Gelauff, J. M., Roest, A. M., & Rosmalen, J. G. M. (2016). Self-Help for Medically Unexplained Symptoms: A Systematic Review and Meta-Analysis. *Psychosomatic Medicine*, *78*(6), 728–739. https://doi.org/10.1097/PSY.0000000000000325

van Kessel, K., Wouldes, T., & Moss-Morris, R. (2016). A New Zealand pilot randomized controlled trial of a web-based interactive self-management programme (MSInvigor8) with and without email support for the treatment of multiple sclerosis fatigue. *Clinical Rehabilitation*, *30*(5), 454–462. https://doi.org/10.1177/0269215515584800

Wiendl, H., & Hohlfeld, R. (2009). Multiple sclerosis therapeutics. *Neurology*, *72*(11), 1008. https://doi.org/10.1212/01.wnl.0000344417.42972.54

Wilson, P. M., Petticrew, M., Calnan, M. W., & Nazareth, I. (2010). Disseminating research findings: What should researchers do? A systematic scoping review of conceptual frameworks. *Implementation Science*, *5*(1), 91. https://doi.org/10.1186/1748-5908-5-91

World Health Organisation. (2008). *Atlas: Multiple sclerosis resources in the world 2008*.

Wrosch, C., & Sabiston, C. M. (2013). Goal adjustment, physical and sedentary activity, and well-being and health among breast cancer survivors. *Psycho-Oncology*, *22*(3), 581–589.

Wrosch, C., Scheier, M. F., & Miller, G. E. (2013). Goal Adjustment Capacities, Subjective Well-being, and Physical Health. *Social and Personality Psychology Compass*, *7*(12), 847–860. https://doi.org/10.1111/spc3.12074

Appendices

Appendix A – Study Advert

Most of us have 'goals' - things that we want to achieve, and we think can be achieved with a bit of effort. However, sometimes we don't make as much progress on our goals as we would like to. Sometimes things get in the way and other times we are not sure what we need to do to make progress. In addition, sometimes we lack motivation because the goals are not clearly linked to what we really value and enjoy.

"Valuing Goals" is an online training programme that helps people to identify and work towards positive personal goals by teaching specific skills step by step. It has been designed by clinical psychologists and based on research that shows goal setting, planning skills and working towards valued goals are associated with high levels of well-being. Previous studies have found this programme to increase well-being and quality of life in the general population as well as in groups of people experiencing lowered well-being. We are now conducting a study to evaluate if the Valuing Goals programme can be as beneficial for people living with MS, and **we are looking for volunteers** to take part in the programme.

If you would like the opportunity to have some online training in how to work towards personal goals, and to take part in some research please go to the link below for further information by clicking it or pasting it into your internet browser **before 31st August 2018.**

http://valuegoals.azurewebsites.net/participant-information/

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ROYAL HOLLOWAY UNIVERSITY OF LONDON

Appendix B - Participant Information Sheet

Department of Psychology: Royal Holloway, University of London
www.royalholloway.ac.uk/psychology

Valuing Goals – Participant information sheet

What is the study about?

"Valuing Goals" is an online training programme that is focused on helping people to identify personal goals and planning actions to achieve them. It is designed by clinical psychologists and based on research that shows goal setting, planning skills and working towards valued goals are associated with high levels of well-being. Previous studies have found this programme to increase well-being and decrease depression in the general population as well as in groups of people who have low levels of well-being. We would like to see if the same results occur for people with MS. To take part you must be an adult with MS, fluent in English, with access to a computer and the internet.

What will it involve?

If you decide to take part, I will ask you to complete the Valuing Goals online programme which will take around 1 hour per week for 5 weeks. I will also ask you to complete some short online questionnaires about your feelings of well-being at 3 different times between July and October 2018 to measure any changes, which will take around 20-30 minutes. As part of the programme you will be asked to identify goals that you would like to achieve, think of steps towards those goals as well as thinking ways round obstacles that might get in the way. It is a self-help programme that you carry out on your own, at times that suit you, but with some contact with me (Emma Britneff). I will telephone you around 1 week after you start the programme to see how you are getting on, and you can also contact me by email or telephone at any point during the study if you have any questions.

To help test whether Valuing Goals works, everyone who is interested will get the programme but to allow us to make comparisons some people will start the training from around July 2018 and a second group will be asked to wait to complete it, and will start from around October 2018. If you decide to take part you will be randomly allocated to one group or the other.

In total, your commitment to the study would be around 7 hours. As a thank you for your time, participants completing the study will be given the chance to enter into a prize draw to win one of two £50 Amazon vouchers.

Will my data be kept private?

Nobody except myself and my supervisor will be allowed to see your personal details and questionnaire data, and in the study you will be known only by a random number. An overall summary of the study's results will be given to your MS centre and to all those who have participated but no individual's data will be shared, so the information is completely confidential. If the study is published in an academic journal, your data will be part of a large group summary and will not be identifiable as referring to you.

Do I have to take part?

You do not have to take part in this study if you don't want to. If you decide to take part you may withdraw at any time without having to give a reason. Your MS centre is kindly supporting this study, but there will be no impact on how you are treated if you decide to take part or not to take part.

How can I find out more?

If you have any questions about the study or want to talk about it before deciding whether or not to take part then please do contact me using the details below. If you have read this information sheet and would like to take part, then please click on the link below to go to the online consent form.

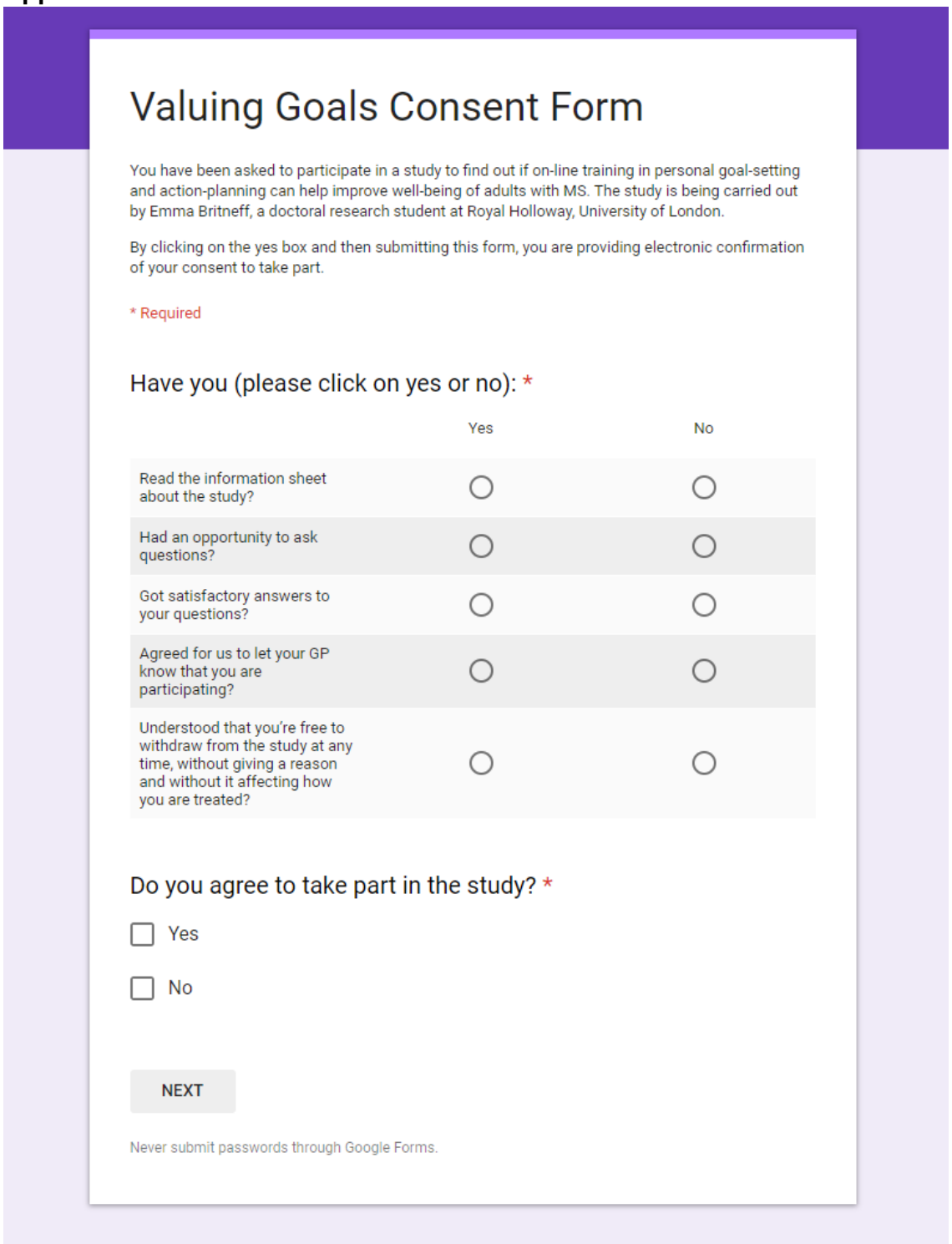
Take me to the online consent form

Contact me

- Email on emma.britneff.2016@live.rhul.ac.uk
- Phone on **01784 414042** (research answer-machine, checked each working day).
- You can also contact my supervisor, Professor Andy MacLeod, on A.MacLeod@rhul.ac.uk or on 01784 414042.

Please print or save this information sheet and keep it for reference.

Appendix C – Online Consent Form



Valuing Goals Consent Form

You have been asked to participate in a study to find out if on-line training in personal goal-setting and action-planning can help improve well-being of adults with MS. The study is being carried out by Emma Britneff, a doctoral research student at Royal Holloway, University of London.

By clicking on the yes box and then submitting this form, you are providing electronic confirmation of your consent to take part.

* Required

Have you (please click on yes or no): *

	Yes	No
Read the information sheet about the study?	○	○
Had an opportunity to ask questions?	○	○
Got satisfactory answers to your questions?	○	○
Agreed for us to let your GP know that you are participating?	○	○
Understood that you're free to withdraw from the study at any time, without giving a reason and without it affecting how you are treated?	○	○

Do you agree to take part in the study? *

☐ Yes

☐ No

NEXT

Never submit passwords through Google Forms.

Valuing Goals Consent Form

* Required

Contact Information

Please also add your name, email address, telephone number, and GP practice below. This allows us to link your consent to your name and provides contact details for us to use when contacting you during the study. This consent form will be stored securely separate from the anonymous information you provide during the study. We will inform your GP that you are taking part in this study but will not share any of your data with them. The only exception to this is if we feel you are at risk of harm, for example experiencing suicidal thoughts, and then we would have a duty to help keep you safe.

Name *

Your answer

Email Address *

Your answer

Telephone Number *

Your answer

Name of GP Practice *

Your answer

BACK SUBMIT

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Appendix D – I-PANAS-SF

Well-being Questionnaires

* Required

Positive and Negative Affect

This scale consists of 10 words that describe different feelings and emotions. Please read each item and indicate the extent you have felt this way over the past week. *

	Very slightly or not at all	A little	Moderately	Quite a lot	Extremely
Upset	○	○	○	○	○
Hostile	○	○	○	○	○
Alert	○	○	○	○	○
Ashamed	○	○	○	○	○
Inspired	○	○	○	○	○
Nervous	○	○	○	○	○
Determined	○	○	○	○	○
Attentive	○	○	○	○	○
Active	○	○	○	○	○
Afraid	○	○	○	○	○

BACK NEXT

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Appendix E – Flourishing Scale

Flourishing Scale

Below are eight statements that you may agree or disagree with. Using the 1 - 7 scale below, indicate your agreement with each item. Please be open and honest in your response.

*

	Strongly disagree	Disagree	Slightly disagree	Neither agree nor disagree	Slightly agree	Agree	Strongly agree
I lead a purposeful and meaningful life	○	○	○	○	○	○	○
My social relationships are supportive and rewarding	○	○	○	○	○	○	○
I am engaged and interested in my daily activities	○	○	○	○	○	○	○
I actively contribute to the happiness and well-being of others	○	○	○	○	○	○	○
I am competent and capable in the activities that are important to me	○	○	○	○	○	○	○
I am a good person and live a good life	○	○	○	○	○	○	○
I am optimistic about my future	○	○	○	○	○	○	○
People respect me	○	○	○	○	○	○	○

Appendix F – FAMS

Functional Assessment of Multiple Sclerosis

Below is a list of statements that other people with MS have said are important. Please mark one response per line to indicate how much it applies to you in the past 7 days.

Mobility *

	Not at all	A little bit	Some-what	Quite a bit	Very much
Because of my physical condition, I have trouble meeting the needs of my family	○	○	○	○	○
I am able to work (include work at home)	○	○	○	○	○
I have trouble walking	○	○	○	○	○
I have to limit my social activity because of my condition	○	○	○	○	○
I have strength in my legs	○	○	○	○	○
I have trouble getting around in public places	○	○	○	○	○
I have to take my condition into account when making plans	○	○	○	○	○

Symptoms *

	Not at all	A little bit	Some-what	Quite a bit	Very much
I have nausea	○	○	○	○	○
I have pain	○	○	○	○	○
I feel ill	○	○	○	○	○
I feel weak all over	○	○	○	○	○
I have pain in my joints	○	○	○	○	○
I am bothered by headaches	○	○	○	○	○
I am bothered by muscle pains	○	○	○	○	○

Emotional Well-being *

	Not at all	A little bit	Some-what	Quite a bit	Very much
I feel sad	○	○	○	○	○
I am losing hope in the fight against my illness	○	○	○	○	○
I am able to enjoy life	○	○	○	○	○
I feel trapped by my condition	○	○	○	○	○
I am depressed about my condition	○	○	○	○	○
I feel useless	○	○	○	○	○
I feel overwhelmed by my condition	○	○	○	○	○

General Contentment *

	Not at all	A little bit	Some-what	Quite a bit	Very much
My work (include work at home) is fulfilling	○	○	○	○	○
I have accepted my illness	○	○	○	○	○
I am enjoying the things I usually do for fun	○	○	○	○	○
I am content with the quality of my life right now	○	○	○	○	○
I am frustrated by my condition	○	○	○	○	○
I feel a sense of purpose in my life	○	○	○	○	○
I feel motivated to do things	○	○	○	○	○

Thinking and Fatigue *

	Not at all	A little bit	Some-what	Quite a bit	Very much
I have a lack of energy	○	○	○	○	○
I feel tired	○	○	○	○	○
I have trouble starting things because I am tired	○	○	○	○	○
I have trouble finishing things because I am tired	○	○	○	○	○
I need to rest during the day	○	○	○	○	○
I have trouble remembering things	○	○	○	○	○
I have trouble concentrating	○	○	○	○	○
My thinking is slower than before	○	○	○	○	○
I have trouble learning new tasks or directions	○	○	○	○	○

Family/Social Well-being *

	Not at all	A little bit	Some-what	Quite a bit	Very much
I feel close to my friends	○	○	○	○	○
I get emotional support from my family	○	○	○	○	○
I get support from my friends	○	○	○	○	○
My family has accepted my illness	○	○	○	○	○
I am satisfied with family communication about my illness	○	○	○	○	○
My family has trouble understanding when my condition gets worse	○	○	○	○	○
I feel "left out" of things	○	○	○	○	○

Appendix H – PHQ9

Patient Health Questionnaire

Over the last 2 weeks, how often have you been bothered by any of the following problems? *

	Not at all	Several days	More than half the days	Nearly every day
Little interest or pleasure in doing things	○	○	○	○
Feeling down, depressed, or hopeless	○	○	○	○
Trouble falling or staying asleep, or sleeping too much	○	○	○	○
Feeling tired or having little energy	○	○	○	○
Poor appetite or overeating	○	○	○	○
Feeling bad about yourself — or that you are a failure or have let yourself or your family down	○	○	○	○
Trouble concentrating on things, such as reading the newspaper or watching television	○	○	○	○
Moving or speaking so slowly that other people could have noticed? Or the opposite — being so fidgety or restless that you have been moving around a lot more than usual	○	○	○	○
Thoughts that you would be better off dead or of hurting yourself in some way	○	○	○	○

Appendix H – Intervention Website

Introduction Timetable Module 1 Module 2 Module 3 Module 4 Module 5 Module 6 Time 1 Questionnaires (G1) Time 2 Questionnaires (G1) Time 3 Questionnaires (G1) Exercise sheets Contact

INTRODUCTION

Welcome to the Valuing Goals Programme. This version of the self-help programme has been developed specifically to help people with MS. You will be asked to identify positive goals that you would like to achieve and to make plans for reaching those goals.

- **'Well-being'** is about how you feel – feeling good and having a positive view of your life.
- **'Goal'** means something you want to achieve and think you can do with a bit of effort. It does not matter how small or 'silly' you think the goal is, or what area of your life it relates to. Any goal that seems positive and important to you matters.
- **'Self-help'** means working on the programme without much support from us. We will check in with you once by phone, but other than that, it is for you to commit to the programme and follow it as much as possible. Feel free to involve friends and family in what you are up to – that can be a good source of motivation.

Have a look at the suggested timetable, then use the menu bar above or the links at the end of each page to navigate through the site.

Appendix I – Keeping up momentum email

Dear [redacted],

It has been roughly three weeks since you began the Valuing Goals Modules. Don't be discouraged if you have lost momentum and not completed the Modules; life can get in the way!

I'll be asking everyone to do their Time 1 Questionnaires in around 3 weeks. If you have lost momentum perhaps now would be a good time to see if you can restart where you left off so that you have a chance to finish before doing the questionnaires.

If you have any questions or problems with the website, please feel free to email or call me anytime.

Good luck!

Kind regards
Emma

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