Curtin School of Population Health

A Collaborative Approach to Designing an Online Nutrition Education Program for People with Multiple Sclerosis

Rebecca Dawn Russell

0000-0002-3554-648X

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Declaration

To the best of my knowledge and belief this thesis contains no material previously published by any other person except where due acknowledgment has been made. This thesis contains no material which has been accepted for the award of any other degree or diploma in any university.

Human Ethics The research presented and reported in this thesis was conducted in accordance with the National Health and Medical Research Council National Statement on Ethical Conduct in Human Research (2007) – updated March 2014. The proposed research study received human research ethics approval from the Curtin University Human Research Ethics Committee (EC00262), Approval Numbers HRE2019-0179 and HRE2020-0484.

Signature:

[signature redacted]

Date: 28th September 2022

Statement from principal supervisor

Rebecca Russell's thesis has been prepared in accordance with the guidelines for a Doctor of Philosophy thesis by publication and I am recommending the thesis now be sent for examination.

Signature:

[signature redacted]

Date: 28th September 2022

Acknowledgement of Country

I acknowledge that Curtin University works across hundreds of traditional lands and custodial groups in Australia, and with First Nations people around the globe. I wish to pay my deepest respects to their ancestors and members of their communities, past, present, and to their emerging leaders. Curtin University’s passion and commitment to work with all Australians and peoples from across the world, including our First Nations peoples, is reflective of the institutions' values and commitment to our roles as leaders in the Reconciliation space in Australia.

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Publications, presentations, awards, and prizes

The following publications, presentations, awards, and prizes were attributed to the work in this thesis. Related publications and presentations that are not central to this thesis are presented in Appendix B: Publications and presentations not central to this thesis.

Peer-reviewed publications (80% Q1)

1. **Russell RD**, Black LJ, Begley A. Nutrition education programs for adults with neurological diseases are lacking: a scoping review. *Nutrients.* 2022;14(8):1577. doi:10.3390/nu14081577.

2. **Russell RD**, Black LJ, Begley A. Navigating dietary advice for multiple sclerosis. *Health Expectations.* 2021;24:853-862. doi:10.1111/hex.13226. (1 early citation, Scopus)

3. **Russell RD**, Black LJ, Begley A. Dietary education programs for adults with neurological diseases: a scoping review protocol. *JBI Evidence Synthesis.* 2021;19(1):853-862. doi:10.11124/JBISRIR-D-19-00394. (1 early citation, Scopus)

4. **Russell RD**, Black LJ, Begley A. The unresolved role of the neurologist in providing dietary advice to people with multiple sclerosis. *Multiple Sclerosis and Related Disorders.* 2020;44:102304. doi:10.1016/j.msard.2020.102304. (5 citations, Scopus)

5. **Russell RD**, Black LJ, Pham NM, Begley A. The effectiveness of emotional wellness programs on mental health outcomes for adults with multiple sclerosis: a systematic review and meta-analysis. *Multiple Sclerosis and Related Disorders.* 2020;44:102171. doi:10.1016/j.msard.2020.102171. (5 citations, Scopus)

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Conference presentations (oral communication)

1. **Russell RD**, Black LJ, Begley A. “Co-designing an online nutrition education program for people with multiple sclerosis”. Paper presented at: Dietitians Australia 2022 Conference; August 14-16, 2022; Adelaide, Australia. doi:10.1111/1747-0080.12758

2. **Russell RD**, Black LJ, Begley A. “Nutrition education programs for adults with neurological diseases are lacking: a scoping review”. Paper presented at: Nutrition Society of Australia, Virtual Annual Scientific Meeting; December 2-3, 2021; Online.

3. **Russell RD**, Black LJ, Begley A. “Navigating dietary advice for multiple sclerosis”. Paper presented at: Nutrition Society of Australia, Virtual Annual Scientific Meeting; December 3-4, 2020; Online. doi:10.3390/proceedings2021072001

Conference presentations (poster communication)

1. **Russell RD**, Black LJ, Begley A. “A collaborative approach to designing an online nutrition education program for people with multiple sclerosis”. Poster presented at: MS Australia Progress in MS Research; April 4-5, 2022; Hobart, Australia.

Grants awarded

1. 2021-2022: MS Australia Incubator Grant (#21-1-072). $24,703. **Russell RD** (CIA), Begley A, Black LJ, Daly A. “Feasibility and efficacy of an online nutrition education program for people with multiple sclerosis”.

Awards and prizes

2022: Premier’s Science Awards: Student Scientist of the Year Finalist

“Awarded to an outstanding postgraduate student who has demonstrated a commitment to science at an early stage and shows great promise in reaching the highest levels of excellence.” https://www.wa.gov.au/organisation/department-of-jobs-tourism-science-and-innovation/premiers-science-awards

ExxonMobil Student Scientist of the Year 2022: Rebecca Russell. https://www.youtube.com/watch?v=cA8phzjLbuc

2021: Best Student Oral Presentation, Nutrition Society of Australia Annual Scientific Meeting

Titled “Nutrition education programs for adults with neurological diseases are lacking: a scoping review.”

2021: Graduate Women of Western Australia (GWWA) Open Scholarship

GWWA scholarships awarded to assist women enrolled in higher degrees by research to enhance the quality of their research and project. The funds were used to enhance the quality of the online nutrition education program, specifically, for professional copyediting, professional videography, and to purchase high-quality images from Shutterstock.

2020: Multiple Sclerosis Western Australia (MSWA) PhD Top-Up Scholarship

2019: Curtin University representative at the Three Minute Thesis (3MT) Asia Pacific Finals (University of Queensland)

2019: Curtin University 3MT winner

Titled "What is a healthy diet anyway? People with multiple sclerosis need a nutrition program."

2019: Australian Government Research Training Program (RTP) Scholarship

Abstract

High-quality diets are important for people with multiple sclerosis (pwMS) to reduce their risk of co-morbidities, including hypertension, dyslipidaemia, and obesity, which have been associated with increased disability progression in multiple sclerosis (MS). High-quality diets can also help to alleviate some of the symptoms of MS, such as fatigue and constipation, by way of adequate nutrients, fibre, and fluids. However, the dietary habits of pwMS are similar to those of the general population whereby the vast majority of Australians do not meet their food group recommendations from the Australian Dietary Guidelines. Furthermore, there are numerous special MS diets (with limited evidence to support their use for pwMS) that are marketed to pwMS with conflicting recommendations and that restrict entire food groups. PwMS need an evidence-based nutrition education program to help them filter through the conflicting information online and improve the quality of their diets with positive health behaviour changes, but no such programs exist in Australia. Nutrition education programs can provide people with the knowledge, skills, and support to achieve high-quality diets and meet the Australian Dietary Guidelines. The best practice principles for effective programs include recognition of co-design, where programs are developed and/or improved in collaboration with consumers. Co-designed nutrition education programs are rated as more acceptable to consumers and result in positive health behaviour changes.

The aim of this thesis was to use a collaborative approach to develop a nutrition education program for pwMS. This aim was fulfilled through four objectives: 1) to identify what nutrition education programs have been implemented for adults with neurological diseases, and the characteristics of those programs; 2) to determine the characteristics of effective education programs for pwMS, namely emotional wellness programs; 3) to explore the perceptions and experiences of neurologists in giving dietary advice and of pwMS in receiving dietary advice; and 4) to develop an evidence-based nutrition education program for pwMS and explore the acceptability and ease of comprehension of one draft module. These objectives were achieved through a series of five manuscripts.

Chapter 1 provides background information on the significance of diet and nutrition education for pwMS and places the contribution of the thesis amongst the identified knowledge gaps in the field.

Chapter 2 details a scoping review that identified what nutrition education programs exist for adults with neurological diseases and examined the characteristics of those programs.

A total of 13 programs were included in the narrative summary and only four programs were for pwMS. There was no evidence of co-design in any of the programs. The most common program characteristics were in-person, group setting, nine hours of total delivery time, six sessions, and one hour per session. The most commonly used behaviour change techniques (BCTs; active components of interventions that support behaviour change) were *instruction on how to perform a behaviour, credible source, behavioural practice/rehearsal, information about health consequences,* and *social comparison.*

Given the scarcity of nutrition education programs for pwMS, it was not possible to examine the characteristics of effective nutrition programs. Therefore, Chapter 3 details a systematic review and meta-analysis that was conducted to explore the characteristics of effective emotional wellness education programs for pwMS. Pooled data from 25 randomised controlled trials (RCTs) and four quasi-experimental studies included 2323 pwMS. The most common program characteristics were in-person, group setting, eight sessions, held weekly, and the duration of sessions ranged from 45 minutes up to three hours. Meta-analyses produced statistically significant results favouring the interventions in relation to waitlist controls, usual care, or another intervention, and the most commonly used BCTs in effective interventions were *behaviour practice/rehearsal, demonstration of the behaviour, social comparison, framing/reframing, social support,* and *goal setting*. The characteristics and BCTs from Chapters 2 and 3 were identified as potentially useful for informing the nutrition education program design.

In Chapter 4, semi-structured interviews were conducted with 11 neurologists to explore their perceptions of the role of diet in MS and what dietary advice they give to their patients. Inductive thematic analysis produced four themes: 1) juggling the evidence on the role of diet in MS; 2) acknowledging the risks and benefits of specific diets; 3) distancing from the diet ‘gurus’; and 4) the unresolved role of the neurologist in providing dietary advice. This chapter highlights the need for an evidence-based nutrition education program that neurologists could refer their patients to, given the neurologists’ self-identified time and knowledge limitations.

In Chapter 5, focus groups were conducted with 33 pwMS plus 1 spouse to explore their experiences when navigating dietary advice, including their attitudes when making dietary decisions and their nutrition education needs. Transcript data were analysed using a general inductive approach and secondary analysis aligned the themes with the constructs of the self-determination theory. Six themes emerged: 1) confusion about where to seek

dietary advice; 2) scepticism towards national dietary guidelines; 3) personalised approaches to dietary change; 4) barriers to dietary changes; 5) judging if dietary changes work; and 6) wanting dietary guidelines for MS. The self-determination theory explained the varying levels of motivation of pwMS to make dietary changes. Building on the characteristics identified in Chapters 2 and 3, the themes from Chapters 4 and 5 were used to develop a framework for a nutrition education program targeted to pwMS.

Chapter 6 details further collaborations with pwMS and MS health professionals and the stages that contributed to developing a full draft program prototype. Stage 1 explored the preferred characteristics and content of a nutrition education program (online survey, n=114 pwMS) and developed a complete nutrition education program. Stage 2 involved revising the potential program with stakeholders (n=10 pwMS and MS health professionals; two meetings) and pwMS (n=6; two online workshops). Stage 3 used cognitive interviews to explore the acceptability and ease of comprehension of one draft module of the program (n=9 (8 pwMS plus 1 spouse). Content analysis of the cognitive interviews produced four themes to direct the program design: 1) positive and targeted messaging to motivate behaviour change; 2) “not enough evidence” is not good enough; 3) expert advice builds in credibility; and 4) engaging and appropriate online design elements are crucial.

This body of work has produced fundamental knowledge regarding the need and characteristics of a nutrition education program for pwMS in Australia and reinforces the importance of using co-design for end-user acceptability. This thesis has also identified content, language, design, and format considerations of an online program that may ultimately improve the motivation of pwMS to make evidence-based, healthy dietary changes. The publications included in this thesis have also identified a theoretical framework and BCTs that may be useful for supporting dietary behaviour change among pwMS and should be considered in development of future nutrition education programs. At the time of writing this thesis, a feasibility study is underway to test the complete prototype nutrition education program with 75 pwMS (Australian New Zealand Clinical Trials Registry ACTRN12622000276752; MS Australia Incubator Grant #21-1-072).

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List of abbreviations

3MT:	Three Minute Thesis
AIMS:	Arthritis Impact Measurement Scales
BCT:	Behaviour Change Technique
BCTs:	Behaviour Change Techniques
BDI:	Beck Depression Inventory
BDI-II:	Beck Depression Inventory II
CCIP:	Consumer and Community Involvement Program
CEOS:	Context, Executive and Operational Systems
CES-D:	Center for Epidemiologic Studies Depression Scale
CG:	Comparator Group
CI:	Confidence Interval
CIS:	Clinically Isolated Syndrome
COREQ:	Consolidated Criteria for Reporting Qualitative Research
DASS-21:	Depression, Anxiety and Stress Scales
DMT:	Disease Modifying Therapy
EQ-5D:	EuroQol
GWWA:	Graduate Women Western Australia
HADS:	Hospital Anxiety and Depression Scale
HAQUAMS:	Hamburg Quality of Life Questionnaire in Multiple Sclerosis
HPLP-II:	Health-Promoting Lifestyle Profile-II
IG:	Intervention Group
IQR:	Interquartile Range
IV:	Inverse Variance
JBI SUMARI:	Joanna Briggs Institute System for the Unified Management, Assessment and Review of Information
JBI:	Joanna Briggs Institute
LiSat-9:	Life Satisfaction Questionnaire
MeSH:	Medical Subject Headings
MHI-18:	Mental Health Inventory
MS:	Multiple Sclerosis
MSQOL-54:	Multiple Sclerosis Quality of Life-54
MSWA:	Multiple Sclerosis Western Australia
NR:	Not Reported

PHQ-9:	Patient Health Questionnaire
PPMS:	Primary Progressive Multiple Sclerosis
PQOLC:	Profile of Health-Related Quality of Life in Chronic Disorders
PRISMA-ScR:	Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for Scoping Reviews
PROMIS:	Patient-Reported Outcomes Measurement Information System
PSS:	Perceived Stress Scale
PwMS:	People with Multiple Sclerosis
QoL:	Quality of life
RCT:	Randomised Controlled Trial
RCTs:	Randomised Controlled Trials
RRMS:	Relapsing-remitting Multiple Sclerosis
RTP:	Research Training Program
SD:	Standard Deviation
SDT:	Self-Determination Theory
SF-12:	Medical Outcomes 12-Item Short-Form Health Survey
SF-36:	Medical Outcomes 36-Item Short-Form Health Survey
SMD:	Standardised Mean Difference
SMDs:	Standardised Mean Differences
SPMS:	Secondary Progressive Multiple Sclerosis
SRQR:	Standards for Reporting Qualitative Research
STAI:	State-Trait Anxiety Inventory
TAFE:	Technical and Further Education

Stakeholder advisory group

At the beginning of this PhD project in 2019, I engaged the Consumer and Community Involvement Program (CCIP) to establish a project stakeholder advisory group. The CCIP is an activity of the Western Australian Health Translation Network, to support consumers, community members, and researchers to work in partnership to improve health outcomes and ensure that community involvement in research is standard practice.[1] The purpose of the stakeholder advisory group was to contribute consumer feedback and professional knowledge essential to the project. The stakeholder advisory group contributed their personal and professional experiences at key stages throughout my PhD project while developing a nutrition education program, to ensure that the values, needs, and practices of people with multiple sclerosis (pwMS) were appropriately considered.

The CCIP advertised two vacant consumer positions on their website, in their e-newsletter, and via social media. Interested consumers submitted an expression of interest via the CCIP website. Mr Ben Horgan, a CCIP Consumer Advocate, telephoned each applicant to discuss their background, including experience on committees or steering groups, current community group connections, and their motivations for being involved in the stakeholder advisory group. Mr Horgan phoned the referees of the applicants to enquire about their communication skills and other strengths that would be valuable to the group dynamic, and he generated a summary report of shortlisted applicants for my consideration. I selected the two pwMS based on their community connections and the diversity of experience that they would potentially bring to the group. The health professional positions were advertised by Multiple Sclerosis Western Australia (MSWA, a primary service and support provider for people living with neurological conditions in Western Australia (WA)) to their staff via email. Health professionals emailed their expressions of interest to me, which included a brief statement on their background, number of years working with pwMS, their interest in diet and multiple sclerosis (MS), and why they wanted to be involved in the stakeholder advisory group. I also invited a prominent WA neurologist who specialises in MS to join the group.

As of August 2022, the members of the project stakeholder advisory group are:

- Rachel Burns (person with MS)

[1] For further information about the Community and Consumer Involvement Program, see: https://cciprogram.org/

- Tracey Tasker (person with MS)
- Professor Allan Kermode (neurologist)
- Katie Pound (dietitian with experience working with pwMS)
- Julie Chandler (MS nurse)
- Michaela Mundy (MS counsellor)
- Dr Lisa Grech (clinical psychologist, research academic, and person with MS)
- Misty Reinkowsky (MS dietitian, joined the group in 2021)
- Jodie Roberts (MS dietitian, joined the group in 2021)
- Gemma Toovey (MS dietitian, joined the group in 2021)

The group was tasked with reviewing the design and language of the participant information sheets and consent forms, the proposed question guides for the qualitative phase of this project, the topics and content in a draft version of the nutrition education program, the proposed program activities and discussion board topics, and the logistics of a proposed feasibility study to test a prototype program, including recruitment considerations. Six meetings were held throughout this PhD project. The two MS consumers were paid $35 per hour, in line with the CCIP Honorarium Guidelines.

Thesis outline

This mixed-methods thesis contains seven chapters: background and methodology (Chapter 1), four peer-reviewed papers that have been published (Chapters 3-5), one manuscript that has been submitted for peer-review (Chapter 6), and a discussion and conclusion (Chapter 7).

Chapter 1: Background

This chapter begins with a brief overview of nutrition education and the best practice principles for effective nutrition education programs. These principles ensure that the literacy, visual design, activities, underlying theories, and behaviour change techniques (BCTs, active components of interventions that support behaviour change, e.g., *goal setting*) are suitable for the target audience, with increasing recognition of co-design. The chapter outlines the overall significance of using co-design principles when developing a nutrition education program for people with multiple sclerosis (pwMS), given that programs that have been developed using principles of co-design are more acceptable and result in positive health behaviour changes.

Chapter 1 then provides background information on multiple sclerosis (MS) and the role that diet can play as a modifiable lifestyle factor in MS. Dietary modifications can affect the risk of some co-morbidities that are more common in pwMS, such as cardiovascular disease, hypertension, dyslipidaemia, obesity, type 2 diabetes, and osteoporosis, and some common symptoms of MS, including fatigue, constipation, bladder dysfunction, and depression. This chapter details the current dietary recommendations for pwMS in Australia concerning those co-morbidities and symptoms, and the role that nutrition education can play in helping pwMS achieve the recommendations. The dietary intakes of pwMS are similar to the general population, in that the majority (>95%) of Australians do not meet the food group recommendations in the Australian Dietary Guidelines; hence, nutrition education programs can help people improve their diet quality and achieve the recommendations. Therefore, this chapter reiterates the importance of nutrition education programs for pwMS.

Prior to this thesis, it was not known **what nutrition education programs had been implemented for pwMS,** including if there was **any evidence of consumer involvement or co-design in the development of such programs,** or the **characteristics of effective education programs for pwMS**. It was also unclear **what dietary advice pwMS receive**

from their neurologists (their preferred and trusted source of health information) and **how pwMS respond to the advice they receive about diet and MS.** This introductory chapter highlights the knowledge gaps in the field that are filled by this PhD project and justifies the need for an evidence-based nutrition education program for pwMS that has been developed using a collaborative approach with MS consumers and stakeholders.

Chapter 2: Methodology

Chapter 2 defines the author's personal context and epistemology, the study design presented in the overall thesis, and the methods used in each of the individual research chapters.

Chapter 3: Nutrition education programs for adults with neurological diseases

The first stage of this PhD project was to map out what nutrition education programs currently exist for adults with neurological diseases and to examine the characteristics of those programs. To fill this knowledge gap, I conducted a scoping review according to a published *a priori* protocol (Appendix D: Published scoping review protocol). The findings from this review revealed that a lack of programs exist, with only 13 programs included – of which only four programs were for pwMS.

None of the programs appeared to adhere to the best practice principles for nutrition education programs, and there was no evidence of co-design. The program characteristics varied widely, but the most common program characteristics were in-person, group setting, the total delivery time of nine hours (median), six sessions (median), and one hour per session (median). Only one program for pwMS reported using a theoretical framework. I coded behaviour change techniques (BCTs) using a 93-item taxonomy to identify BCTs that may be suitable for the MS nutrition education program that was developed for this PhD project. The most commonly used BCTs were *instruction on how to perform a behaviour, credible source, behavioural practice/rehearsal, information about health consequences,* and *social comparison.*

Chapter 2 justifies the need to first identify characteristics of effective education programs for the MS population (Chapter 3), and then to develop a nutrition education program for pwMS that aligns with best practice principles, including collaborating with MS consumers and other stakeholders.

Chapter 4: Effective emotional wellness programs for adults with multiple sclerosis

The plausible next step would have been to examine the characteristics of effective nutrition education programs for pwMS using a systematic review and meta-analysis. However, given the scarcity of programs as revealed in Chapter 2, I was unable to conduct such a review. Therefore, the purpose of this chapter was to explore the characteristics of effective wellness education programs for pwMS to inform the design of the nutrition education program. The United States National MS Society has defined three domains of wellness for MS: diet, exercise, and emotional wellness. Exercise programs and the characteristics of effective programs have been extensively published in the MS literature; therefore, in Chapter 2 I explored emotional wellness programs for pwMS.

I conducted a systematic review and meta-analysis to determine the effectiveness of emotional wellness programs on mental health outcomes (depression, anxiety, quality of life, and stress) for adults with MS, and to assess the BCTs used in effective programs. The most common characteristics of the programs were: in-person, weekly, in a group setting, and eight sessions. The duration of sessions ranges from 45 minutes up to three hours. The most commonly used BCTs in effective interventions were *behaviour practice/rehearsal, demonstration of the behaviour, social comparison, framing/reframing, social support,* and *goal setting*.

The findings from Chapters 2 and 3 identified characteristics and BCTs that may be suitable for a nutrition education program for pwMS.

Chapter 5: The perceived role of neurologists in providing dietary advice to people with multiple sclerosis

PwMS prefer to receive health information from their neurologists but prior to this PhD project, it was not known how neurologists perceive the role of diet in MS, that is, if they consider diet to be important or not, and what dietary advice they give to their patients with MS. These gaps in knowledge were important to contextualise during the development of a nutrition education program for this PhD project, as neurologists are the most trusted and preferred source of health information for pwMS. To fill these knowledge gaps, I interviewed 11 neurologists who were diagnosing and/or treating pwMS in Western Australia (WA).

The interview discussions revealed that the lack of data from randomised controlled trials made it difficult for neurologists to juggle the evidence on the role that diet plays in MS. The neurologists very rarely acknowledged any potential effects of dietary changes on

symptoms of MS or co-morbidities, except for weight loss. The dietary advice that they provided to their patients ranged from no advice at all, to counselling about specific diets; often based on anecdotes from their other patients or their personal experiences. None of the neurologists specifically mentioned the Australian Dietary Guidelines. Despite acknowledging their limitations in providing dietary advice, including lack of knowledge and time, the majority of the neurologists did not refer their patients to dietitians.

These interviews provided me with an understanding of what dietary advice pwMS may receive from their neurologist, which was not always evidence-based, as well as some insights into the reasons underpinning such advice. This chapter justifies the need for an evidence-based nutrition education program that neurologists could refer their patients to.

Chapter 6: How people with multiple sclerosis navigate dietary advice

In the same time period while interviewing neurologists, I also conducted focus groups with pwMS to explore their experiences when navigating dietary advice, including their attitudes when making dietary decisions and their nutrition education needs. The focus groups revealed that pwMS were confused about where to seek dietary advice and how to judge the reliability of information about diet, but they were highly motivated to learn about dietary changes that could help them.

The focus group participants were wary of general healthy eating advice and were sceptical about the suitability of the Australian Dietary Guidelines. They wanted to be told what to eat for their MS by way of MS-specific dietary guidelines that they could tailor to their own needs. The focus group discussions explored barriers to making healthy dietary changes, and ways that pwMS overcome those barriers. These barriers and solutions along with the overarching themes were incorporated into the developing nutrition education program, namely: confusion about where to seek dietary advice (and how to judge the credibility of information); scepticism towards national dietary guidelines; barriers to dietary changes; and wanting dietary guidelines that are tailored for MS.

The lens of the self-determination theory was applied as a secondary analysis of the focus group transcript data. This theory explained the varying degrees of motivation among pwMS for making dietary changes. This theory formed the underlying theoretical basis for the developing nutrition education program, where the content and activities would leverage the types of motivation (external and intrinsic) to support and drive dietary behaviour change in pwMS.

Building on the characteristics identified in Chapters 2 and 3, the qualitative findings from Chapters 4 and 5 were used to develop a framework for a nutrition education program.

Chapter 7: Using co-design principles to develop an online nutrition education program for people with multiple sclerosis

This final research chapter details how I continued to collaborate with pwMS and MS health professionals to develop a complete program prototype and then test the acceptability and ease of comprehension of one draft module of the prototype program. The chapter begins by detailing how the findings from the previous chapters and the relevant literature were used to iteratively develop the framework for the draft nutrition education program.

This chapter has three distinct stages. Stage 1 involved an online survey to explore the interest and preferred content and characteristics of a nutrition program (n=114 pwMS). A draft of a nutrition program was created by building on the program framework with the survey findings, best practice recommendations for nutrition interventions, and input from the project stakeholder advisory Group. Stage 2 described how the draft program was revised based on from feedback from the stakeholder advisory group in two meetings and six pwMS in two workshops to produce a full program prototype. Stage 3 details how one module was produced in an online format by an instructional designer (whom I sourced and briefed on the project, including preferred design concepts and style). I used cognitive interviews to explore the acceptability and ease of comprehension of the module (n=9 (8 pwMS plus 1 spouse)). Content analysis from the cognitive interviews produced themes that highlighted the key elements that influenced acceptability and/or ease of comprehension. The main findings were that: positive and targeted messaging motivates behaviour change; phrases that are perceived as negative and demotivating, such as “not enough evidence”, should be avoided; expert advice, including pwMS as experts, provides credibility; and engaging and appropriate online design elements are crucial.

This chapter ties together the findings from the previous chapters and highlights the importance of collaborating with consumers and stakeholders at multiple stages during the development of a nutrition education program. It reiterates the importance of using co-design principles by revealing key insights into factors that could affect the acceptability of a program. These factors included language and visual design elements, as these appeared to influence the engagement with the online module and may ultimately influence the motivation to make evidence-based, healthy dietary changes.

Chapter 8: Discussion

The final chapter of this thesis summarises and discusses the relevance of each of the chapters in informing the development of the prototype nutrition education program. The overall significance of using co-design principles when developing a nutrition education program for pwMS is highlighted. This chapter also contains the implications for future research, including outlining the 2021-2022 MS Australia-funded feasibility study to test the program prototype nutrition education program (Australian New Zealand Clinical Trials Registry ACTRN12622000276752).

Chapter 1. Background

1.1 Nutrition education

Nutrition education involves a series of educational strategies that aim to improve nutrition-related behaviours and dietary intakes that are conducive to health and wellbeing.[1] These programs have been shown to be effective at improving nutrition-related knowledge and behaviours in the general population[2] and are recommended by the World Health Organization to promote healthy diets in line with national dietary guidelines to reduce the risk of diet-related chronic diseases.[3]

People with multiple sclerosis (pwMS) are interested in learning about nutrition[4] and they perceive that various dietary factors can influence the severity of their symptoms or their disease activity.[5] However, evidence suggests that the overall dietary habits of pwMS are either similar to the general population[6-8] or less nutrient-dense,[9] whereby more than 96% of Australians do not meet their food group recommendations from the Australian Dietary Guidelines.[10] This highlights the need for an evidence-based nutrition education program that has significant scope to improve the dietary habits of pwMS, and thus improve quality of life through dietary management of some symptoms. **However, prior to this PhD project, it was unclear what nutrition education programs existed for pwMS.**

1.1.1 Best practice in nutrition education

Nutrition education programs that are implemented using best practice principles, through linking research, theory, and practice, are more likely to be effective at achieving their outcomes.[11] To promote effective nutrition education programs, there are 28 recognised best practices organised into five domains (Table 1.1).[12] Under the program design domain, it is recognised that an appropriate theoretical basis should be used for the program development, which would guide the constructs of the behaviour change techniques (BCTs, active components of interventions that support behaviour change, e.g., goal setting)[13] that are used in the program.

Table 1.1: The best practice principles in nutrition education under the five key domains, adapted from Baker et al[12] (2020).

Domain	Examples of best practices principles
Program design	Evidence-based content
	Theoretical basis
	Design appropriate for the target audience
	Considers the literacy of the target audience
	Goal setting and objectives
	Recognition for co-designed programs to ensure visuals, language, activities, and BCTs[13] are suitable for the target audience[14]
Program delivery	Experiential activities
	Learning styles
	Program fidelity
Educator characteristics	Expert in the content area
	Expertise in the teaching methods
	Relate to the target audience
Educator training	Initial training
	Ongoing training
Evaluation	Formative, process, and outcome evaluations
	Assess the program impact
	Sustained behaviour change

BCTs: behaviour change techniques.

There is an increasing recognition for nutrition education programs to be developed using co-design.[14] Co-design is defined as “meaningful end-user engagement in research design and includes instances of engagement that occur across all stages of the research process and range in intensity from relatively passive to highly active and involved.”[15(pp2-3)] Hence, co-design can be achieved by using participatory research methods and collaborating with consumers.[16] Co-design can ensure that the visuals, language, activities, underlying theories, and BCTs are appropriate for the audience,[12] and the lived experience of consumers is captured, to produce programs that meet their needs and considers their beliefs, values, and practices.[16-18] Nutrition interventions that have been co-designed with consumers and stakeholders are reported as being more acceptable and result in positive health behaviour change.[14] Hence, the recommendation for nutrition

interventions to adopt this “bottom-up”, consumer-focussed approach.[14] **Prior to this PhD project, evidence of consumer and stakeholder involvement in the development of nutrition education programs for pwMS was unknown.** Furthermore, given that it was unclear prior to this PhD project what nutrition education programs existed for pwMS, the **characteristics of effective nutrition education programs for pwMS, including what BCTs have been used, was also unknown**.

1.2 Multiple sclerosis

Multiple sclerosis (MS) is an immune-mediated disease of the central nervous system.[19, 20] Inflammation, scarring, and demyelination of the nerve cell sheaths disrupt the transmission of nerve signals, which can affect any of the visual, sensory, or motor systems.[21] As a result, the symptoms can range widely, and may include optic neuritis, vertigo, fatigue, bladder and bowel dysfunction, pain, or disrupted motor control such as spasticity or limb numbness.[22] The average age of diagnosis globally is around 30 years,[20, 23] and there are at least twice as many females with MS compared to males (69% and 31%, respectively), although, those statistics vary with global regions, and in the Western Pacific region, 78% of pwMS are female.[23] MS is the most common cause of non-traumatic disability in young adults[24], affecting an estimated 2.8 million people globally, an increase from the 2013 estimate of 2.3 million,[23] although this is likely to be an underestimate.[25] In Australia in 2017, over 25,000 people had MS, with an estimated economic cost of $1.75 billion per annum.[21]

There are four clinical course descriptions (phenotypes) of MS: relapsing-remitting MS (RRMS), secondary progressive MS (SPMS), primary progressive MS (PPMS) (Figure 1.1), and clinically isolated syndrome (CIS). A small proportion of people are diagnosed with CIS, which is characterised as a single, inflammatory demyelinating event, followed by partial or complete recovery. If a second demyelinating event is experienced or evidence of lesions on magnetic resonance imaging are identified, the person will be diagnosed with one of the other MS phenotypes.[26] The most common phenotype is RRMS which accounts for around 80% of cases. RRMS is characterised by a sudden onset of symptoms (a relapse), followed by periods of remission where symptoms resolve, and there is a gradual accumulation of disability over time.[27] In 15% to 30% of cases, RRMS develops into SPMS, where there is an ongoing accumulation of disability and no periods of remission in the symptoms. Approximately 15% of people are diagnosed with PPMS at the onset: a phenotype similar to SPMS, where there are no periods of remission, but in contrast, disease and disability progression is continual from the onset.[25]

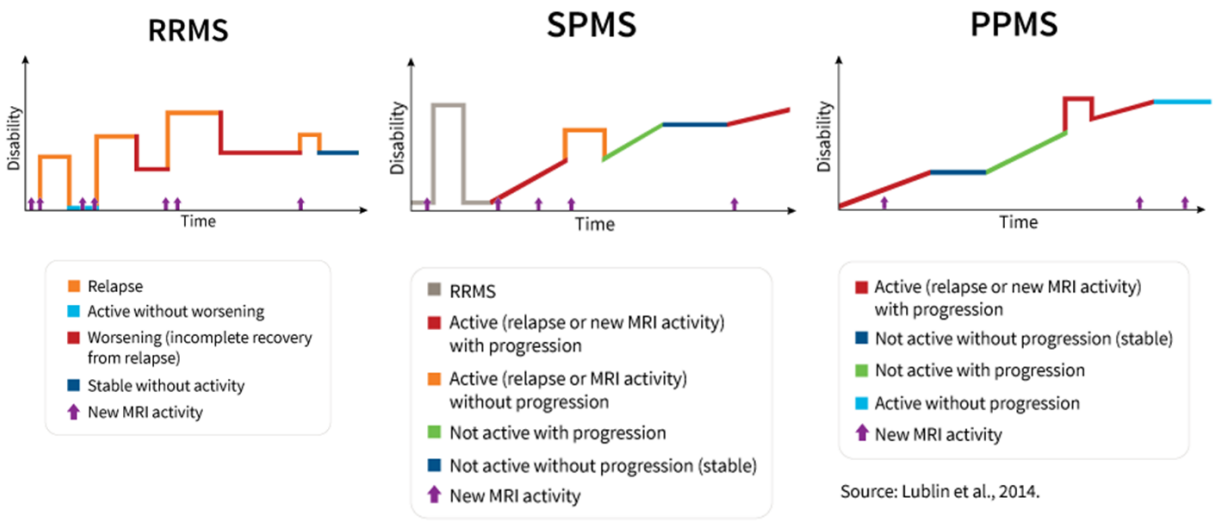

Figure 1.1: Three main phenotypes of multiple sclerosis, graphically represented by disability accumulation over time, relapses, and new magnetic resonance imaging (MRI) activity. Images from the National MS Society[28] (data sourced from Lublin et al., 2014[26]).

Currently, there is no known cure for MS, though disease-modifying therapies (DMTs) are becoming more effective at slowing the progression of the disease and reducing or preventing the accumulation of disability.[29] However, newer DMTs with higher efficacy in reducing disease activity are often accompanied by undesirable side effects due to the greater levels of immunosuppression, including flu-like symptoms, gastrointestinal issues, elevated liver enzymes, headaches, hair loss, and chest pains.[29] These adverse side effects impede adherence to treatment.[30] Despite the lack of evidence of efficacy, some pwMS use complementary and alternative medicines, including dietary modifications, to help manage their disease either in conjunction with their DMTs[31] or as an alternative to taking a DMT.[32]

1.3 Diet as a modifiable lifestyle factor in MS

1.3.1 Dietary recommendations for people with MS

Currently, there is not enough high-quality empirical evidence to support any specific therapeutic diet for MS.[33, 34] However, emerging evidence supports diet as a modifiable lifestyle factor that may influence disability and disease progression.[35, 36] A healthy, balanced diet is considered complementary to the use of DMTs for more efficient management of MS.[36] Due to the recognition that diet may play a role in the pathogenesis and disease course of MS,[37] maintaining or adopting a healthy diet has been recognised as a priority by MS Australia in their 'Roadmap to Defeat MS in Australia'. As such, MS

organisations in Australia advise pwMS to follow the Australian Dietary Guidelines as an example of a healthy dietary pattern.[35, 38, 39]

The Australian Dietary Guidelines, published by the National Health and Medical Research Council, provide evidence-based recommendations on the amounts and kinds of foods to eat for health and wellbeing.[40] Following the dietary advice in the Australian Dietary Guidelines can ensure sufficient nutrient intake and a reduced risk of chronic diseases, including cardiovascular disease, type 2 diabetes, obesity, and some cancers.[40] There are five principal guidelines, quoted here from the Australian Dietary Guidelines:

1) To achieve and maintain a healthy weight, be physically active and choose amounts of nutritious food and drinks to meet your energy needs

2) Enjoy a wide variety of nutritious foods from these five groups every day

3) Limit intake of foods containing saturated fat, added salt, added sugars and alcohol

4) Encourage, support, and promote breastfeeding

5) Care for your food; prepare and store it safely[40]

The Australian Guide to Healthy Eating is a visual representation of the food group recommendations within the Australian Dietary Guidelines, showing the recommended proportions of each group that should be consumed, along with some examples of foods within each of the food groups (Figure 1.2).[40]

Figure 1.2: The Australian Guide to Healthy Eating food selection guide, showing the proportions to eat from each of the food groups every day from the Australian Dietary Guidelines.[40]

1.3.2 Diet-related co-morbidities and MS

Adherence to a dietary pattern that aligns with the Australian Dietary Guidelines can reduce the risk of diet-related co-morbidities, including vascular co-morbidities (hypertension and dyslipidaemia), obesity, and osteoporosis.[40] Reducing the risk of these co-morbid diseases is especially important for pwMS, as multiple co-morbidities have been associated with greater odds of 30-day hospital readmission[41] and poorer quality of life in pwMS.[42] Vascular co-morbidities have been associated with increased disability progression in pwMS,[43] obesity in MS has been associated with increased pain, fatigue, and disability progression,[42, 44, 45] higher risk of relapse,[46] hypertension, and type 2 diabetes,[42, 47] and pwMS are at a higher risk of fractures from falls due to osteoporosis.[48] The prevalence of these co-morbidities in the MS population is similar to, and/or greater than, the prevalence in the general population.[45, 49-51]

1.3.3 Diet and MS symptoms

Symptom management is a major area that can influence the quality of life for pwMS.[52] PwMS who consume a higher quality diet (indicating greater adherence to the Australian Dietary Guidelines) have reported a lower severity in some symptoms, including depression, pain, bowel, vision, and cognitive symptoms[53] (compared to pwMS who had a lower quality diet). While people with less symptom severity may find it easier to consume a higher-quality diet, as opposed to a higher-quality diet reducing the impact of the MS symptoms, this potential bias in the reporting does not detract from the other potential benefits from adhering to dietary patterns in line with the Australian Dietary Guidelines, as previously described in this chapter. From a medical nutrition perspective, adherence to the Australian Dietary Guidelines may provide a reduction in some of the common symptoms of MS, by way of adequate nutrient, fibre, and fluid intake and maintaining a healthy weight.[40] Such symptoms include fatigue, constipation, bladder dysfunction, and depression (Table 1.2).

Table 1.2: Prevalence and nutritional considerations for some common symptoms of MS

	Prevalence in pwMS	Nutritional considerations
Fatigue	Up to 90%[54]	May be exacerbated by an inadequate energy/kilojoule intake, dehydration, and deficiency of micronutrients that are involved in energy generation, such as folate, iron, and vitamin B12.[52, 55] Excess body weight may also exacerbate fatigue in pwMS.[52]
Constipation	50%[56]	May reduce by consuming enough fibre and fluids, and eating frequently.[52]
Bladder issues, such as urgency and incontinence	70-80%[57, 58]	Limiting alcohol, spicy foods, citrus fruits, carbonated beverages, and caffeine, and adequate hydration (dehydration can increase the risk of urinary tract infections and irritate a neurogenic bladder), may ease bladder issues.[52, 57]
Depression	31%[59]	Higher intakes of fruits and vegetables correlated with lower depression and better mental health in three large cross-sectional studies of nearly 6000 pwMS.[8, 9, 60] Randomised controlled trial evidence has shown that adherence to a dietary pattern that is compliant with the Australian Dietary

	Guidelines (the Mod*i*MedDiet) statistically reduced depressive symptoms in people with clinically diagnosed depression from the general population.[61]

MS, multiple sclerosis; pwMS, people with MS.

1.4 Special diets that are marketed to people with MS

Despite the dietary recommendations for pwMS to follow the national dietary guidelines, numerous special MS diets with limited evidence to support their use in MS are marketed to pwMS.[62] Many of the diets are contradictory in the lists of foods that they encourage or restrict,[62] and some of the diets eliminate entire food groups from the Australian Dietary Guidelines (Table 1.3). Such restrictive diets may put pwMS at risk of nutritional deficiencies, such as calcium, vitamin B12, and iron, depending on the food group that they restrict; nutrients that are especially important for pwMS. Insufficient calcium intake is one factor that can contribute to osteoporosis, of which there is a higher prevalence in older adults with MS (13%, compared to 8% in the general population).[49, 50] PwMS have a higher tendency of falls and fractures due to issues with spasticity, spatial awareness, and other motor functioning deficits in MS, even in early diagnosis.[48, 63] Vitamin B12 is used by nerve cells as a component of the myelin sheaths that enhance nerve transmission. Some symptoms of vitamin B12 deficiency are similar to those of MS, which can exacerbate MS symptoms or even accelerate demyelination and/or slow myelin repair in pwMS.[64, 65] Many of the B group vitamins and iron are important nutrients for energy generation[66]; hence, deficiencies could plausibly exacerbate MS-related fatigue.

Table 1.3: Comparison of the Australian Dietary Guidelines[40] and the diets marketed to pwMS by food groups (and associated key nutrients).

	Australian Dietary Guidelines [40]	Swank Diet [a]	Overcoming MS Diet [b]	McDougall Program [c]	Wahls Protocol [d]	Autoimmune Protocol [e]	Best Bet Diet [f]
Vegetables Key nutrients: carbohydrates, vitamin A, vitamin C, folate, and fibre.	✓	✓	✓	✓	✓	✓	✓
Fruit Key nutrients: carbohydrates, vitamin C and fibre.	✓	✓	✓	✓	✓	✓	✓
Grain (cereal) foods Key nutrients: carbohydrates, protein, iron, fibre, B group vitamins, vitamin E, zinc, and magnesium.	✓	✓	✓	✓	✗	✗	Gluten-free only
Meat and poultry Protein, iron, zinc, and vitamin B12.	✓	Limit	✗	✗	✓	✓	Limit
Fish and seafood Protein, iodine, zinc, selenium, vitamin A, and omega-3 fatty acids.	✓	Limit oily	✓	✗	✓	✓	✓
Legumes/beans Protein, iron, zinc, some essential fatty acids, and fibre.	✓	✓	✓	✓	✗	✗	✗
Eggs Protein, vitamin B12, and choline.	✓	White only	White only	✗	✗	✗	Limit
Dairy Protein, calcium, iodine, B group vitamins, zinc, and vitamin B12	✓	Low-fat only	✗	✗	✗	✗	✗

MS: multiple sclerosis; pwMS: people with multiple sclerosis.
[a]**Swank diet:** low-fat, limits saturated fat to <15g/day, and restricts red meat.[67]
[b]**Overcoming MS diet:** plant-based, excludes animal products except for seafood and egg whites.[68(p284)]
[c]**McDougall Program:** a low-fat, vegan diet, and excludes all oils, white flour, white rice, chocolate, and coffee.[69]
[d]**Wahls Protocol:** based on a paleolithic diet - high in meat and saturated fat, excludes dairy, legumes, grains, and sugar.[70]
[e]**Autoimmune Protocol:** strict elimination phase (excludes grains, legumes, nightshade vegetables, eggs, dairy, nuts, seeds, alcohol, coffee, and refined/processed sugars) and foods are reintroduced after a set period.[71]
[f]**Best Bet diet:** based on seafood, game meats, and vegetables, and restricts red meat, eggs, dairy, gluten-containing foods, legumes, and soy.[72]

Given the abundance of conflicting nutrition advice on the internet,[62] often with a readability level above the educational level attained by pwMS,[73] pwMS find it difficult to

work out what information is credible or trustworthy.[74-76] The potential risks from following diets that are marketed to pwMS should not be discounted, such as potential nutrient deficiencies,[77, 78] social isolation due to difficulty maintaining the diet when eating out of the home,[74] and possible mental health repercussions if the diet expectations are not achieved (i.e., improvement in symptoms, reversal of damage from existing lesions, and/or halting MS progression).[74] There is a need to promote, and educate pwMS about, the likely benefits of following the Australian Dietary Guidelines, and to provide nutrition education that empowers them with the skills to assess the credibility of the information that they read. However, **prior to this PhD project, there was very little known about how pwMS respond to the nutrition information they retrieve, including their attitudes when making dietary decisions and their nutrition education needs**.

1.5 Dietary changes made by people with MS

The have been very few studies that have reported on the dietary changes made by pwMS in Australia. Around 40% of Australians with MS reported making dietary modifications within one year of a first clinical diagnosis of a demyelinating event, a common precursor to MS, (total n=244).[79] The majority (94%) of pwMS living in Australia have reported making an effort to eat healthily, and around 21% were currently following a particular diet (total n=1490 pwMS; over 70% had been diagnosed with MS 10+ years prior).[53] The proportion of pwMS outside of Australia who have reported making dietary changes ranges from 12% in Italy (total n=435; mean disease duration 10 years),[4] to 42% in Germany (total n=337; mean disease duration 7 years),[80] and 45% in the United States (total n=6990; mean disease duration 19 years).[7] Reasons for making dietary changes included overall well-being/general health, weight loss,[7] slow disease progression, reducing relapses,[80] feeling in control of their disease, improving their symptoms, and curing their MS.[74]

Some of the dietary changes described in the literature were in line with the Australian Dietary Guidelines advice, for example, eating more fruits and/or vegetables, eating less discretionary foods,[79] consuming less added sugar,[7, 79] reducing saturated fat intake,[4, 80] and eating less meat and more fish.[80] However, not all changes reported were in line with the guidelines, including eliminating dairy and/or meat,[53, 80] following low-carbohydrate diets, low-calorie diets, and adhering to weight loss-plan diets (such as Jenny Craig or Nutrisystem).[7]

1.6 Sources of dietary advice

While physicians remain the preferred, trusted, and most important source of health information for pwMS,[75, 76, 81-83] the majority of pwMS seek out information about diet and MS online[75]; hence, the internet is commonly used as a source of information.[81, 84] There can be hesitancy amongst pwMS to discuss the information that they retrieve online with their neurologists,[84] which is concerning given the low readability/high educational level required to understand MS-related websites.[73] However, **prior to this PhD project, it was not known how neurologists perceived the role of diet in MS** (i.e., if they consider diet to be important or not for pwMS) and **what dietary advice they gave to their patients with MS**. These gaps in knowledge were important to contextualise during the development of a nutrition education program for this thesis, given the degree of trust and preference for neurologists as a source of health advice for pwMS.

1.7 Aim and objectives

The aim of this thesis was to use a collaborative approach to develop a nutrition education program for people with multiple sclerosis (pwMS). To ensure that the program addressed the beliefs and behaviours of pwMS and that the content and characteristics suited their preferences, it was imperative to answer the following questions:

1. What nutrition education programs currently exist for pwMS?
2. What are the characteristics of effective nutrition education programs for pwMS?
3. What dietary advice is provided to pwMS by their neurologists, and how do neurologists perceive the role of diet in multiple sclerosis (MS)?
4. How do pwMS respond to the nutrition information that they receive?

To answer the research questions and address the knowledge gaps that were highlighted in Chapter 1, this PhD project involved four distinct phases which were addressed by the following research objectives:

1. To identify what nutrition education programs have been implemented for adults with neurological diseases, and the characteristics of those programs
2. To determine the characteristics of effective education programs for people with MS, namely emotional wellness programs
3. To explore the perceptions and experiences of neurologists in giving dietary advice and of pwMS in receiving dietary advice

4. To develop an evidence-based nutrition education program for people with MS and explore the acceptability and ease of comprehension of one draft module

Five studies were conducted to address the thesis aim and objectives, which are detailed in Chapters 3 to 7 and outlined below (Figure 1.3).

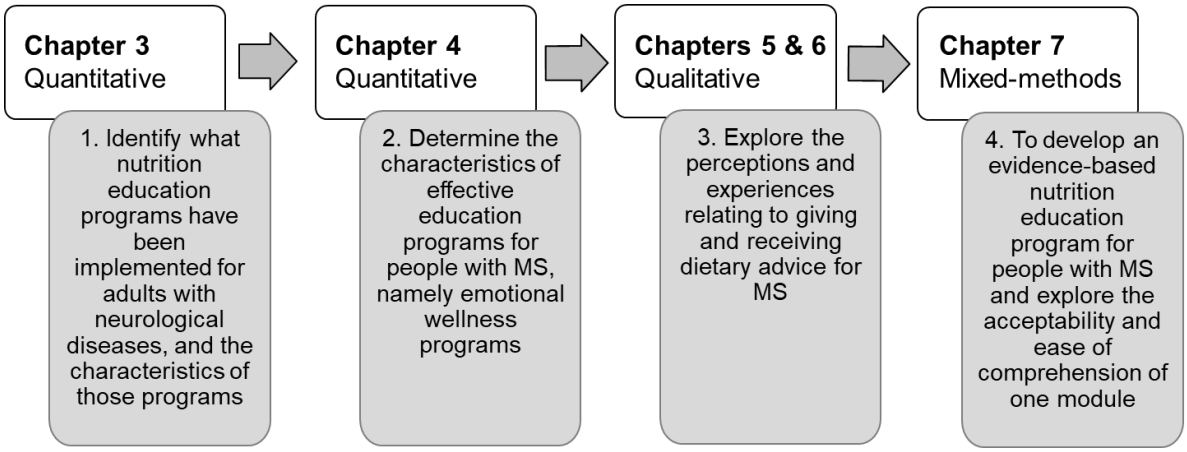

Figure 1.3: Research objectives addressed in each of the thesis chapters.

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Chapter 2. Methodology

2.1 Personal context and epistemology

Defining my positionality within the research may assist the reader to understand how my personal context may have influenced the research topic, the conclusions that I have formed, and the lens through which I view this topic. My choice of epistemology and methodologies that underpin this thesis stem from rigorous self-reflection and questioning. I have adopted a relativist ontological position, where reality and knowledge are constructed within the human mind and through lived experience.[85] What is perceived as reality or knowledge is also shaped by social constructs, including the social constructs of nutrition and disability; hence I have used social constructionism as the epistemological approach in this thesis.[86] It is imperative to understand the subjective realities and experiences of pwMS and regarding how they perceive diet and dietary advice. By listening to the realities of pwMS and their neurologists, I anticipated that I would begin to understand what is important to them and how knowledge would translate into a valuable nutrition education program.

I identify as a white, cis-female, Australian adult who is a mother to three young children. I have completed a chef trade apprenticeship (Certificate III in Commercial Cookery) and achieved tertiary-level education through completion of a Bachelor of Science (Nutrition) and a Bachelor of Science (Health Science) (Honours). Throughout my nutrition degree I developed a keen interest in the sociocultural factors that influence what and how we eat and relationships between diet and diseases. When the time came to choose an Honours project, I was driven by my interest in food and disease and was intrigued to read about a potential project to explore the dietary attitudes and experiences of people newly diagnosed with multiple sclerosis (MS). After a brief search, I learnt that people who were most commonly diagnosed with MS were women, aged 20-40 years. It struck me that I embodied those statistics, as a 29-year-old female. At the time, I was raising a young family, had a mortgage, worked part-time, and as I neared the end of my university studies, I was thinking about my career direction. This was a profound period in my life, and as I pondered about what the future would be like I had the thought of ‘what if’ – what if I was suddenly diagnosed with an incurable, lifechanging disease? What would that be like, and what would that mean for my future?

As part of my Honours research in 2017, I had the privilege of interviewing 11 people who had been diagnosed with MS within the previous 15 months.[74] They shared with me how

they frustratingly received very little dietary advice, and any advice they did receive was perceived as too vague: essentially, general healthy eating information. They described how they conducted their own searches for nutrition information, based on the assumption that diet plays a vital role in overall health and therefore surely must affect MS symptoms and progression. Yet pwMS were confronted with conflicting information and found it difficult to determine what was credible. The participants interviewed were driven and motivated to self-experiment with diet in the hopes that they could alleviate their symptoms, reverse the damage from MS, or even cure themselves – factors that they had been led to believe from their quest for information about diet. They were grateful to have the opportunity to share their experiences and have their voices heard.

> *What you're doing right now with me here [the interview] is vital to any MS person, and I'm just one person. I just feel really grateful that you cared enough to, you know, take me on. Thank you.*
>
> — Honours research participant, 2017.

From the responses in the Honours interviews and the feedback about how important the research into diet is for pwMS, this indicated to me that this was not only a knowledge gap, but it was also a gap that was significant and vital to pwMS. My motivations for undertaking this research were influenced by my personal position, my expert knowledge base in this field, and the likely significance that this research would have for pwMS. The 2017 interviews prompted me to reflect upon my highly privileged position of tertiary education, with which came a fundamental knowledge of food, nutrition, and how to judge the credibility of information. I reflect on my biases and assumptions in this space and find myself questioning that if I were diagnosed with MS, would I be level-headed enough to make sense of the nutrition misinformation, or would I self-experiment with different diets on the slim chance that it *might* help my MS? The lack of nutrition education resources for people newly diagnosed with MS was evident and became a driving force for me to continue from my Honours research to develop a practical, useful, and viable nutrition education program through research involving participants (both pwMS and MS health professionals). The realities of living with MS and the symptoms had to be contextualised, and the impact of these symptoms formed part of my research design for pragmatic reasons: to develop an evidence-based program was that end-user-appropriate. Therefore, it was imperative to base the development of the nutrition education program on methods that would support the experiences, values, and opinions of pwMS and MS health professionals.

In this section I have defined my positionality to justify and explain my relativist ontological position and social constructionism as the epistemological approach that have informed the methods used in this thesis. A mixed-methods methodological approach, explored in the following section, allowed me to position and amplify the voices of pwMS as the primary experts of living with MS.

2.2 Study design

The research design for this thesis was a mixed-methods study design with sequential phases that were conducted over time within one overall study.[87] The methods for each of the phases are detailed in the relevant thesis chapters (Chapters 3-7). This section will briefly outline the methods relating to each of the research objectives (Figure 2.1).

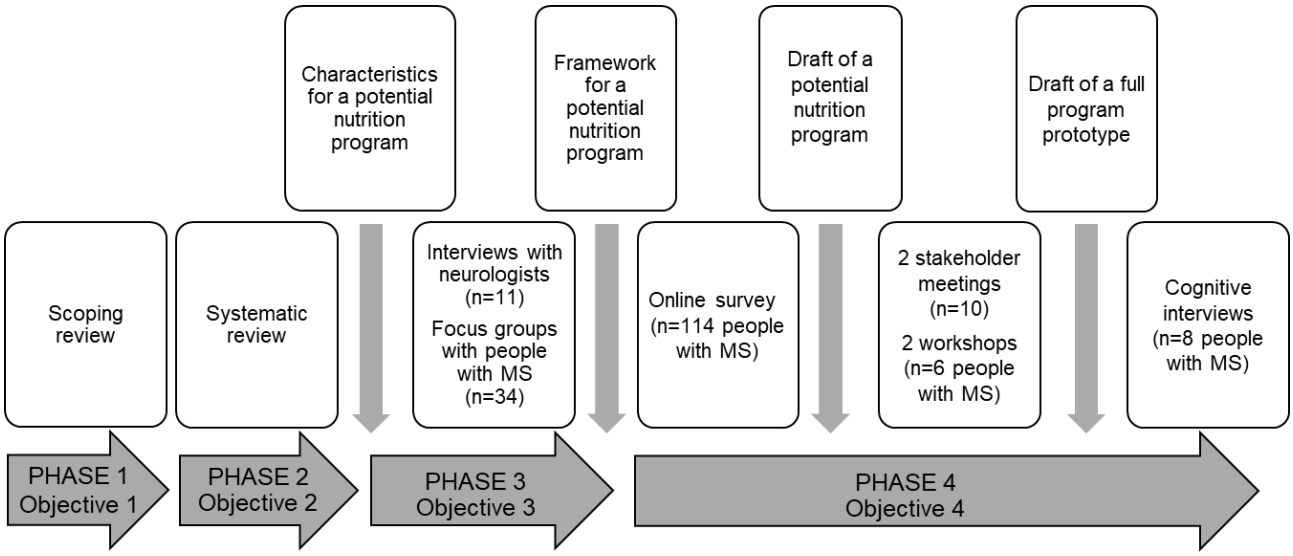

Figure 2.1: Overview of the methods showing the four sequential phases of this PhD project.
MS, multiple sclerosis

2.3 Phase 1 (objective 1)

2.3.1 Study 1 Scoping review

A systematic scoping review was conducted for this objective, as per a published *a priori* protocol,[88] following Joanna Briggs Institute guidelines.[89] Qualitative and intervention studies of nutrition education programs for adults with dementia, epilepsy, Huntington's disease, motor neurone disease, multiple sclerosis (MS), Parkinson's disease, and stroke were retrieved following a three-stage search strategy of six databases for published studies and three databases for unpublished studies and grey literature. 60 relevant

neurological disease organisations were contacted by email. The studies were screened against the inclusion and exclusion criteria and the following data were extracted: author(s), year of publication, country of origin, context, participants (age, sex, and neurological disease), comparator group details (if applicable), sample size, nutrition education program details (topics, format, duration, underlying theories used, and behaviour change techniques (BCTs) used, and nutrition-related outcomes. The findings were presented in a narrative summary.

2.4 Phase 2 (objective 2)

2.4.1 Study 2 Systematic review

A systematic review and meta-analysis were conducted for this objective. The review was conducted in accordance with a registered protocol (PROSPERO registration number CRD42019131082) and following the Joanna Briggs Institute methodology for systematic reviews of effectiveness.[90] Quasi-experimental trials and randomised controlled trials of emotional wellness programs for adults with MS were retrieved following a three-step search strategy. Five databases were searched for published studies and three databases were searched for grey literature. The studies were screened against the inclusion and exclusion criteria and the following data were extracted: study characteristics, aim, participant details (type of MS, sample size, age, sex, duration of MS), intervention details (type, number of study arms, description of intervention, type of comparator group, duration and number of sessions, delivery method), behaviour change theory/ used, BCTs used, tools used to measure outcomes, and results. Methodological quality was assessed using the Joanna Briggs Institute critical appraisal checklists for quasi-experimental trials and randomised controlled trials. Random effects meta-analyses were performed for outcomes assessed in at least five studies. Meta-regression was used to investigate potential predictors that may explain high heterogeneity for outcomes with at least ten studies.

Findings from Phase 1 and Phase 2 were used to develop a list of potential characteristics and BCTs for a potential nutrition education program for pwMS.

2.5 Phase 3 (objective 3)

Qualitative methods were used to address this objective with two distinct studies. The first study involved semi-structured interviews with neurologists and the second study involved focus groups with people with MS (pwMS).

2.5.1 Study 3 Interviews with neurologists

The study was guided by a general inductive approach.[91] Purposive and snowball sampling was used to recruit neurologists working in Western Australia (WA). Individual semi-structured interviews were conducted either in-person or online using web-conferencing software (Skype) until data saturation was reached. Interviews were audio-recorded and transcribed verbatim. Inductive thematic analysis was used to analyse the transcript data.[91]

2.5.2 Study 4 Focus groups with people with MS

PwMS were purposively recruited from a local MS organisation (Multiple Sclerosis Western Australia [MSWA]). Focus groups were held at MSWA branch locations around the Perth metropolitan region, and the focus group discussions were guided by a topic guide. Focus groups were conducted until data saturation was reached. The group discussions were audio-recorded and transcribed verbatim, and data were analysed using a general inductive approach.[91]

The Curtin University Human Research Ethics Committee approved these studies (HRE2019-0179).

The findings from Phases 1 to 3 were used to develop a framework for a potential nutrition education program.

2.6 Phase 4 (objective 4)

2.6.1 Study 5 Using co-design principles to develop a nutrition education program

This objective was addressed using a mixed-methods study, with sequential phases conducted over a period of time.[87] There were three distinct stages in this study.

2.6.1.1 Stage 1 Quantitative survey

A cross-sectional online survey was used to determine the interest and preferred characteristics and content of a nutrition education program for pwMS. Participants were purposively sampled from MSWA. The survey contained 22-34 questions, depending on participants' responses, with questions capturing dietary changes made since diagnosis, reasons for making dietary changes, sources of dietary information, interest in a nutrition education program, and the preferred characteristics (mode of delivery and length, number, and frequency of sessions) and content of a nutrition education program. Differences in participant characteristics were explored by "interested in a nutrition education program" versus "not interested" using t-tests, Wilcoxon rank sum tests, and chi squared tests, as appropriate, and multivariable logistic regression was used to explore potential predictors of interest in a nutrition education program (covariates: sex, age, time since diagnosis, level of education, employment status, if dietary were changes made since diagnosis, and current adherence to a specific diet). The Curtin University Human Research Ethics Committee approved this study (HRE2020-0484).

A draft of a potential online nutrition education program was developed by building on the framework outlined at the end of Phase 3 and findings from the quantitative survey.

2.6.1.2 Stage 2 Qualitative stakeholder meetings and consumer workshops

The stakeholder advisory group and consumers (n=6 pwMS) provided feedback on the draft of the potential nutrition education program in two online meetings and two online workshops, respectively. The workshop participants were recruited through the Consumer and Community Involvement Program (CCIP; details about the CCIP are outlined in the stakeholder advisory group chapter). The Curtin University Human Research Ethics Committee approved this study (HRE2019-0179).

The participants provided input and feedback on the program topics, content, and proposed supporting activities. Informal data analysis involved the confirmation or rejection of the topics discussed and the inclusion of any missing information into the program.

All the feedback from the stakeholders and consumers was incorporated into the program to create a full nutrition education program prototype. The online program was proposed to be asynchronous and run for six weeks, with one module released each week. An instruction designer produced an online version of one module using a range of modes, including text, images, interactive graphics, and videos. The content of the module included how diet may affect the progression and symptoms of MS and tips for managing fatigue while shopping, preparing, and cooking foods.

2.6.1.3 Stage 3 Qualitative cognitive interviews

Cognitive interviews were used to assess the acceptability and ease of comprehension of one module of the online nutrition education program prototype. Participants were purposively sampled from pre-existing sampling frames,[92] to select potential participants based on time since diagnosis, sex, age, and level of education.

The participants took part in individual online interviews via Microsoft Teams, and used a 'think aloud' technique throughout the interview.[92] Verbal probing was used to determine the participants' thought processes concerning the study objectives. The interviews were recorded and transcribed verbatim, and data collection continued until the sampling criteria were achieved. The Framework method[92] was used to summarise the data, and the data were analysed using deductive content analysis.[93]

The Curtin University Human Research Ethics Committee approved this study (HRE2019-0179).

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Chapter 3. Nutrition education programs for adults with neurological diseases

Thesis objective addressed in this chapter:

Objective 1: To identify what nutrition education programs have been implemented for adults with neurological diseases, and the characteristics of those programs.

The content of this chapter is covered by Publication 1:

Russell, R. D., Black, L. J., & Begley, A. Nutrition education programs for adults with neurological diseases are lacking: a scoping review. *Nutrients*. 2022;14:1577. https://doi.org/10.3390/nu14081577

The version that appears in this thesis is of an article that has been through peer-review with *Nutrients* but has not been through the copyediting process.

The contribution of co-authors, A/Prof Andrea Begley and A/Prof Lucinda Black are detailed in the author attribution statements in Appendix A: Attribution statements.

This review was conducted according to a published protocol, Appendix D: Published scoping review protocol.

Russell, R. D., Black, L. J., & Begley, A. Dietary education programs for adults with neurological diseases: a scoping review protocol. *JBI Evid Synth*. 2020;19(1): 170-176. https://doi.org/10.11124/JBISRIR-D-19-00394

The contribution of co-authors, A/Prof Andrea Begley and A/Prof Lucinda Black are detailed in the author attribution statements in Appendix D3.

Review: Nutrients

Nutrition Education Programs for Adults with Neurological Diseases Are Lacking: A Scoping Review

R. D. Russell[a], L. J. Black[a,b], A. Begley[a]

Affiliations:

[a]School of Public Health, Curtin University, Perth, Australia

[b]Curtin Health Innovation and Research Institute

Corresponding Author:

Andrea Begley

School of Public Health, Kent St, Bentley WA 6102

a.begley@curtin.edu.au

+61 8 9266 2773

Abstract:

The nutrition recommendation for most common neurological diseases is to follow national dietary guidelines. This is to mitigate malnutrition, reduce the risk of diet-related diseases, and to help manage some common symptoms, including constipation. Nutrition education programs can support people in adhering to guidelines; hence the aim of this scoping review was to explore what programs have been implemented for adults with neurological diseases. We con-ducted this review according to a published a priori protocol. From 2555 articles screened, 13 were included (dementia n=6; multiple sclerosis n=4; stroke survivors n=2; Parkinson's n=1). There were no programs for epilepsy, Huntington's, and motor neurone disease. Program duration and number of sessions varied widely; however, weekly delivery was most common. Just over half were delivered by dietitians. Most did not report using a behavior change theory. Commonly used behavior change techniques were instruction on how to perform a behavior, credible source, and behavioral practice/rehearsal. Evidence of nutrition education programs for adults with neurological diseases is lacking. Of those that are published, many do not meet best practice principles for nutrition education regarding delivery, educator characteristics, and evaluation. More programs aligning with best practice principles are needed to assess characteristics that lead to behavior change.

Keywords:

Behavior change techniques; behavior change theories; dietary guidelines; neurological diseases; nutrition education.

1. Introduction

Neurological diseases are an increasing cause of morbidity and mortality and they are now the leading cause of disability and the second leading causing of death, globally [1, 2]. Common neurological diseases include Alzheimer's disease and other dementias, epilepsy, Huntington's disease, motor neurone disease, multiple sclerosis, Parkinson's disease, and stroke [1, 3, 4], and they affect up to one billion people worldwide [2]. The nutrition recommendation for most of the common neurological diseases is to follow national dietary guidelines. Such guidelines vary between countries in the specific details, but overall they promote consumption of a wide range of nutritious foods from each of the defined food groups, and to limit consumption of highly-processed foods and drinks that are high in added sugars, salt, saturated fat, and alcohol [5, 6]. These recommendations are given to people with neurological diseases to ensure they consume a high-quality diet and achieve optimal dietary intakes to prevent malnutrition, which may help to manage some common symptoms of neurological diseases, including weight loss or gain and constipation [5, 6]. Furthermore, adherence to national dietary guidelines has been shown to reduce the risk of diet-related noncommunicable diseases, including cardiovascular disease, type 2 diabetes, and obesity [5, 6], which are common comorbidities of neurological diseases [7-10]. Epidemiological evidence has also indicated that type 2 diabetes, obesity, and vascular risk factors such as hypertension and dyslipidemia, are potentially modifiable risk factors for Alzheimer's disease and dementia [11]. As the majority of the general population does not achieve the food group recommendations within national dietary guidelines [12-15], there is an opportunity for people newly diagnosed with any neurological disease to focus on improving dietary intakes in ways that are tailored to their symptoms, to improve vascular and brain health and prevent comorbidities.

Nutrition education involves a set of educational strategies to improve nutrition-related behaviors and dietary intakes that are beneficial for health and wellbeing [16]. The World Health Organization recommends nutrition education programs as a way of promoting healthy diets that are in line with national dietary guidelines to reduce the risk of noncommunicable diseases [17]. Nutrition education programs have been shown to improve nutrition-related knowledge and behaviors in the general population [18], and programs that are tailored to an individual's dietary needs appear to be more promising at improving diet quality than non-tailored programs [19]. Best practice principles for nutrition

education programs fall within five domains: 1) pro-gram design (including content areas, evidence based, goal setting, appropriate for audience including increasing recognition for co-designed programs [20], and theoretical basis); 2) program delivery (including experiential activities and fidelity); 3) educator characteristics (including expertise in content and teaching methods); 4) educator training (including initial and ongoing training); and 5) evaluation (including formative, process, and outcome evaluations, and sustained behavior change) [21].

Disease-specific programs could provide participants with messages that are tailored to their situation and enable them to share their experiences with peers who can empathize to build social support. For such programs to be effective at prompting behavior change, they must be grounded in evidence-based research and incorporate ap-propriate theories [22, 23] and behavior change techniques (BCTs) [24]. To develop effective nutrition education programs, it is important to identify which theories and BCTs have the most potential to support the desired changes in dietary behaviors in the target population [25]. To date, it is not clear what nutrition programs exist for adults with neurological diseases, or the characteristics of such programs, including theories and BCTs.

Scoping reviews aim to map the current evidence on a topic when an area of re-search has not been comprehensively reviewed. Unlike systematic reviews, the purpose of this review was not to assess the effectiveness of the retrieved studies [26]. The aim of this scoping review was to explore what nutrition education programs have been implemented for adults with neurological diseases. The objectives of this review were to determine: 1) which neurological disease populations nutrition education programs have been implemented in; 2) the characteristics of nutrition education programs; and 3) which behavior change theories and techniques have been used in the programs. A preliminary search of PROSPERO, MEDLINE, the Cochrane Database of Systematic Reviews, and JBI Evidence Synthesis revealed that there were no current or underway systematic or scoping reviews on this topic.

2. Methods

This scoping review was carried out according to an a priori protocol [27], in accordance with the Joanna Briggs Institute (JBI) methodology for scoping reviews [28] and the Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for Scoping Reviews [PRISMA-ScR] [29] (Table S1).

2.1. Inclusion criteria

2.1.1. Participants

This review considered studies that included adults (≥ 18 years) with any of the following neurological diseases: dementia (including Alzheimer's disease), epilepsy, Huntington's disease, motor neurone disease, multiple sclerosis, Parkinson's disease, and stroke. Exclusions were: mixed-disease populations where data for the neurological disease/s of interest could not be extracted; adults requiring medical nutrition therapy such as percutaneous gastrostomy or nasogastric tubes; and ketogenic dietary therapy for epilepsy.

2.1.2. Concept

The concept considered in this review was nutrition education programs, i.e., education strategies to improve nutrition-related behaviors. Programs could be in any format (group or individual; in-person, web-based, or teleconference), of any duration (set duration or self-paced), and run for any number of sessions (single or multiple sessions). Studies reporting any of the following outcomes were considered: dietary behaviors, attitudes, or knowledge; diet quality; dietary patterns; biomarker data for nutrient or food intake; or change in intake of nutrients, energy, or food groups. We excluded: 1) dietary clinical trials with a focus on therapy or treatment and with no education component (e.g., vitamin supplementation trials); and 2) lifestyle interventions where <50% of the program pertained to nutrition.

2.1.3. Context

This review considered studies that implemented a nutrition education program in any setting, including educational institutions, community centers, hospitals, care facilities, and home settings.

2.1.4. Types of studies

Both qualitative and intervention studies, with and without comparators, were considered in this review.

2.2. Search strategy

A three-stage search strategy was adopted for this review, and has been described in detail in our protocol [27]. Briefly, MEDLINE and CINAHL were initially searched to identify

relevant terms, which were used to develop a full search strategy (Table S2). We searched CINAHL, Cochrane, Emcare, MEDLINE, ProQuest, and PsycInfo for published studies. We further searched Google Scholar and ProQuest Theses and Dissertations for unpublished studies and grey literature. Finally, the reference lists of all included studies were searched for additional studies. We only considered studies published in English and we did not apply any date restrictions. The initial search was conducted in August 2019 and updated in January 2022. Relevant neurological disease organizations were identified from an internet search using Google, using the terms “[disease] site:org”, “[disease] international”, and “[disease] national”, for all included neurological diseases. All organizations were contacted by email (Table S3).

2.3. Study selection

Search results were uploaded into EndNote X9 (Clarivate Analytics, USA). One reviewer (Author A) screened all titles and abstracts. The full texts of studies were imported into the JBI System for the Unified Management, Assessment and Review of Information (JBI SUMARI) (2019, Joanna Briggs Institute, Adelaide, Australia), and screened independently by two reviewers (Author A and Author C). Disagreements were resolved through discussion.

2.4. Data extraction

Data were extracted using the data extraction tool specified in the protocol [27], and included: study details (author(s), year of publication, country of origin, context, study design, fidelity/drop-outs), target population (neurological disease, age, sex, sample size, comparator group details (if applicable)), and characteristics of the education program (topics, format, duration, nutrition-related outcome measures, behavior change theories, and BCTs used - assessed against Michie et al.’s 93-item taxonomy [24]). Authors were contacted to request missing or additional data, and a follow-up request sent four weeks later, as required.

3. Results

3.1. Search results

The search strategy retrieved 3121 articles, and 2555 articles were screened by title and abstract once duplicates were removed. Full text articles were accessed for the remaining studies, and 26 were excluded (Table S4). We emailed 61 international neuro-logical

disease organizations from the United States, Canada, the United Kingdom, Europe, Australia, and New Zealand (8 dementia; 11 epilepsy; 7 Huntington's disease; 6 motor neurone disease; 10 multiple sclerosis; 9 Parkinson's disease; and 10 stroke), and received responses from 35 (Table S3); no relevant information was retrieved. Thirteen studies were included in this review (Figure 1).

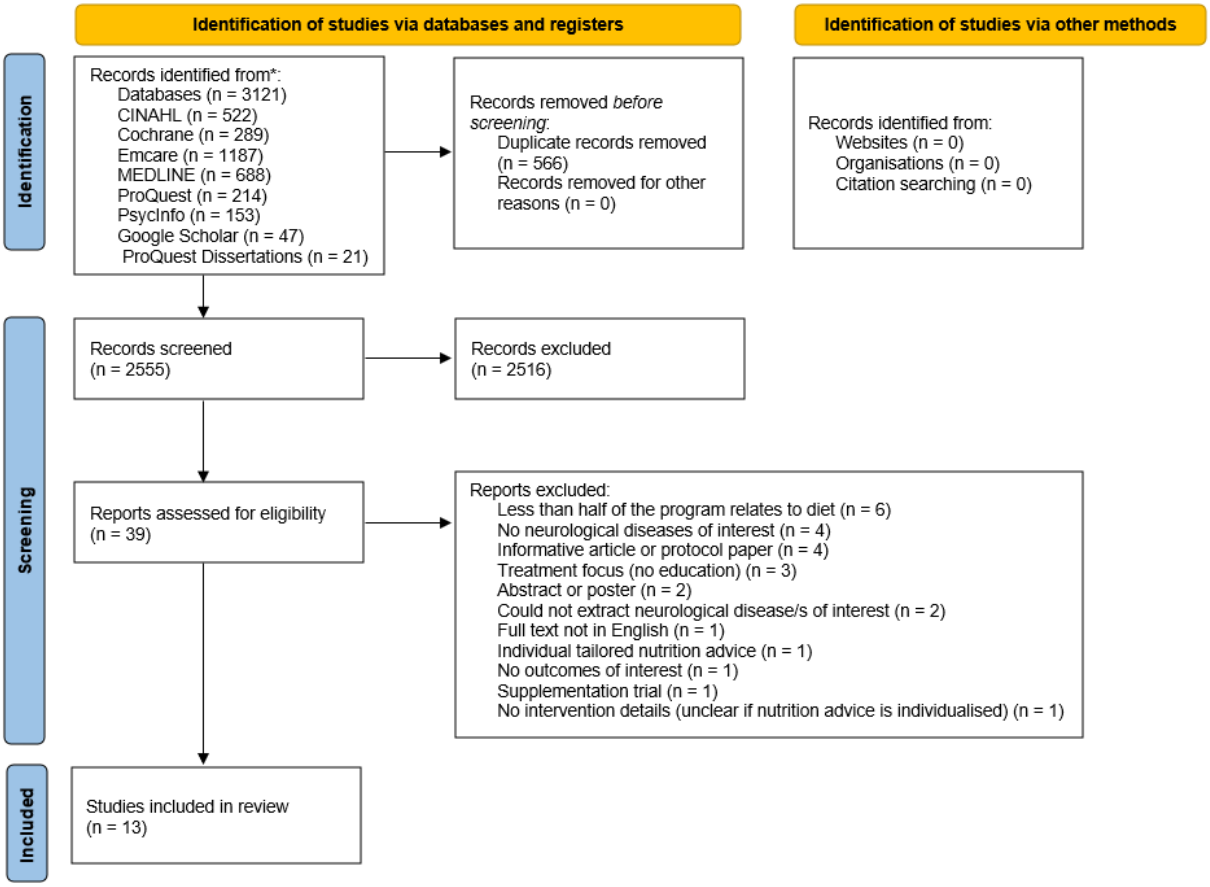

Figure 1. Flowchart showing the scoping review searching and screening processes [30].

3.2. Study details

Table 1 shows the characteristics of the 13 included studies. The studies involved people with dementia (six studies [31-36]), people with multiple sclerosis (four studies [37-40]), stroke survivors (two studies [41, 42]), and people with Parkinson's disease (one study [43]). The studies were conducted in the United States [39-43], Brazil [33], Korea [31], Sweden [32], the United Kingdom [37], Germany [38], Spain [35], and Taiwan [36], and one study was conducted in three countries (France, Italy, and Spain) [34]. Eight of the studies compared an intervention group to a comparator group/s, in either a quasi-controlled trial [32], a randomized [35] or non-randomized cluster trial [34], or a

randomized controlled trial [33, 36, 39, 41, 42]. Five studies did not have a comparator group [31, 37, 38, 40, 43]. Of the eight studies with comparator groups, six enlisted treatment as usual/waitlist control groups [33-36, 41, 42], and three had active compar-ator groups [32, 33, 39]. One study had two comparator groups: one treatment as usual group, and one active comparator group [33]. Participants in the active comparator groups received nutritional supplements [32, 33] or participated in education seminars [39]. There were no qualitive studies that met the inclusion criteria.

Table 1. Characteristics of the 13 studies that met the inclusion criteria of a scoping review of nutrition education programs for adults with neurological diseases.

Year	Author	Study design	Sample size (n)	Age mean (SD) (years)	Intervention description	Delivery method	Intervention duration and frequency	Comparator	Behavior change theory used	Number of BCTs used	Diet/nutrition outcome (tool)
	Dementia										
2019	Cho and colleagues [31]	Pre-post	23	83.5 (4.9)	Physical activity and nutrition education for people with mild dementia. Nutrition topics: the concept of health, proper eating habits, nutrition and nutrients, and the problems of hyper-nutrition and nutrient deficiency.	NR	20 minutes; 16 sessions over 16 weeks	None	NR	3	Nutritional status (Mini Nutritional Assessment)
2002	Faxen-Irving and colleagues [32]	Quasi-controlled trial	33 (IG 21; CG 12)	84.0 (4.0)	Nutrition education for caregivers, plus nutritional supplements for people with dementia for 6 months. Education included practical exercises. Topics: malnutrition, food and nutritional requirements, dental care, detecting swallowing difficulties, altering food consistency.	Group, in-person	12 hours; 1 session	Nutritional supplement only	NR	4	Nutritional status (serum albumin, transferrin, B12, and hemoglobin)
2020	Hsaio and colleagues [36]	RCT	57 (IG 30; CG27)	74.0 (10.2)	Nutrition education for people with dementia and their caregivers, including practical exercises and demonstrations. Topics: altered eating, nutritional imbalances, Mediterranean diet preparing food, healthy fast food., videos.	Group, in-person	1 hour plus 10-15 minutes phone calls; 6 sessions plus 3 phone calls over 3 months	Treatment as usual plus telephone counselling	Knowledge-attitude-behavior Model, Bandura's Social Learning Theory, and the integrative model of mediators of health behavior change	6	Caregiver's nutritional knowledge (Family Caregivers Nutritional Knowledge of Dementia); caregiver's healthy eating behavior (Family Caregiver's Healthy Eating Behavior for Dementia Checklist); and nutritional status (Mini Nutritional Assessment)

2011	Pivi and colleagues [33]	RCT	78 (IG 25; CG1 27; CG2 26)	75.2 (76*)	Nutrition education for people with dementia and their caregivers. Topics: nutrition in disease, behavioral changes during meals, attractive meals, constipation, hydration, administration of drugs, swallowing, food supplementation, lack of appetite.	Group, in-person	NR; 10 sessions over 6 months	CG1: treatment as usual CG2: nutritional supplement twice daily	NR	1	Nutritional status (total protein and serum albumin)
2001	Riviere and colleagues [34]	Non-randomized cluster trial	225 (IG 151; CG 74)	76.3 (8.0)	Nutrition education for caregivers of people with dementia at a day hospital. Topics: weight loss consequences, eating behavior disorders, enriching food, nutritional recommendations, increasing protein and energy intake.	Group, in-person	1 hour; 9 sessions over 1 year	Treatment as usual (patients and caregivers from day hospitals in France and Spain)	NR	9	Nutritional status (Mini Nutritional Assessment); and caregiver's nutritional knowledge (Family Caregivers Nutritional Knowledge of Dementia)
2011	Salva and colleagues [35]	Cluster randomized trial	946 (IG 448; CG 498)	79 (7.3)	NutriAlz nutrition program for families and caregivers of people with dementia. Topics: weight loss, nutritional monitoring, the food pyramid, menu creation, cooking methods, food substitution, eating behavior problems.	Group, in-person	NR; 4 sessions over 1 year	Treatment as usual (five patient day care centers)	NR	4	Nutritional status (Mini Nutritional Assessment)
	Multiple sclerosis										
1993	Doidge and colleagues [37]	Pre-post	48	46.9 (9.9)	Nutrition education for people with multiple sclerosis. Topics: *The Action and Research for Multiple Sclerosis* healthy eating plan, saturated and polyunsaturated fat, preparing food at home, understanding food labels, suitable convenience food, vitamins and minerals,	Group, in-person	90 minutes; 8 sessions over 8 weeks	None	NR	8	Diet composition (daily energy intake and nutrient intakes)

					weight maintenance, recipe tasting.						
2019	Katz Sand and colleagues [39]	Pilot RCT	34 (IG 18; CG 16)	43 (NR)	Nutrition education for people with multiple sclerosis (groups of five); Mediterranean Diet. Topics: shopping tips, sample menu plan, reading food labels, eating at restaurants. Participants returned monthly (or dialed in) to discuss issues with following the diet.	Group, in-person and/or telehealth	NR; 6 sessions over 6 months	MS education seminars	NR	6	Dietary adherence and food group intake (food frequency questionnaire); and perceived benefits
2016	Riemann-Lorenz and colleagues [38]	Single aim, post	11	38.5 (12.3)	Nutrition education for people with multiple sclerosis (1 session), including 2 short group discussions. Topics: epidemiology, research study designs, study endpoints and problems, experiences with multiple sclerosis diets, common multiple sclerosis diets, RCTs of diet and multiple sclerosis.	Group, in-person	2 hours; 1 session	None	NR	3	Novelty of information/ knowledge; importance of information; and impact of information
2020	Wingo and colleagues [40]	Single arm, post	18	46.0 (11.6)	Nutrition education and physical activity education for people with multiple sclerosis, for the low glycemic index diet, including online modules and calls from tele-coaches. Nutrition topics: meal planning, foods to eat and limit, cooking basics, healthy eating on a budget. Weeks 1-5 were standardized information. Weeks 6-12 were tailored to address barriers and goals.	Individual, telehealth	12 online modules (time NR) and 12 20-45 minute phone calls over 12 weeks	None	Health Action Process Approach	10	Diet quality (24-hour food recall); and fat mass (dual-energy X-ray absorptiometry scan)
	Stroke										

2000	Rimmer and colleagues [41]	RCT	35 (IG 18; CG 17)	53.2 (8.3)	Health Promotion program for stroke survivors (exercise, nutrition, and health behavior classes), including cooking demonstration and practice. Nutrition topics: low-fat and low-cholesterol foods, preparation of healthy meals, healthy food substitutes.	Group, in-person	1 hour; 36 sessions over 12 weeks	Waitlist controls	Transtheoretical (Stage of Change) Model	11	Dietary fat intake (Rate Your Plate Eating Pattern Assessment) and blood lipid profile (total cholesterol, high-density and low-density lipoprotein cholesterols, triglycerides)
2020	Towfighi and colleagues [42]	RCT	100 (IG 49; CG 51)	58.0 (9.0)	*Healthy Eating and Lifestyle After Stroke* program for stroke survivors. Nutrition topics: healthy dietary patterns, monitoring food intake, food label reading, shopping, purchasing healthy foods, diet as a means of secondary stroke prevention.	Group, in-person	2 hours; 6 sessions over 6 weeks	Treatment as usual	Transtheoretical (Stage of Change) Model, Health Belief Model, and Social Cognitive Theory	11	Serves of fruits/vegetables per day; waist circumference; and blood lipid profile (total cholesterol, high-density and low-density lipoprotein cholesterols, triglycerides, hemoglobin A1c)
	Parkinson's disease										
2000	Brenes [43]	Pre-post	15	69.0 (NR)	Virtual nutrition education program for people with Parkinson's disease and their caregivers. Included lesson videos, handouts and recipes (video and written). Topics: basic nutrition, healthy eating, Parkinson's disease and the gut, inflammation and Parkinson's disease, constipation and hydration, and 'protein and Levodopa.	Individual, online	Self-paced; 6 sessions over 6 weeks	None	Self-Determination Theory	11	Nutritional status (Mini Nutritional Assessment); intake of macronutrients, micronutrients, and food groups (Diet History Questionnaire 3); nutrition knowledge (nutrition knowledge questionnaire); motivation about nutrition knowledge

SD, standard deviation BCTs, behavior change techniques; NR, not reported; IG, intervention group; CG, comparator group; RCT, randomized controlled trial. *Median report

There was very limited reporting on adherence to the nutrition interventions (fidelity), which was only reported by two studies: 27% of participants did not attend any classes, and 53% attended five out of the six classes in one study (total n = 49) [42], and 90% of participants completed all of the scheduled calls with the telehealth coach in another study, but they did not report what percentage of participants completed the online modules prior to the coaching calls [40]. The rate of drop-out from the studies was under 15% from all studies except for Brenes [43] (46.4% drop-out; Parkinson's disease), Salva and colleagues [35] (29.4%; dementia), and Hsiao and colleagues [36] (17.4%; dementia). The rate of drop-out was unclear in one study [34] (dementia).

3.3. Target populations

There were 1623 participants in the 13 included studies: 1362 with dementia and/or caregivers of people with dementia; 111 with multiple sclerosis; 135 stroke survivors, and 15 with Parkinson's disease. The nutrition education programs were conducted with people with the disease [31, 33, 37-42], people with the disease and their caregivers [33-36, 43], and one program was conducted with only caregivers [32]. There were 875 participants in the intervention groups, and 748 participants in the comparator groups. The total number of participants in the studies ranged from 11 [38] to 656 [35]. The median (interquartile range, IQR) age of participants was 69 (31) years, and the range of reported mean ages was 39 [38] to 84 years [31, 32]. The median (range) proportion of females was 68% (38% [42] - 100% [39]); one study did not report sex [31]. Other socio-demographic data were infrequently reported: seven studies reported education levels [33, 35, 36, 39, 40, 42, 43]; four studies reported comorbidities [32, 35, 36, 41]; and only three studies reported the length of time since diagnosis [33, 39, 43].

3.4. Characteristics of the nutrition education programs

Most of the programs were focused solely on nutrition [32-39, 43], while four pro-grams included physical activity education alongside nutrition education [40-42]. Seven of the programs were delivered by dietitians [32, 34, 35, 37, 39-41] (one of those was alongside physicians [32], and one was alongside facilitators with degrees in health education or kinesiology [40]); one was delivered by a medical student [38]; one was delivered by occupational therapists [42]. There were no details reported regarding nutrition training for non-nutrition professionals. Four programs did not specify the credentials of the facilitators [31, 33, 36, 43]. In eleven education programs [31-34, 36, 37, 39-43], participants were given instructions on how to perform the nutrition-related behaviors, including reading and

interpreting food labels, preparing healthy meals, and detecting swallowing difficulties in people with dementia. Seven studies reported that participants had the opportunity to practice the skills being taught during the education sessions [32, 36-38, 40-42], and only four studies stated that the participants engaged in goal-setting activities [40-43]. There was no evidence of codesign in any of the nutrition education programs. Only participatory research [20] was evident was in two studies, and was used to inform the program topics: one program for people with multiple sclerosis used the findings from a survey [38]; and the program for people with Parkinson's disease used focus groups [43].

Nearly all of the education programs were delivered in-person and in a group format [32-38, 41, 42], except for one telehealth intervention [40] and one online intervention [43]. One study did not report the method of delivery or program format [31]. Of the ten studies that were conducted in a group setting, only six specified that the participants engaged in group discussions [34, 36-38, 41, 42]. The online program used a discussion board to facilitate group discussions [43]. Three studies had missing information on the duration of the education sessions [33, 35, 39] and one program was self-paced with no expected duration reported [43]. Of the studies with complete data, the total hours of program delivery ranged from 2 [38] to 36 [41] (median, IQR: 9.0, 6.6). The shortest nutrition education session lasted for 20 minutes [31], and the longest lasted 12 hours [32] (median, IQR: 1.0, 1.3). The number of sessions for the nutrition education programs ranged from one single session (two studies, lasting two hours [38] and 12 hours [32], respectively) to 36 sessions [41] (lasting one hour) (median, IQR: 6, 8). The most common frequency of delivery was weekly (six programs [31, 36, 37, 40, 42, 43]). One program was delivered three days per week [41], one program involved five sessions in month one, then one session in months two, three, and six [34] and one program was delivered fortnightly [39]. The frequency of delivery was unclear in two programs: both of these programs also did not report the duration of the sessions [33, 35].

The studies included a range of outcome measures to evaluate the effectiveness of the nutrition education programs. Dietary intake was assessed in six studies [37, 39-43] using food diaries, food frequency questionnaires, and/or 24-hour recalls. Nutritional status was measured using the Mini Nutritional Assessment in five studies [31, 34-36, 43], and two studies used biomarkers of nutritional status, including serum albumin, transferrin, total protein, vitamin B12, and/or hemoglobin [32, 33]. Blood lipid biomarkers of dietary intake, such as triglycerides, total cholesterol, and high-density and low-density lipoprotein cholesterols, were used in two studies [41, 42]. Three studies [34, 36, 43] evaluated the

nutrition knowledge of the participants, using the Family Caregivers Nutritional Knowledge of Dementia or a nutrition knowledge questionnaire. Only one study evaluated the program effectiveness with measures of perceived benefits [39], and only one study measured perceived novelty, importance, and impact of information [38].

3.4.1. Nutrition education programs for people with dementia

Of the six nutrition education programs for people with dementia, four included the caregivers of people with dementia [33-36] and one program was for the caregivers only [32]. The topics included in the programs were: malnutrition and weight loss, nutritional requirements, eating behavior problems, detecting swallowing difficulties, enriching the nutritional quality of foods, altering the consistency of foods, constipation, lack of appetite, and cooking methods. Only two studies specified that the participants practiced during the sessions the nutrition-related behaviors that were being taught [32, 36].

One nutrition education program was aimed at people with dementia, as opposed to their caregivers [31]. The topics included in the programs were: the concept of health, proper eating habits, nutrition and nutrients, and the problems of hyper-nutrition and nutrient deficiency. Participants were given instructions on how to perform nutrition-related behaviors, such as reading and interpreting food labels, but it was not specified if they practiced those skills during the sessions.

3.4.2. Nutrition education programs for people with multiple sclerosis

All four of the nutrition education programs were delivered to people with multiple sclerosis as opposed to caregivers. Three were in-person, group programs [37-39] and one was a telehealth intervention [40]. Three of the programs were focused on specific diet plans: the Action and Research for Multiple Sclerosis healthy eating plan [37], the Mediterranean diet [39], and a low glycemic-load diet [40]. The topics included in the programs were: healthy eating, reading and interpreting food labels, eating out/convenience foods, meal planning, shopping tips, and cooking at home [37, 39, 40]. One program also included information on common study designs used in nutrition research, popular diets for multiple sclerosis, and results from clinical trials of diet and multiple sclerosis [38]. In three of the studies, instructions on how to perform the de-sired nutrition-related behaviors were provided to participants [37, 39, 40]; two of those studies also gave participants the opportunity to practice those skills during the sessions [37, 40]. In half of the nutrition education programs, participants engaged in group discussions [37, 38].

3.4.3. Nutrition education programs for stroke survivors

The two education programs for stroke survivors were based on physical activity and nutrition education [41, 42]. The nutrition topics included: preparing healthy meals, food substitutions, meal planning, and how to read and interpret food labels. The participants engaged in goal setting activities and discussed their experiences with other participants. Both programs included visual demonstrations, and participants were given time during the sessions to practice the skills being taught.

3.4.4. Nutrition education programs for people with Parkinson's disease

The one nutrition education program for people with Parkinson's disease included the caregivers alongside people with Parkinson's disease [43]. It was a self-paced, online program consisting of short videos and handouts for each of the weekly topics, as well as videos and handouts with recipe suggestions. The weekly topics were developed from focus group discussions with people with Parkinson's disease and included: basic nutrition; healthy eating; Parkinson's disease and the gut; inflammation; constipation and hydration; and the protein-Levodopa interaction. The participants engaged in goal setting activities and were encouraged to regularly revise their goals. There was an online discussion board to allow participants to share their experiences with other participants.

3.5. Theories and behavior change techniques

Five studies reported using at least one underlying behavior change theory: both of the programs for stroke survivors [41, 42] used the Transtheoretical (Stage of Change) Model and one program [42] also used the Health Belief Model and the Social Cognitive Theory; one program for people with multiple sclerosis [40] used the Health Action Process Approach; one program for people with dementia and their caregivers [36] used the Knowledge-attitude-behavior Model, Bandura's Social Learning Theory, and the integrative model of mediators of health behavior change; and the program for people and caregivers of people with Parkinson's disease [43] used the Self-Determination Theory. However, explanations on how the theory was applied was lacking in the Brenes study [43]. A total of 22 different BCTs were used in the 13 studies in this review (Table 2). During the data extraction stage, two BCTs from two different studies were identified by only one author. The coding of these BCTs was discussed by returning to the BCT taxonomy definitions [24] and a mutual agreement was reached on whether or not the BCT was coded. The median number of BCTs used per program was six (range 1 [33] to 11 [41-43]); however,

some of the studies lacked comprehensive details on the contents of the programs, hence it is possible that more BCTs were used but were unable to be coded. The most commonly used BCTs were: instruction on how to perform a behavior (eleven studies [31-34, 36, 37, 39-43]), credible source (nine studies [32, 34, 35, 37-42]), be-havioral practice/rehearsal (eight studies [32, 36-38, 40-43]), information about health consequences (seven studies [31, 34-36, 41-43]), and social comparison (seven studies [34, 36-38, 41-43]). These BCTs indicate that the majority of the programs gave the participants information on how to change their behavior in relation to dietary intake, were delivered by people with relevant expertise, included practical exercises to give the participants the opportunity to practice the skills being taught, and were a source of peer ex-change by way of group discussions.

Table 2. Behavior change techniques used in each of the 13 studies that met the inclusion criteria of a scoping review of nutrition education programs for adults with neurological diseases.

	Cho and colleagues [31]	Doidge and colleagues [37]	Faxen-Irving and colleagues [32]	Hsiao and colleagues [36]	Katz Sand and colleagues [39]	Pivi and colleagues [33]	Riemann-Lorenz and colleagues [38]	Rimmer and colleagues [41]	Riviere and colleagues [34]	Salva and colleagues [35]	Towfighi and colleagues [42]	Wingo and colleagues [40]	Brenes [43]	**Total n**
Instruction how to perform a behavior	■	■	■	■	■	■		■	■		■	■	■	**11**
Credible source		■	■		■		■	■	■	■	■	■		**9**
Behavioral practice/rehearsal		■	■	■			■	■			■	■	■	**8**
Information about health consequences	■			■				■	■	■	■		■	**7**
Social comparison		■		■			■	■	■		■		■	**7**
Self-monitoring behavior				■	■						■	■	■	**5**
Demonstration of the behavior	■		■	■				■					■	**5**

Problem solving								■		■	■	■		**4**
Adding objects to the environment									■		■	■	■	**4**
Social support (unspecified)								■	■		■			**3**
Goal setting (outcome)								■			■	■		**3**
Framing/reframing					■			■	■					**3**
Feedback on behavior		■										■	■	**3**
Action planning												■	■	**2**
Reduce negative emotions								■	■					**2**
Prompts/cues					■							■		**2**
Review behavior goal(s)											■		■	**2**
Monitoring behavior by others without feedback									■					**1**
Monitoring outcome(s) by others without feedback										■				**1**
Biofeedback		■												**1**
Social support (practical)		■												**1**
Goal setting (behavior)													■	**1**
Total n	**3**	**8**	**4**	**6**	**6**	**1**	**3**	**11**	**9**	**4**	**11**	**10**	**11**	

4. Discussion

We identified the nutrition education programs that have been implemented for adults with neurological diseases, and the characteristics of those programs. In the 28 years ranging from 1993 to 2021, we only found 13 studies that met the criteria of this review. The studies included nutrition education programs for people with dementia, people with multiple sclerosis, stroke survivors, and people with Parkinson's disease. The nutrition topics taught in the programs were evidence-based and relevant to the target group participants, but evidence of co-design was lacking. This review has identified several characteristics of programs that reflect poor design and do not align with the best practices for nutrition education programs [21]: 1) the duration and number of sessions varied between programs and the session duration was missing from nearly a third of the studies: best practice principles state that both sufficient duration and frequency are required to achieve the desired learning outcomes [21]; 2) fidelity (percent-age completion of the nutrition program sessions) was rarely reported; 3) nearly half of the nutrition education

programs were not delivered by dietitians or nutritionists; 4) there was no information about the initial and ongoing training for those delivering the programs; and 5) varying evaluation measures were used which indicates that dietary behavior change was not the focus for evaluation: changes in dietary intakes were not measured in more than half of the studies. The missing data and differences in the program characteristics meant that we were unable to make recommendations for future nutrition education programs for adults with neurological diseases, although weekly delivery was the most common. To facilitate evaluation and improvement of these programs, more programs need to be developed in accordance with best practice principles [21].

No nutrition education programs for people with epilepsy, Huntington's disease, or motor neurone disease met the inclusion criteria of this review, and only one program for people with Parkinson's disease was included. With the exception of intractable epilepsy, for which there is evidence to support the ketogenic diet as a treatment in some cases [44], national and international organizations for these neurological diseases recommend adhering to national dietary guidelines to achieve optimal nutritional intake [45-47]. Malnutrition and weight loss or gain are common problems for adults with neurological diseases: achieving dietary recommendations mitigates this problem and can improve quality of life for people with Huntington's disease, motor neurone disease, and Parkinson's disease [45, 48, 49]. Given that nutrition education programs can support people in meeting the dietary guidelines and achieving nutritional adequacy [18], there is a need for dietitians and nutritionists to be actively involved in using best practice principles to develop nutrition education programs for neurological diseases, particularly for people in early diagnosis to prevent malnutrition. Ultimately this could improve patient education, dietary behaviors, and quality of life for people living with different neurological diseases.

Nutrition education programs should be based on relevant theoretical frameworks [22] for enhanced efficacy [23] and better outcomes for participants [50]. Only five (38%) of the programs in this review were based on theories, and four of those were recently published. Similarly, Plow and colleagues reported that only 24% of nutrition and weight loss interventions for adults with neurological and musculoskeletal conditions were based on a behavior change theory in their systematic review [51].

Interestingly, we found that different theories were used for each of the neuro-logical diseases (dementia, multiple sclerosis, stroke, and Parkinson's disease), but the two programs for stroke survivors were based on the same theory despite being published

twenty years apart. Similar to our findings, a systematic review reported that a range of theories have been used in nutrition education programs, including the Transtheoretical (Stages of Change) Model, Social Learning Theory, Social Cognitive Theory, Adult Learning Theory, and the Health Belief Model [23]. Theories should be used when designing and implementing nutrition interventions [21, 23]; therefore, future nutrition education programs for adults with neurological diseases should adhere to best practice principles by being driven by appropriate theories, to facilitate and support the behavior changes desired by participants.

The median number of BCTs in the included studies was six; however, due to the lack of detail provided for many of the nutrition education programs in this review, it is likely that more BCTs were used in the programs. For example, we only coded social comparison if it was explicitly stated that participants engaged in group discussion; we did not infer this from a group setting [24]. While almost all the programs in this review were conducted in a group setting, only half specified that participants engaged in group discussions. The most common BCTs in the included studies were: instruction on how to perform a behavior; credible source; behavioral practice/rehearsal; and social comparison. Future programs for adults with neurological diseases should consider using these BCTs. Our findings are supported by other reviews of nutrition education interventions: instruction on how to perform a behavior and social comparison have been used in effective nutrition interventions for adults [52]; and instruction on how to perform a behavior and behavioral practice/rehearsal have been used in effective lifestyle interventions (diet and/or physical activity) for adults with chronic kidney disease [53] and obesity [54]. Fur-thermore, our previous systematic review of emotional wellness programs for people with multiple sclerosis found that behavioral practice/rehearsal was the most commonly used BCT in efficacious interventions [55]. Since it is important to identify the combination of BCTs that supports health-related behaviors [25], reporting of nutrition education programs should be in sufficient detail to all allow BCTs to be identified. This would enable researchers to establish a list of BCTs that are effective for adults with neurological diseases and could be used to inform the future development of nutrition education programs.

This review was conducted following the recommendations outlined in the JBI guidelines for conducting scoping reviews, and the PRISMA-ScR checklist for scoping reviews. The search strategy was developed in consultation with a Health Sciences librarian and included published and unpublished literature. This review has some limitations. Firstly, we only included studies in the English language. Secondly, a limitation of scoping reviews is

that they do not include a critical appraisal of the quality of included studies or evaluate efficacy; however, the aim was to map out what programs exist, and not assess the quality or efficacy of the studies.

5. Conclusions

Published evidence of nutrition education programs for adults with neurological diseases is lacking, and those that are published either do not meet best practice guidelines for nutrition education programs, and/or have inconsistent characteristics. Given the role that optimal nutritional intake can play in these diseases, there is a need for dietitians and nutritionists to be involved in designing and implementing nutrition education programs that adhere to best practice guidelines, using codesign to ensure the participants' needs are met. Such programs may help to improve patient education and dietary behaviors, therefore reducing the risk of malnutrition and comorbid diseases, which may improve quality of life. Specifically, the reporting of such programs should include the underlying behavior change theories and be in sufficient detail to allow all BCTs to be identified, including those that we have identified as most commonly used in this review. This would enable future programs to be based on appropriate theories and BCTs that appear to be effective for this population. The nutrition topics that were taught in the programs in this review were appropriate and relevant to the target group participants, and weekly delivery was most common. These characteristics should be considered when developing future nutrition education programs for adults with neurological diseases.

Supplementary Materials: The following supporting information can be downloaded at: www.mdpi.com/xxx/s1, Table S1: Preferred Reporting Items for Systematic reviews and Me-ta-Analyses extension for Scoping Reviews (PRISMA-ScR) Checklist; Table S2: Search strategy for MEDLINE (Ovid) and CINAHL databases. January 2022; Table S3: Neurological disease organizations contacted by email; Table S4: Studies excluded at full text stage of screening and reasons for exclusion.

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Table S1 Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR) Checklist

SECTION	ITEM	PRISMA-ScR CHECKLIST ITEM	REPORTED ON PAGE #
TITLE			
Title	1	Identify the report as a scoping review.	1
ABSTRACT			
Structured summary	2	Provide a structured summary that includes (as applicable): background, objectives, eligibility criteria, sources of evidence, charting methods, results, and conclusions that relate to the review questions and objectives.	1
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known. Explain why the review questions/objectives lend themselves to a scoping review approach.	1-2
Objectives	4	Provide an explicit statement of the questions and objectives being addressed with reference to their key elements (e.g., population or participants, concepts, and context) or other relevant key elements used to conceptualize the review questions and/or objectives.	2
METHODS			
Protocol and registration	5	Indicate whether a review protocol exists; state if and where it can be accessed (e.g., a Web address); and if available, provide registration information, including the registration number.	3
Eligibility criteria	6	Specify characteristics of the sources of evidence used as eligibility criteria (e.g., years considered, language, and publication status), and provide a rationale.	3
Information sources*	7	Describe all information sources in the search (e.g., databases with dates of coverage and contact with authors to identify additional sources), as well as the date the most recent search was executed.	3
Search	8	Present the full electronic search strategy for at least 1 database, including any limits used, such that it could be repeated.	Table S2
Selection of sources of evidence†	9	State the process for selecting sources of evidence (i.e., screening and eligibility) included in the scoping review.	4
Data charting process‡	10	Describe the methods of charting data from the included sources of evidence (e.g., calibrated forms or forms that have been tested by the team before their use, and whether data charting was done independently or in duplicate) and any processes for obtaining and confirming data from investigators.	4 and published protocol paper
Data items	11	List and define all variables for which data were sought and any assumptions and simplifications made.	4
Critical appraisal of individual sources of evidence§	12	If done, provide a rationale for conducting a critical appraisal of included sources of evidence; describe the methods used and how this information was used in any data synthesis (if appropriate).	N/A
Synthesis of results	13	Describe the methods of handling and summarizing the data that were charted.	4
RESULTS			
Selection of sources of evidence	14	Give numbers of sources of evidence screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally using a flow diagram.	4-5. and Table S4

SECTION	ITEM	PRISMA-ScR CHECKLIST ITEM	REPORTED ON PAGE #
Characteristics of sources of evidence	15	For each source of evidence, present characteristics for which data were charted and provide the citations.	5-10
Critical appraisal within sources of evidence	16	If done, present data on critical appraisal of included sources of evidence (see item 12).	N/A
Results of individual sources of evidence	17	For each included source of evidence, present the relevant data that were charted that relate to the review questions and objectives.	5-14
Synthesis of results	18	Summarize and/or present the charting results as they relate to the review questions and objectives.	10-14
DISCUSSION			
Summary of evidence	19	Summarize the main results (including an overview of concepts, themes, and types of evidence available), link to the review questions and objectives, and consider the relevance to key groups.	14-15
Limitations	20	Discuss the limitations of the scoping review process.	15
Conclusions	21	Provide a general interpretation of the results with respect to the review questions and objectives, as well as potential implications and/or next steps.	15-16
FUNDING			
Funding	22	Describe sources of funding for the included sources of evidence, as well as sources of funding for the scoping review. Describe the role of the funders of the scoping review.	16

JBI = Joanna Briggs Institute; PRISMA-ScR = Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews.
* Where *sources of evidence* (see second footnote) are compiled from, such as bibliographic databases, social media platforms, and Web sites.
† A more inclusive/heterogeneous term used to account for the different types of evidence or data sources (e.g., quantitative and/or qualitative research, expert opinion, and policy documents) that may be eligible in a scoping review as opposed to only studies. This is not to be confused with *information sources* (see first footnote).
‡ The frameworks by Arksey and O'Malley (6) and Levac and colleagues (7) and the JBI guidance (4, 5) refer to the process of data extraction in a scoping review as data charting.
§ The process of systematically examining research evidence to assess its validity, results, and relevance before using it to inform a decision. This term is used for items 12 and 19 instead of "risk of bias" (which is more applicable to systematic reviews of interventions) to include and acknowledge the various sources of evidence that may be used in a scoping review (e.g., quantitative and/or qualitative research, expert opinion, and policy document).

From: Tricco AC, Lillie E, Zarin W, O'Brien KK, Colquhoun H, Levac D, et al. PRISMA Extension for Scoping Reviews (PRISMAScR): Checklist and Explanation. Ann Intern Med. 2018;169:467–473. doi: 10.7326/M18-0850.

Table S2 Search strategy for MEDLINE (Ovid) and CINAHL databases. January 2022

MEDLINE (Ovid)		
No.	Search terms	Retrieved
1	exp "autoimmune diseases of the nervous system"/ or exp autonomic nervous system diseases/ or exp central nervous system diseases/ or exp cranial nerve diseases/ or exp demyelinating diseases/ or exp neurodegenerative diseases/ or exp neuromuscular diseases/	1971299
2	Multiple sclerosis.mp. or Parkinson's.mp. or Huntington's.mp. or motor neurone.mp. or Alzheimer's.mp. or dementia.mp. or amyotrophic lateral sclerosis.mp. or stroke.mp. or epilepsy.mp.	924146
3	1 or 2	2279571
4	exp Diet/ or diet*.ti,ab.	735446
5	exp Nutrition Therapy/ or nutrition*.ti,ab.	378612
6	exp Diet Therapy/	59231
7	(diet* adj2 (behavio?r* or quality or pattern* or change)).ab.	26866
8	4 or 5 or 6 or 7	968556
9	exp Health Education/	256186
10	exp Patient Education as Topic/ or patient education.ti,ab.	99169
11	education* program*.ti,ab.	46469
12	exp Health Promotion/	82293
13	diet* education.ti,ab.	682
14	nutrition* education.ti,ab.	6001
15	9 or 10 or 11 or 12 or 13 or 14	305150
16	3 and 8 and 15	773
17	limit 16 to (English language and humans)	688

CINAHL		
No.	Search terms	Retrieved
S1	(MH "Nervous System Diseases") OR (MH "Autoimmune Diseases of the Nervous System+") OR (MH "Autonomic Nervous System Diseases+") OR (MH "Central Nervous System Diseases+") OR (MH "Cranial Nerve Diseases+") OR (MH "Demyelinating Diseases+") OR (MH "Neurodegenerative Diseases+") OR (MH "Parkinsonian Disorders+") OR (MH "Neuromuscular Diseases+")	503637
S2	AB Multiple sclerosis or Parkinson's or Huntington's or motor neurone or Alzheimer's or dementia or prion or amyotrophic lateral sclerosis or stroke or epilepsy	187517
S3	1 or 2	559013
S4	AB diet*	117813
S5	(MH "Diet+")	133347
S6	(MH "Diet Therapy+")	35430
S7	AB "diet* therapy" or "nutrition therapy"	1340
S8	AB "diet* behavio?r*" or "diet* quality" or "diet* pattern*" or "diet* change"	9230
S9	S4 or S5 or S6 or S7 or S8	207,624
S10	(MH "Health Education+")	137306

S11	(MH "Patient Education+")	83196
S12	(MH "Health Promotion+")	76111
S13	AB "diet* education"	314
S14	AB "education* program*"	26054
S15	AB "nutrition* education"	3073
S16	S10 or S11 or S12 or S13 or S14 or S15	223,079
S17	S3 and S9 and S16	522

Table S3 Neurological disease organizations contacted by email

Neurological disease	Organization name	Web address	Response received (yes/no)
Dementia	Dementia Australia	https://www.dementia.org.au/	Yes
	Alzheimer's WA	https://www.alzheimerswa.org.au/	Yes
	Alzheimer's Association	https://www.alz.org/	Yes
	Alzheimer's Disease International	https://www.alz.co.uk/	Yes
	Alzheimer's Association Australia	https://www.alz.org/au/dementia-alzheimers-australia.asp	No
	Alzheimer's Queensland	https://www.alzheimersonline.org/	No
	Alzheimer's Society	https://www.alzheimers.org.uk/	No
	Alzheimer's Foundation of America	https://alzfdn.org/	No
Epilepsy	Epilepsy Action Australia	https://www.epilepsy.org.au/	Yes
	Epilepsy Society	https://www.epilepsysociety.org.uk/	Yes
	Epilepsy Foundation END EPILEPSY	https://www.epilepsy.com/	Yes
	American Epilepsy Society	http://www.aesnet.org/	Yes
	Epilepsy Foundation	http://epilepsyfoundation.org.au/	No
	International League Against Epilepsy and International Bureau for Epilepsy	https://www.epilepsy.org/	No
	Epilepsy Society	http://www.epilepsysociety.org.uk/	No
	The Epilepsy Centre	https://epilepsycentre.org.au/	No
	Epilepsy Foundation	https://epilepsyfoundation.org/	No
	Epilepsy Australia	http://www.epilepsyaustralia.net/	No
	International League Against Epilepsy	https://www.ilae.org/	No
Huntington's disease	Huntington's Disease Society of America	https://hdsa.org/	Yes
	Huntington's NSW ACT	https://www.huntingtonsnsw.org.au/	Yes
	Huntington's Disease Association	https://www.hda.org.uk/	Yes
	Huntington Society of Canada	https://www.huntingtonsociety.ca/	Yes
	Huntington's Western Australia	https://www.huntingtonswa.org.au/	Yes
	The International Huntington Association	https://huntington-disease.org/	No
	Huntingtons Queensland	https://huntingtonsqld.org.au/	No
Motor neurone disease (MND)	Motor Neurone Disease Association	https://www.mndassociation.org/	Yes
	A Life Story Foundation	https://www.alifestoryfoundation.org/	Yes
	MND Australia	https://www.mndaust.asn.au/Home	Yes
	MND Victoria	https://www.mnd.asn.au/	Yes
	International Alliance of ALS/MND Associations	https://www.alsmndalliance.org/	Yes

	MND NSW	https://www.mndnsw.asn.au/	No
Multiple sclerosis (MS)	MS	https://www.ms.org.au/	Yes
	MS Research Australia	https://msra.org.au/	Yes
	National Multiple Sclerosis Society	https://www.nationalmssociety.org/	Yes
	Multiple Sclerosis New Zealand	https://www.msnz.org.nz/	Yes
	The MS Society	https://mssociety.ca/	Yes
	MS Australia	https://www.msaustralia.org.au/	Yes
	MSWA	https://www.mswa.org.au	Yes
	The Multiple Sclerosis Society of South Australia and Northern Territory	https://www.ms.asn.au/	Yes
	MS Society	https://www.mssociety.org.uk/	Yes
	MS International Federation	https://www.msif.org/	No
Parkinson's disease	Parkinson's Foundation	https://www.parkinson.org/	Yes
	International Parkinson and Movement Disorder Society	https://www.movementdisorders.org/MDS.htm	Yes
	American Parkinson Disease Association	https://www.apdaparkinson.org/	Yes
	European Parkinson's Disease Association	https://www.epda.eu.com/	Yes
	Parkinson's NSW	https://www.parkinsonsnsw.org.au/	Yes
	The Michael J. Fox Foundation	https://www.michaeljfox.org/	No
	Parkinson's Australia	https://www.parkinsons.org.au/	No
	ParkinsonNet	https://www.parkinsonnet.com/	No
	NeuroWellness	https://www.parkinsonsinternational.com/	No
Stroke	American Stroke Association	https://www.stroke.org/	Yes
	Stroke Foundation	https://strokefoundation.org.au/	Yes
	Stroke Association	https://www.stroke.org.uk/	Yes
	American Heart Association	https://www.heart.org/	Yes
	American Stroke Foundation	https://americanstroke.org/	No
	Stroke Foundation NZ	https://www.stroke.org.nz/	No
	Stroke SA	https://www.stroke.org.au/	No
	World Stroke Organization	https://www.world-stroke.org/	No
	Safe Implementation of Treatments in Stroke	https://www.sitsinternational.org/	No
	Stroke Care International	https://www.strokecareinternational.org/	No

Table S4 Studies excluded at full text stage of screening and reasons for exclusion

Citation	Reason for exclusion
1. Almeida CS, Stanich P, Salvioni CC, et al. Assessment and nutrition education in patients with amyotrophic lateral sclerosis. *Arquivos de Neuro-Psiquiatria* 2016; 74: 902-908. DOI: 10.1590/0004-282X20160145.	No intervention details (unclear if individualised)
2. Arshia H and Jeyaraj SS. Assessment of nutritional status and Expanded Disability Status Scale in women with multiple sclerosis in Chennai (South India) and the impact of a nutrition education program. *Multiple Sclerosis and Related Disorders* 2018; 26: 253. DOI: 10.1016/j.msard.2018.10.074.	Abstract or poster
3. Becker H, Stuifbergen A, Taxis C, et al. The use of goal attainment scaling to facilitate and assess individualized change in a wellness intervention for women with fibromyalgia syndrome. *Journal of Holistic Nursing* 2009; 27: 232-240.	Less than half of the program relates to diet
4. Block P, Skeels SE, Keys CB, et al. Shake-It-Up: Health promotion and capacity building for people with spinal cord injuries and related neurological disabilities. *Disability & Rehabilitation* 2005; 27: 185-190.	Less than half of the program relates to diet
5. Chang C-C, Wykle ML and Madigan EA. The effect of a feeding skills training program for nursing assistants who feed dementia patients in Taiwanese nursing homes. *Geriatric Nursing* 2006; 27: 229-237. DOI: 10.1016/j.gerinurse.2006.03.007.	Treatment focus (not education)
6. Gross B, Anderson EF, Busby S, et al. Using culturally sensitive education to improve adherence with anti-hypertension regimen. *Journal of Cultural Diversity* 2013; 20: 75-79.	No neurological disease/s of interest
7. Harris L, Hankey C, Jones N, et al. A cluster randomised control trial of a multi-component weight management programme for adults with intellectual disabilities and obesity. *British Journal of Nutrition* 2017; 118: 229-240. DOI: 10.1017/S0007114517001933	No neurological disease/s of interest
8. Hill VA, Vickrey BG, Cheng EM, et al. A pilot trial of a lifestyle intervention for stroke survivors: Design of healthy eating and lifestyle after stroke (HEALS). *Journal of Stroke & Cerebrovascular Diseases* 2017; 26: 2806-2813. DOI: 10.1016/j.jstrokecerebrovasdis.2017.06.058	Informative article or protocol paper
9. Ifejika NL, Noser EA, Grotta JC, et al. Swipe out Stroke: Feasibility and efficacy of using a smart-phone based mobile	Informative article or protocol paper

application to improve compliance with weight loss in obese minority stroke patients and their carers. *International Journal of Stroke* 2016; 11: 593-603. DOI: 10.1177/1747493016631557.	
10. Kim H and Kim O. The lifestyle modification coaching program for secondary stroke prevention. *Journal of Korean Academy of Nursing* 2013; 43: 331-340.. DOI: 10.4040/jkan.2013.43.3.331.	Full text not in English
11. Kim J-I, Lee S and Kim J-H. Effects of a web-based stroke education program on recurrence prevention behaviors among stroke patients: A pilot study. *Health Education Research* 2013; 28: 488-501.	Less than half of the program relates to diet
12. Larsen G. *Dietary outcomes from the V-STOP Stroke Program*. Dissertation. M.S., Texas Woman's University, Ann Arbor, 2012.	Cannot extract neurological disease/s of interest
13. Marck CH, De Livera AM, Brown CR, et al. Health outcomes and adherence to a healthy lifestyle after a multimodal intervention in people with multiple sclerosis: three year follow-up. *PLoS ONE* 2018; 13: e0197759.	Less than half of the program relates to diet
14. Nordvik I, Myhr KM, Nyland H, et al. Effect of dietary advice and n-3 supplementation in newly diagnosed MS patients. *Acta Neurologica Scandinavica* 2000; 102: 143-149. DOI: 10.1034/j.1600-0404.2000.102003143.x.	Supplementation trial
15. Rashvand F, Abtahi M, Moshtagh Eshgh Z, et al. Improvement in activity of daily living and fatigue in multiple sclerosis patients: the impact of nutrition education. *Nursing and Midwifery Studies* 2016; 5. DOI: 10.17795/nmsjournal32862.	No outcomes of interest
16. Riemann-Lorenz K, Eilers M, Schulz KH, et al. Multiple sclerosis and diet: systematic review, internet-based survey and pilot-testing of an evidence based patient education programme. *Multiple Sclerosis* 2016; 22: 390. DOI: 10.1177/1352458516663081.	Abstract or poster
17. Rimmer JH and Hedman G. A health promotion program for stroke survivors. *Topics in Stroke Rehabilitation* 1998; 5: 30-44.	Informative article or protocol paper
18. Sabour H, Javidan AN, Soltani Z, et al. The effect of behavioral intervention and nutrition education program on serum lipid profile, body weight and blood pressure in Iranian individuals with spinal cord injury: A randomized clinical trial. *Journal of Spinal Cord Medicine* 2018; 41: 28-35. DOI: 10.1080/10790268.2016.1209890.	No neurological disease/s of interest
19. Sajatovic M, Tatsuoka C, Welter E, et al. A targeted self-management approach for reducing stroke risk factors in African	Less than half of the program relates to diet

American men who have had a stroke or transient ischemic attack. *American Journal of Health Promotion* 2018; 32: 282-293.	
20. Sakakibara BM, Lear SA, Barr SI, et al. Development of a chronic disease management program for stroke survivors using intervention mapping: The Stroke Coach. *Archives of Physical Medicine & Rehabilitation* 2017; 98: 1195-1202.	Informative article or protocol paper
21. Suominen MH, Kivisto SM and Pitkala KH. The effects of nutrition education on professionals' practice and on the nutrition of aged residents in dementia wards. *European Journal of Clinical Nutrition* 2007; 61: 1226-1232. DOI: 10.1038/sj.ejcn.1602639.	Treatment focus (not education)
22. Suominen MH, Puranen TM, Jyvakorpi SK, et al. Nutritional guidance improves nutrient intake and quality of life, and may prevent falls in aged persons with alzheimer disease living with a spouse (NuAD Trial). *Journal of Nutrition, Health & Aging* 2015; 19: 901-907.	Individual tailored nutrition advice
23. Teuschl Y, Matz K, Firlinger B, et al. Preventive effects of multiple domain interventions on lifestyle and risk factor changes in stroke survivors: evidence from a two-year randomized trial. *International Journal of Stroke* 2017; 12: 976-984. DOI: 10.1177/1747493017702662.	Less than half of the program relates to diet
24. Wallace R, Lo J and Devine A. Tailored nutrition education in the elderly can lead to sustained dietary behaviour change. *Journal of Nutrition, Health & Aging* 2016; 20: 8-15. DOI: 10.1007/s12603-016-0669-2.	No neurological disease/s of interest
25. Weber B, Bersch-Ferreira ÂC, Torreglosa CR, et al. Implementation of a Brazilian Cardioprotective Nutritional (BALANCE) Program for improvement on quality of diet and secondary prevention of cardiovascular events: A randomized, multicenter trial. *American Heart Journal* 2019; 215: 187-197. DOI: 10.1016/j.ahj.2019.06.010.	Cannot extract neurological disease/s of interest
26. Wong YY. The effectiveness of an educational intervention on managing feeding difficulties for residents with dementia. *Dissertation Abstracts International Section A: Humanities and Social Sciences* 2018; 79.	Treatment focus (not education)

Chapter 4. Effective emotional wellness programs for adults with multiple sclerosis

Thesis objective addressed in this chapter:

Objective 2: To determine the characteristics of effective education programs for people with MS, namely emotional wellness programs.

The content of this chapter is covered by Publication 2:

Russell, R. D., Black, L. J., Pham, N. M., & Begley, A. The effectiveness of emotional wellness programs on mental health outcomes for adults with multiple sclerosis: a systematic review and meta-analysis. *Mult Scler Relat Disord*. 2020;44:102171. https://doi.org/10.1016/j.msard.2020.102171

The version that appears in this thesis is of an article that has been through peer-review with *Multiple Sclerosis and Related Disorders* but has not been through the copyediting process.

The contribution of co-authors, A/Prof Andrea Begley, A/Prof Lucinda Black, and Dr Minh Pham are detailed in the author attribution statements in Appendix A: Attribution statements.

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Review: Multiple Sclerosis and Related Disorders

The effectiveness of emotional wellness programs on mental health outcomes for adults with multiple sclerosis: a systematic review and meta-analysis

R. D. Russell[a], L. J. Black[a] , N. M. Pham[a,b], A. Begley[a]

Affiliations:

[a]School of Public Health, Curtin University, Perth, Australia

[b]Thai Nguyen University of Medicine and Pharmacy, Thai Nguyen, Vietnam

Corresponding Author:

Andrea Begley

School of Public Health, Kent St, Bentley WA 6102

a.begley@curtin.edu.au

+61 8 9266 2773

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Conflicts of interest

The authors declare no conflict of interest.

Author contributions

R. D. Russell: Conceptualization, Formal analysis, Writing - Original Draft, Visualization.

L. J. Black: Conceptualization, Supervision, Writing - Review & Editing.

N. M. Pham: Formal analysis, Writing - Review & Editing

A. Begley: Conceptualization, Supervision, Writing - Review & Editing, Validation

Keywords: Anxiety; behavior change techniques; depression; emotional wellness; quality of life; stress

Highlights

- Mental health outcomes in MS were improved by emotional wellness programs
- Emotional wellness programs were most effective at reducing stress (large effect)
- Emotional wellness programs were moderately effective for depression and anxiety
- Behaviour practice and social comparison were common behaviour change techniques

Abstract

Background: People with multiple sclerosis (MS) have a greater prevalence of depression and anxiety than the general population. Emotional wellness programs (any psychological or psychosocial interventions that focus on awareness, acceptance, managing, or challenging thoughts and feelings) could be important for people with MS. However, there have been no reviews on the effectiveness of emotional wellness programs for people with MS. The objective of this review was to determine the effectiveness of emotional wellness programs on mental health outcomes for adults with MS.

Inclusion criteria: Randomised controlled trials (RCTs) and quasi-experimental trials evaluating emotional wellness programs for adults with any form of MS were included. Mental health outcomes included were depression, anxiety, quality of life, and stress. The comparator groups were waitlist controls, usual care, or another intervention.

Methods: This review was registered with PROSPERO (registration number CRD42019131082) and conducted in accordance with PRISMA guidelines. CINAHL, Cochrane, MEDLINE, PsycInfo, Web of Science, ProQuest Dissertations and Theses, Cochrane register of Controlled Trials, and Google Scholar were searched for English-language publications. Titles and abstracts were initially screened, followed by a screen of full text articles. Studies were critically appraised for methodological quality using the JBI standardised critical appraisal checklists. Data were extracted on intervention details, study outcome measures, behaviour change techniques, and results. Random effects meta-analyses were performed for outcomes assessed in at least five studies, with results reported as the standardised mean difference (SMD).

Results: This review comprised 25 RCTs and four quasi-experimental studies (n participants=2323); 21 were included in meta-analyses. Meta-analyses produced statistically significant results favouring the interventions (SMD (95% CI) for depression -0.55 (-0.87, -0.24); anxiety -0.42 (-0.70, -0.14); quality of life 0.28 (0.14, 0.43); and stress -1.00 (-1.58, -0.43)). The most commonly used behaviour change techniques were behaviour practice/rehearsal, social comparison, and social support.

Conclusions: This review provides evidence to support the effectiveness of emotional wellness programs for improving mental health outcomes in adults with MS. However, these findings should be interpreted with caution given the high degree of heterogeneity between the studies, and potential for biases in analysis due to missing data and/or incomplete reporting.

1. Introduction

The prevalence of depression and anxiety is greater among people with MS (pwMS) than in the general population.[1] These mental health co-morbidities are underdiagnosed and undertreated in pwMS,[2] impacting on quality of life.[3] These co-morbidities impose limitations on daily life activities[4] and are strongly associated with fatigue,[5] which is described as the most common and disabling symptom of MS.[6] According to a recent systematic review, higher levels of stress (as measured by basal cortisol levels) may be associated with depression, anxiety, and MS progression.[7] Given the relationship between mental health and quality of life, interventions that address depression and anxiety may reasonably improve quality of life for pwMS.[3]

Wellness is a high priority for pwMS,[8] and may enhance health-related quality of life.[9] There is interest from pwMS in learning how to manage their MS with diet and exercise, and to develop strategies to manage depression and other mood changes to achieve emotional wellness,[8] i.e. the ability to manage and adapt to stresses and difficult circumstances in one's life.[10] Given this need, the United States National MS Society established the Wellness Research Working Group, which has defined three approaches for wellness in MS: diet, exercise, and emotional wellness.[11] Determining the effectiveness of these approaches has been identified as areas of future research priority.[8] Effective education programs employ a number of recognised techniques to support change in the targeted behaviours, as identified by Michie et al. in their 93-item behaviour change technique (BCT) taxonomy.[12] Identifying which BCTs are used in emotional wellness programs for pwMS could help characterise elements of effective programs. This review will focus on emotional wellness programs, defined as any psychological (e.g. cognitive behavioural therapy) or psychosocial (e.g. supportive group interactions or non-directive counselling) interventions that focus on awareness, acceptance, managing, or changing/challenging thoughts and feelings, including feelings of depression, anxiety, and stress.[13] Such programs (including those using cognitive behaviour therapy[14] and mindfulness techniques[15 16]) have been reported as effective for improving mental health in pwMS.

To our knowledge, there have been no systematic reviews focusing solely on the effectiveness of emotional wellness programs for pwMS. Several reviews have examined

self-management interventions or strategies for pwMS (skills to manage the daily emotional, physical, and social aspects of living with a chronic condition);[17-19] wellness interventions (nutrition, exercise, and emotional wellness, for people with progressive MS,[20] and people with chronic disabling conditions including MS[21]); mindfulness;[22] and stress-management.[23] Overall, accumulating evidence from reviews supports such interventions for improving mental health; however, it is difficult to make definitive conclusions due to the small number of included studies and methodological heterogeneity. Furthermore, identification of BCTs used in this field is lacking. The primary objective of this review was to determine the effectiveness of emotional wellness programs on mental health outcomes (depression, anxiety, quality of life, and stress) for adults with MS. The secondary objective was to assess BCTs used in emotional wellness programs for adults with MS.

2. Methods

This systematic review was carried out according to an *a priori* protocol (registration number: PROSPERO CRD42019131082), in accordance with the Joanna Briggs Institute (JBI) methodology for systematic reviews of effectiveness.[24]

2.1 Inclusion criteria

This review considered studies involving adults with a clinical diagnosis of MS. The included interventions were emotional wellness programs (any structured psychological or psychosocial interventions) running for more than one session. The interventions were in any format (in-person, online, or via telephone), and individual or group-based. To be eligible for inclusion, content/topics of programs must have been standardised for all participants (i.e. individualised programs were excluded). Programs based on exercise or diet were excluded. Eligible comparators were: waitlist control group, usual care comparator group (no intervention), or another intervention. Outcomes of interest were depression, anxiety, quality of life, and stress. This review included quantitative studies (randomized controlled trials (RCTs) and quasi-experimental trials) published in the English language.

2.2 Search strategy

A three-step search strategy was adopted following JBI guidelines. In brief, an initial search limited to MEDLINE and CINAHL was undertaken to identify articles (Appendix A), followed by a full search strategy. The search was conducted in April 2019 and updated in

September 2019. No limitations were applied based on publication date. To account for differences in Medical Subject Headings (MeSH) terms and Boolean operators, the search strategy was adapted for each information source. For published literature, we searched CINAHL, Cochrane, MEDLINE, PsycInfo, and Web of Science; for unpublished studies and grey literature, we searched Cochrane Central Register of Controlled Trials, ProQuest Dissertations and Theses, and Google Scholar. Reference lists of all included studies and were screened for additional studies.

2.3 Study selection

All citations were uploaded into EndNote X9 (Clarivate Analytics, PA, USA). Titles and abstracts were screened by RDR. Potentially relevant studies were imported into the JBI System for the Unified Management, Assessment and Review of Information (JBI SUMARI) (2019, Joanna Briggs Institute, Adelaide, Australia). Two independent reviewers (AB and RDR) screened full text articles for final inclusion. Any disagreements between the reviewers were resolved through discussion.

2.4 Assessment of methodological quality

The first author (RDR) assessed methodological quality using the JBI critical appraisal checklists for quasi-experimental trials and RCTs.[24] For a study to receive a positive ('yes') rating for each question, the required information had to be clearly stated in the article. If the reporting was vague, the item was rated as 'unclear'. If reporting was insufficient, the study received a poor ('no') rating. Studies scoring less than 50% overall were excluded from statistical synthesis due to poor methodological quality, but were included in the narrative review.

2.5 Data extraction

The following data were extracted: aim, study characteristics (authors, year, country), participant details (type of MS, sample size, age, sex, duration of MS), intervention details (type, number of study arms, description of intervention, type of comparator group, duration and number of sessions, delivery method), BCTs (classified according to the BCT taxonomy by Michie and colleagues[12]), behaviour change theory used, tools used to measure outcomes (Appendix B), and results. Authors were contacted to request missing data, and a second request was sent four weeks later, where required. Missing post-intervention standard deviation (SD) scores were calculated using confidence interval (CI) values with the following formula (sample sizes were less than 60):

SD = $\sqrt{n}$ x (upper limit CI - lower limit CI)/t value

t values were obtained by entering =TINV(1-0.95,n-1) into a Microsoft Excel spreadsheet.[25]

2.6 Data synthesis and meta-analysis

Data were pooled with statistical meta-analysis using JBI SUMARI (2019, Joanna Briggs Institute, Adelaide, Australia). Effect sizes were expressed as post-intervention standardized mean differences (SMDs), and their 95% confidence intervals (CIs). An SMD of 0.2 = small effect size; 0.5 = medium; and 0.8 = large.[26] Statistical analyses were performed using a random effects meta-analysis regression model with inverse variance. Statistical heterogeneity was assessed using the standard chi-squared test (Cochran Q test; $P < 0.10$ signified significant heterogeneity[27]), and the I^2 index (where 25%, 50%, and 75% indicated low, moderate, and high degrees of heterogeneity, respectively[28]). Subgroup analyses were conducted as follows: intervention duration (eight weeks or more); method of delivery (in-person); comparator type (waitlist control); and intervention type (mindfulness only). Using meta-regression, we investigated potential predictors to explain high degrees of heterogeneity for outcomes with at least ten studies (depression and anxiety).[29] For each outcome, the following covariates were included in a single meta-regression model: mean participant age (years), mean time since diagnosis (years), percentage of females, in-person intervention (*vs.* teleconference/videoconference), minimum eight week intervention (*vs.* less than eight weeks), studies with waitlist comparators (*vs.* active comparators), and mindfulness intervention (*vs.* other). To test for publication bias, funnel plots were generated, and the Egger's test for asymmetry (where $P<0.05$ indicates bias) using the "trim and fill" method was performed for outcomes with at least ten studies[30] (depression, anxiety, and quality of life). Stata software (StataCorp, College Station, TX, USA) was used for meta-regression analyses and tests of publication bias.

3. Results

3.1 Search results

Database searches retrieved 9168 articles. Once duplicates were removed, 6839 articles were screened by title and abstract. Full text articles were accessed for the remaining studies, and 69 were excluded (Appendix C). We included 29 studies in the narrative review, with 21 studies included in the meta-analyses (16 reporting depression; 16 anxiety;

12 quality of life; and 7 stress) (**Figure 1**). Eight studies were not used in meta-analyses for the following reasons: three studies reported median and interquartile range (IQR) instead of mean and standard deviation (SD);[31-33] three scored too low on assessment of methodological quality;[34-36] and two had missing data.[37, 38]

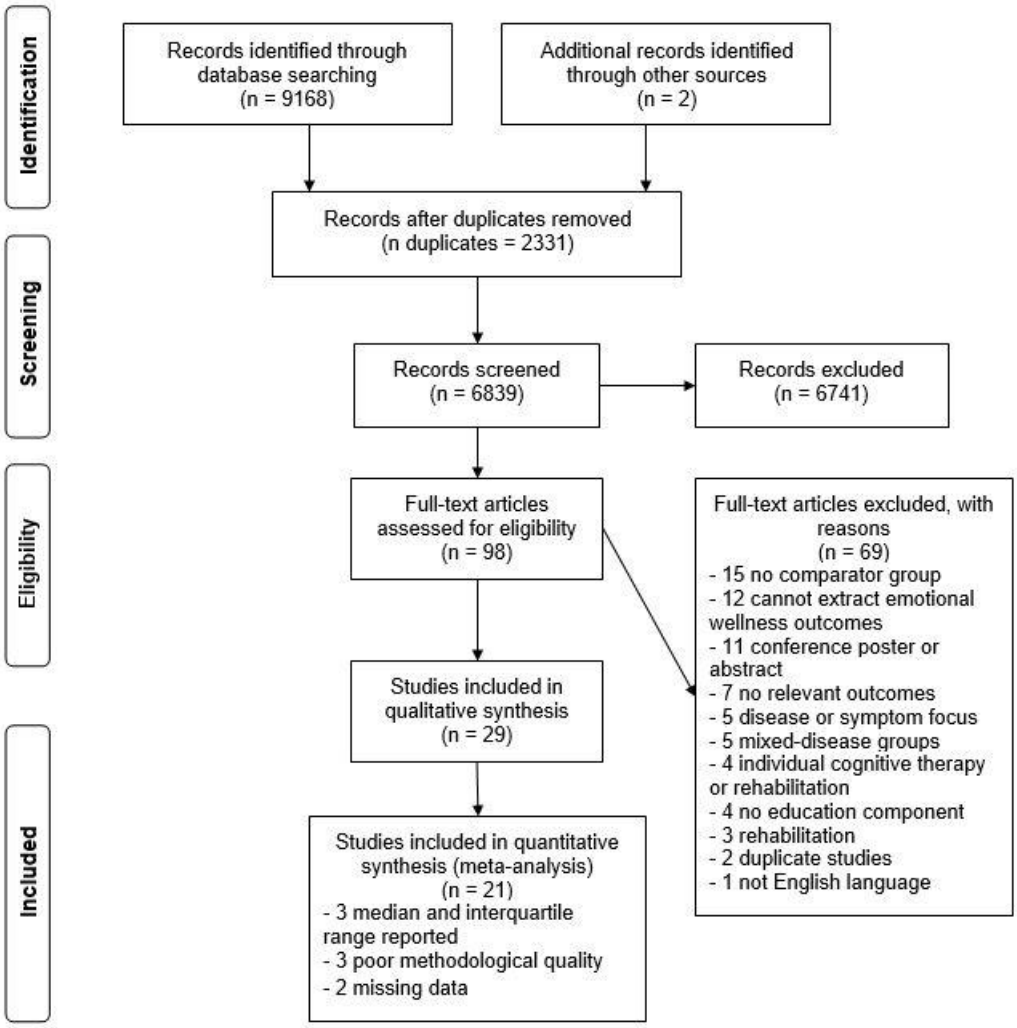

Figure 1 PRISMA flowchart of article screening process[39]

3.2 Methodological quality and publication bias

Studies were appraised for methodological quality using the JBI critical appraisal checklists for quasi-experimental studies and RCTs.[24] Four studies were quasi-experimental trials[40-43] (**Table 1**), and the remaining studies were RCTs[14-16, 31-38, 44-57] (**Table 2**). None of the quasi-experimental trials included multiple measurements of the outcome both pre- and post- intervention (Q5, **Table 1**), and only two trials stated the reliability of the tools.[40, 42]

Table 1 Assessment of methodological quality for quasi-experimental studies

Study	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Score %
Calandri *et al.*[40]	Y	Y	Y	Y	N	Y	Y	Y	Y	**89**
Crescentini *et al.*[41]	Y	Y	Y	Y	N	N	Y	U	Y	**67**
Hoogerwerf *et al.*[42]	Y	Y	Y	Y	N	Y	Y	Y	Y	**89**
Tesar *et al.*[43]	Y	Y	Y	Y	N	Y	Y	U	Y	**78**
Total %	100	100	100	100	0	75	100	50	100	

Y, yes; N, no; U, unclear.
JBI critical appraisal checklist for quasi-experimental studies: Q1: Is it clear in the study what is the 'cause' and what is the 'effect' (i.e. there is no confusion about which variable comes first)?; Q2: Were the participants included in any comparisons similar?; Q3: Were the participants included in any comparisons receiving similar treatment/care, other than the exposure or intervention of interest?; Q4: Was there a control group?; Q5: Were there multiple measurements of the outcome both pre and post the intervention/exposure?; Q6: Was follow up complete and if not, were differences between groups in terms of their follow up adequately described and analysed?; Q7: Were the outcomes of participants included in any comparisons measured in the same way?; Q8: Were outcomes measured in a reliable way?; Q9: Was appropriate statistical analysis used?

After contacting authors for missing data, three RCTs[34-36] were excluded due to scoring less than 50% overall (Appendix D). The excluded studies did not report randomization, allocation concealment, blinding of outcome assessors, or potential differences between completers and drop-out participants. Blinding of those delivering the interventions was not possible in any of the studies. Participant blinding was achieved in only one study: Ehde and colleagues[48] informed participants that both the self-management intervention and the comparator educational program were equivalent treatments as a way of blinding to the intervention. Seventeen studies either did not adequately report whether follow-up was complete, or did not describe differences between groups in relation to drop-outs.[15, 16, 32, 33, 36-38, 44, 47-51, 53-55, 57]

Table 2 Assessment of methodological quality for experimental studies

Study	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10	Q11	Q12	Q13	Score %
Alschuler *et al.*[44]	Y	Y	Y	N	N	U	Y	Y	Y	Y	Y	Y	Y	**77**
Amiri *et al.*[38]	U	U	Y	N	U	U	Y	Y	Y	Y	Y	Y	Y	**61**
Bahrani *et al.*[45]	Y	Y	Y	N	N	Y	Y	N	Y	Y	Y	Y	Y	**77**
Barlow *et al.*[46]	Y	Y	Y	N	N	U	Y	N	Y	Y	Y	Y	Y	**69**
Bogosian *et al.*[47]	Y	Y	Y	N	N	Y	Y	Y	Y	Y	U	Y	Y	**77**
Cavalera *et al.*[16]	Y	Y	Y	U	N	U	Y	Y	Y	Y	U	Y	Y	**69**
das Nair *et al.*[31]	Y	Y	Y	N	N	Y	Y	N	Y	Y	U	Y	Y	**69**
Ehde *et al.*[48]	Y	Y	Y	Y	N	Y	Y	Y	Y	Y	U	Y	Y	**85**
Ennis *et al.*[49]	Y	Y	Y	N	N	Y	Y	Y	Y	Y	U	Y	Y	**77**

Forman *et al.*[32]	Y	Y	Y	N	N	Y	Y	Y	Y	Y	Y	Y	Y	**85**
Grossman *et al.*[50]	Y	Y	Y	N	N	Y	Y	Y	Y	Y	Y	Y	Y	**85**
Graziano *et al.*[51]	U	Y	Y	N	N	U	Y	Y	Y	Y	Y	Y	Y	**69**
Kolahkaj *et al.*[52]	Y	Y	Y	N	N	U	Y	N	Y	Y	Y	Y	Y	**69**
Lincoln *et al.*[53]	Y	Y	Y	N	N	Y	Y	Y	Y	Y	U	Y	Y	**77**
Nordin *et al.*[33]	Y	Y	Y	N	N	Y	Y	Y	Y	Y	U	Y	Y	**77**
Pahlavanzadeh *et al.*[14]	Y	U	Y	N	N	N	Y	N	Y	Y	Y	Y	Y	**61**
Sanaeinasab *et al.*[54]	U	U	Y	N	N	U	Y	Y	Y	Y	U	Y	Y	**54**
Schwartz *et al.*[37]	U	U	Y	N	N	U	Y	Y	Y	Y	U	Y	Y	**54**
Senders *et al.*[55]	Y	Y	Y	N	N	Y	Y	Y	Y	Y	Y	Y	Y	**85**
Shahdadi *et al.*[56]	Y	Y	Y	N	N	U	U	N	Y	Y	Y	U	Y	**54**
Simpson *et al.*[15]	Y	Y	Y	N	N	Y	Y	Y	Y	Y	Y	Y	Y	**85**
Stuifbergen *et al.*[57]	Y	Y	U	N	N	U	Y	Y	Y	Y	Y	Y	Y	**69**
Excluded studies														
Haji-Adineh *et al.*[36]	N	U	U	N	N	U	Y	Y	Y	Y	Y	Y	N	**46**
Khayeri *et al.*[34]	U	U	Y	N	N	U	U	U	Y	Y	Y	Y	Y	**46**
Rigby *et al.*[35]	U	U	Y	N	N	U	Y	N	Y	Y	U	Y	Y	**46**
Total %	72	72	92	4	0	44	92	68	100	100	60	96	96	

Y, yes; N, no; U, unclear.
Q1: Was true randomization used for assignment of participants to treatment groups?; Q2: Was allocation to groups concealed?; Q3: Were treatment groups similar at the baseline?; Q4: Were participants blind to treatment assignment?; Q5: Were those delivering treatment blind to treatment assignment?; Q6: Were outcomes assessors blind to treatment assignment?; Q7: Were treatment groups treated identically other than the intervention of interest?; Q8: Was follow up complete and if not, were differences between groups in terms of their follow up adequately described and analysed?; Q9: Were participants analysed in the groups to which they were randomized?; Q10: Were outcomes measured in the same way for treatment groups?; Q11: Were outcomes measured in a reliable way?; Q12: Was appropriate statistical analysis used?; Q13: Was the trial design appropriate for the topic, and any deviations from the standard RCT design accounted for in the conduct and analysis?

Figures 2A and **2B** suggest the presence of publication bias for depression and anxiety (Egger's *P*=0.02, and 0.04, respectively). We undertook sensitivity analyses using the trim and fill method:[58] the resulting funnel plots were asymmetrical, indicating the potential presence of publication bias (**Figures 2C** and **2D**). Publication bias was not evident for quality of life (Egger's *P*=0.29) (**Figure 3**).

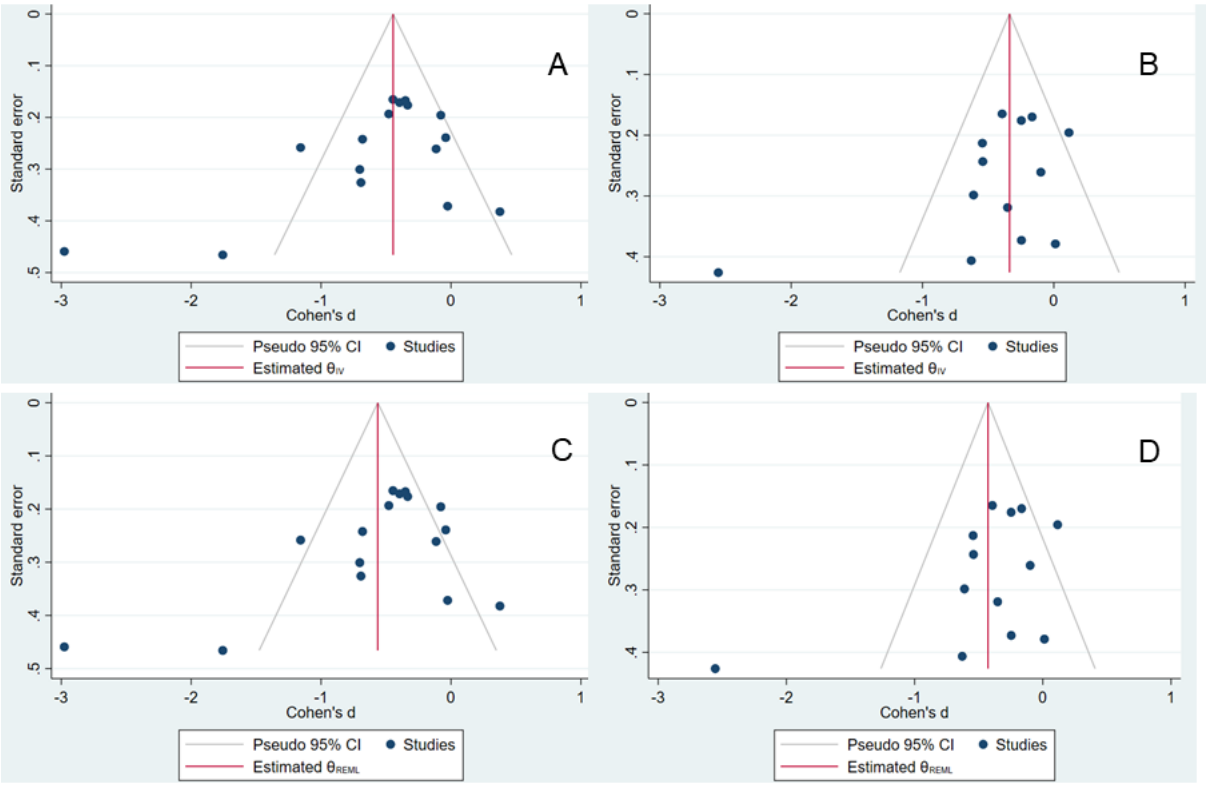

Figure 2 Funnel plots for depression and anxiety without trim and fill (A and B, respectively), and with trim and fill (C and D, respectively)

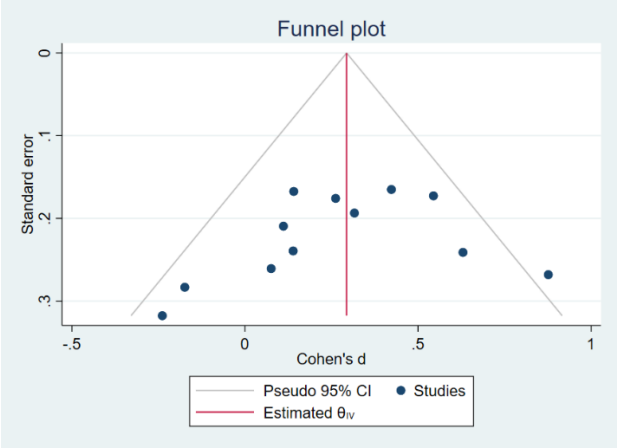

Figure 3 Funnel plot for quality of life, without trim and fill

3.3 Study characteristics

Studies included in this review were conducted in Iran,[14, 34, 36, 38, 45, 52, 54, 56] the United Kingdom,[15, 31, 32, 35, 46, 47, 49, 53] the United States,[37, 44, 48, 55, 57] Italy,[16, 40, 51] Austria,[43] the Netherlands,[42] Sweden,[33] and Switzerland.[50] The majority compared the intervention to a treatment as usual group[14, 34, 36, 38, 41, 43, 45, 50, 52, 54, 56] or a waitlist control group.[15, 32, 40, 42, 44, 46, 47, 49, 53, 57] The remaining studies used other programs or information sessions as the comparators.[16, 31, 33, 35, 37, 48, 51] Two-thirds of the included studies reported power calculations or adequately justified the sample size.[14, 15, 34-37, 42, 44, 45, 47, 48, 50, 52, 53, 55-57]

However, four were underpowered at post-intervention analysis due to drop-outs,[14, 44, 53, 56] and one did not state if all participants completed the trial.[34] Consequently, less than half of the studies reported sufficient power to detect intervention effects. See Appendix E for characteristics of included studies.

3.4 Participant characteristics

Baseline data were collected from 2323 participants (*n* intervention=1142; *n* comparator=1181). Data were missing from nine studies: eight did not report participant disease duration;[14, 32, 34, 36, 38, 48, 51, 52] one did not report participant age;[14] mean age was not available for one study;[52] and one did not report sex.[53] From the studies with complete data, the median (IQR) number of participants in the intervention and comparator groups was 35 (40.5) and 31 (46.0), respectively. The mean (SD) age was 43.7 (7.6) years for participants in the intervention groups, and 44.1 (7.9) years for participants in the comparator groups. Mean (SD) disease duration was 9.0 (3.9) and 9.7 (4.4) years in the intervention and comparator groups, respectively. Participants were mostly female in both the intervention (77%) and comparator groups (76%). The majority of the studies included participants with all types of MS;[14-16, 31, 32, 34-36, 38, 40, 41, 43-46, 51-57] five included participants with only relapsing-remitting and progressive MS;[33, 37, 42, 48, 50] and one included participants with only progressive MS.[47] Seven studies did not report MS type.[34, 35, 43, 46, 52, 54, 56]

3.5 Intervention characteristics

Intervention programs were based on the following concepts according to their authors: mindfulness,[15, 16, 36, 38, 41, 42, 45, 47, 50, 52, 55] adjustment to MS,[31, 32, 53] cognitive behavioural principles,[14, 40, 51] other psychological therapies,[33-35, 43, 44] coping skills,[37, 54] self-management,[46, 48] health promotion/wellness,[49, 57] and self-care.[56] The duration of sessions ranged from 45 minutes[33, 56] up to three hours.[49, 57] The shortest regular session lasted 45-60 minutes per session,[48] and the longest lasted three hours per session.[49] Two interventions included a day-long retreat mid-way through the program, lasting six[55] or seven[50] hours. One intervention did not report session duration.[33] The shortest intervention lasted two weeks[56] and the longest was 15 weeks.[33] The total number of sessions ranged from three[35] to nine;[56] eight sessions was the most common.[14-16, 34, 36-38, 40-42, 45, 47, 49, 50, 52, 55, 57] The majority of interventions ran once a week[14-16, 35-38, 41, 43-50, 52, 54, 55, 57] or once a fortnight.[31, 32, 51, 53] Nearly all of the interventions were conducted in group settings[14-16, 32-38, 40-47, 49-55, 57] and nearly all of the interventions were in-person.[14, 15, 31-38, 40-43, 45, 46, 49-55, 57]

Two interventions were individual programs using standardised content/topics,[31, 48] and delivery method was not specified in one study.[56] Two programs were telephone-based,[44, 48] and one was conducted via videoconference.[47]

3.6 Behaviour change techniques and theories

There were sufficient details in 28 studies to code BCTs (one study did not provide any intervention information[56] so BCTs could not be assigned). Of the 93 different BCTs, a total of 37 were used across the interventions (**Table 3**). The mean number of BCTs used was eight (range, four to 18). The most commonly used BCTs were: behaviour practice/rehearsal (25 studies[14-16, 31-34, 36-38, 40-43, 45-55]); social comparison (17 studies[14-16, 32, 35-37, 40, 43-45, 49, 51, 53-55, 57]); social support (unspecified) (15 studies[16, 31, 32, 34, 35, 37, 38, 40, 44-46, 49, 51, 53, 55]); credible source, i.e. program facilitated by an accredited, relevant health professional (14 studies[15, 16, 33, 35, 37, 40, 42, 44, 45, 47, 49, 51, 52, 57]); and reduce negative emotions (14 studies[14, 15, 34, 37, 38, 40, 41, 43-45, 48, 51, 55, 57]).

Table 3 Behaviour change techniques used in included studies

	Alschulaer *et al.*	Amiri *et al.*	Bahrani *et al.*	Barlow *et al.*	Bogosian *et al.*	Calandri *et al.*	Cavalera *et al.*	Crescentini *et al.*	das Nair *et al.*	Edhe *et al.*	Ennis *et al.*	Forman *et al.*	Graziano *et al.*	Grossman *et al.*	Haji-Adineh *et al.*	Hoogerwerf *et al.*	Khayeri *et al.*	Kolahkaj *et al.*	Lincoln *et al.*	Nordin *et al.*	Pahlavanzadeh *et al.*	Rigby *et al.*	Sanaeinansab *et al.*	Schwartz *et al.*	Senders *et al.*	Shahdadi *et al.*	Simpson *et al.*	Stuifbergen *et al.*	Tesar *et al.*	Total n (%)
Goal setting (behaviour)																							■	■						2 (6.9)
Problem solving				■		■				■		■	■				■	■	■			■	■				■	■	■	13 (44.8)
Goal setting (outcome)	■		■	■		■				■	■	■	■	■	■				■									■		12 (41.4)
Action planning	■					■				■			■										■					■		6 (20.7)
Review behaviour goal(s)			■	■						■														■						4 (13.8)
Review outcome goal(s)										■				■														■		3 (10.3)
Behavioural contract			■																								■			2 (6.9)
Monitoring behaviour by others without feedback																												■		1 (3.4)
Feedback on behaviour			■							■					■						■						■			5 (17.2)
Self-monitoring behaviour			■	■	■			■				■		■					■	■					■		■	■		11 (37.9)
Self-monitoring outcome(s)		■	■													■		■							■		■			6 (20.7)
Monitoring outcome(s) by		■														■											■			3 (10.3)

others without feedback		■														■											■				
Biofeedback											■																				1 (3.4)
Social support (unspecified)	■	■	■	■		■	■		■		■	■	■				■		■			■		■	■						15 (51.7)
Social support (emotional)					■																			■							2 (6.9)
Instruction how to perform a behaviour		■	■	■	■			■	■						■						■			■	■		■	■	■		13 (44.8)
Information on antecedents								■																							1 (3.4)
Information about health consequences			■	■						■							■							■	■		■		■		8 (27.6)
Information about social and environmental consequences																					■										1 (3.4)
Monitoring emotional consequences																														■	1 (3.4)
Information about emotional consequences			■				■	■										■		■											5 (17.2)
Demonstration of the behaviour			■	■	■	■	■	■				■	■				■		■		■									■	12 (41.4)
Social comparison	■		■			■	■				■	■	■		■				■		■	■	■	■	■		■	■	■	17 (58.6)
Prompts/cues									■	■		■					■		■	■	■	■	■		■		■		■	12 (41.4)
Reduce prompts/cues																	■													1 (3.4)
Behavioural practice/ rehearsal		■	■	■	■	■	■	■	■	■	■	■	■	■	■	■	■	■	■	■	■		■	■	■		■		■	25 (86.2)
Credible source	■		■		■	■	■				■		■			■		■		■		■		■			■	■		14 (48.3)

Non-specific reward	■				■																										2 (6.9)
Reduce negative emotions	■	■	■			■		■		■			■				■				■			■	■		■	■	■		14 (48.3)
Avoidance/ reducing exposure to cues		■			■																			■							3 (10.3)
Adding objects to the environment			■					■													■			■			■				5 (17.2)
Framing/ reframing		■	■			■	■		■	■		■	■		■				■		■			■				■			13 (44.8)
Valued self-identity																				■											1 (3.4)
Verbal persuasion about capability			■	■		■					■		■									■						■	■		8 (27.6)
Mental rehearsal of successful performance											■																				1 (3.4)
Focus on past success			■												■									■					■		4 (13.8)
Self-talk	■	■																										■			3 (10.3)
Total	**8**	**9**	**18**	**10**	**8**	**11**	**7**	**8**	**5**	**11**	**8**	**9**	**11**	**4**	**7**	**4**	**8**	**5**	**9**	**6**	**9**	**6**	**6**	**14**	**9**	**0**	**14**	**13**	**11**		

Five studies reported an underlying behaviour change theory: either cognitive behaviour therapy principles[31, 32, 53] or self-efficacy theory.[46, 57] Of those, two out of four studies reported improvement in depression;[32, 53] one out of three reported improvement in anxiety;[53] and two out of three reported greater quality of life.[53, 57] The studies measuring stress did not report any behaviour change theories.

3.7 Review findings

Results have been grouped according to the outcomes of interest: depression, anxiety, quality of life, and stress. **Table 4** presents a summary of the findings relating to program effectiveness and outcomes.

Table 4: Effectiveness of emotional wellness programs on mental health outcomes

		Evidence of effectiveness	
Outcome	Number of studies	Improvement	No improvement
Depression	25	14[a]	12[b]
Anxiety	21	10[c]	13[b,c]
Quality of life	13	6	7
Stress	8	6	2

[a]Lincoln *et al*[53] reported depression scores from the Beck Depression Inventory and the Hospital Anxiety Depression Scale. Both results are included in the table.
[b]Rigby *et al*[35] evaluated the intervention group against two comparator groups (group one: social discussion group plus booklet; group two: information booklet only). Both comparisons are included in the table.
[b]Crescentini *et al.*[41] reported anxiety from both the state and trait scores of the State-Trait Anxiety Inventory. Both results are included in the table.

3.7.1 Depression

Twenty five studies measured depression.[14-16, 31-38, 40-48, 50-53, 55] Relative to the comparators, 13 studies reported statistically significant improvements in depression scores.[14-16, 32, 34, 36, 38, 42, 45, 47, 50, 52, 53] One study reported an improvement in the comparator group, but only from one of the two tools they used to measure depression.[33] The most frequently used tool to measure depression was the Hospital Anxiety and Depression Scale (HADS; reported in nine studies[16, 31-33, 35, 42, 46, 47, 53]) followed by the Beck Depression Inventory (BDI; seven studies[31, 33, 36, 38, 41, 43, 53]). Three studies[31, 33, 53] reported two measures of depression (BDI and HADS). The most frequently used BCTs were: behavioural practice/rehearsal (23 studies[14-16, 31-34, 36-38, 40-43, 45-48, 50-53, 55]; social comparison (14 studies[14-16, 32, 35-37, 40, 43-45, 51, 53, 55]); and social support (unspecified) (14 studies[16, 31, 32, 34, 35, 37, 38, 40, 44-46, 51, 53, 55]). Of the 13 effective interventions, all used behaviour practice/rehearsal as a BCT, followed by demonstration of the behaviour (seven

studies[14, 16, 32, 34, 45, 47, 53]), social comparison (seven studies[14-16, 32, 36, 45, 53]), and framing/reframing (seven studies[14, 16, 32, 36, 38, 45, 53]).

3.7.2 Depression meta-analysis

Sixteen studies were included in statistical meta-analysis.[14, 16, 40-48, 50-53, 55] One study[53] reported multiple measures of depression (BDI and HADS); the HADS score was included in meta-analysis as this tool was more frequently used by other included studies.[16, 46, 47] Meta-analysis included 1265 participants (629 received the intervention), and resulted in a statistically significant medium effect, favouring the intervention (SMD -0.55; 95% CI -0.87, -0.24; *P*=0.001) (**Figure 4**). Heterogeneity was high (I^2=86%; chi-squared *P*<0.001). Subgroup analysis was performed to examine the robustness of the findings. Overall, there was minimal change in the findings when grouped by: minimum eight-week interventions (SMD -0.59; 95% CI -0.97, -0.21; *P*=0.002; I^2=87%; chi-squared *P*<0.001); in-person interventions (SMD -0.51; 95% CI -0.92, -0.11; *P*=0.013; I^2=88%; chi-squared *P*<0.001); waitlist control/usual care comparators (SMD -0.69; 95%CI -1.12, -0.26; *P*=0.002; I^2=89%; chi-squared *P*<0.001); and mindfulness interventions (SMD -0.63; 95%CI -1.22, -0.04; *P*=0.037; I^2=92%; chi-squared *P*<0.001).

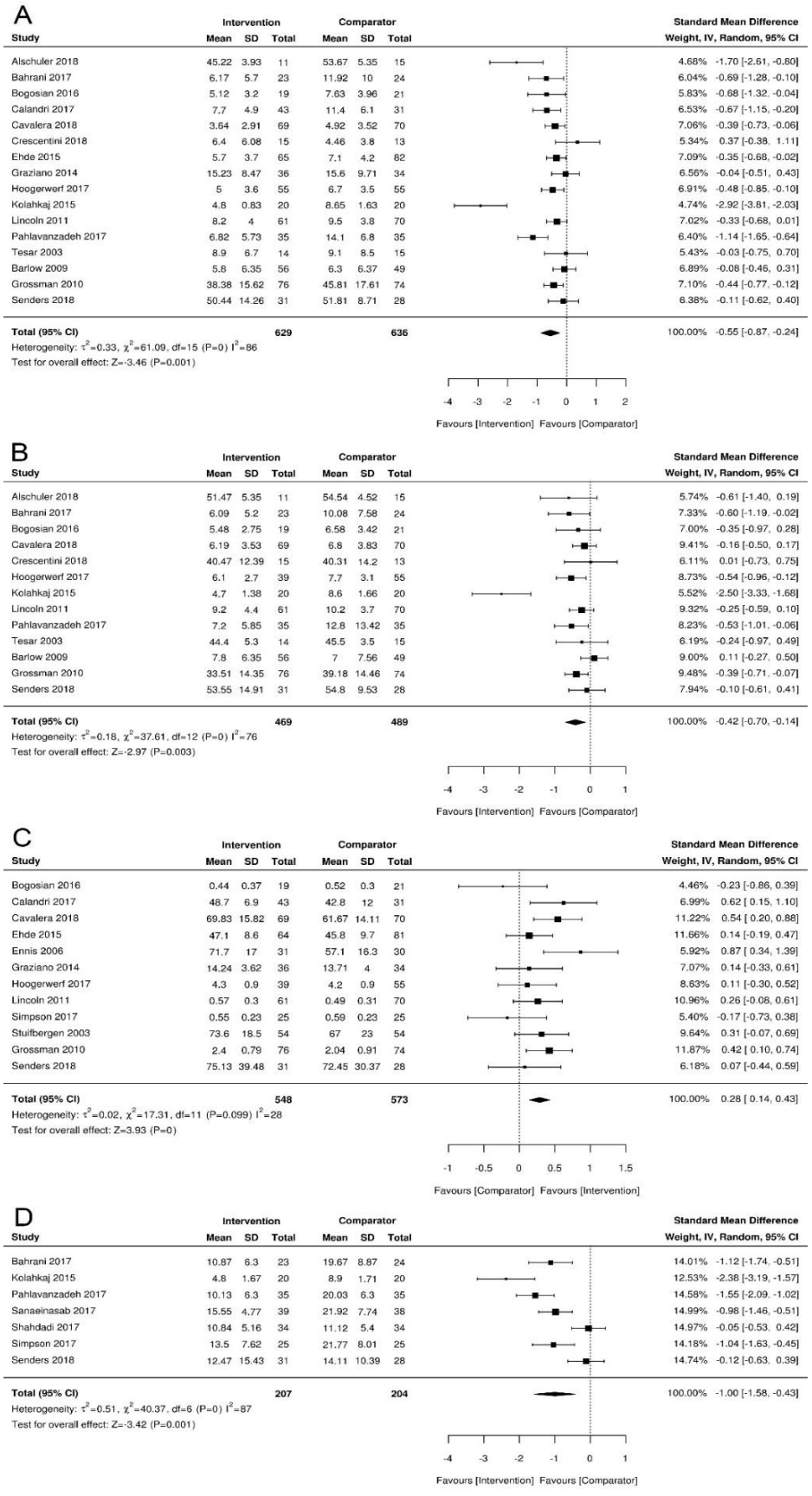

Figure 4 Forest plots for mental health outcomes: depression (A), anxiety (B), quality of life (C), and stress (D)

CI, confidence interval; IV, inverse variance; SD, standard deviation

3.7.3 Depression meta-regression

Studies with missing data on time since diagnosis,[14, 48, 52] mean age,[14, 52] and percentage of females were excluded from meta-regression.[53] Higher percentage of females, minimum eight week intervention (*vs*. less than eight weeks), and waitlist comparator (*vs*. active comparator), were statistically significant inverse predictors of depression. In-person interventions and mindfulness interventions were statistically significantly less effective, compared to teleconference/videoconference and non-mindfulness interventions, respectively, at reducing depression (**Table 5**). These five factors accounted for all variability in effect size estimates between studies (residual I^2 = 0%, adjusted R^2 =100%).

Table 5 Multivariable meta-regression showing statistically significant predictors of depression[a]

Predictor	Estimate	95% CI	*P* value
Percentage of females, per 1%	-0.16	-0.26, -0.01	0.002
In-person (*vs.* teleconference/videoconference)	0.73	0.30, 1.17	0.001
Minimum 8 weeks (*vs.* less than eight weeks)	-0.95	-1.67, -0.24	0.009
Waitlist comparator (*vs.* active comparator)	-0.62	-1.14, -0.10	0.019
Mindfulness intervention (*vs.* other)	0.69	0.08, 1.31	0.026

[a]Depression was measured using the following tools: the Beck Depression Inventory; the Center for Epidemiologic Studies Depression Scale; the Depression, Anxiety and Stress Scales; the Hospital Anxiety and Depression Scale; and the Patient-Reported Outcomes Measurement Information System. Higher scores indicate greater severity.

3.7.4 Anxiety

Twenty one studies measured anxiety.[14-16, 31-35, 37, 38, 41-47, 50, 52, 53, 55] Relative to comparators, ten studies reported statistically significant improvements in anxiety scores[14, 16, 35, 38, 41, 42, 45, 50, 52, 53] (including one study that reported a beneficial effect in trait anxiety but not state anxiety,[41] and another that reported improved anxiety compared to only one of two comparator groups – the 'information booklet only' group, but not the 'social discussion plus booklet' group[35]). The most frequently used tool to measure anxiety was the HADS (used in nine studies[16, 31-33, 35, 42, 46, 47, 53]), followed by the State-Trait Anxiety Inventory (STAI; used in four studies[38, 41, 43, 50]) The most frequently used BCTs were: behavioural practice/rehearsal (18 studies[14-16, 31-34, 37, 38, 41-43, 45-47, 50, 53, 55, 59]); social support (unspecified) (12 studies[16, 31, 32, 34, 35, 37, 38, 44-46, 53, 55]); and social comparison (11 studies[14-16, 32, 35, 37, 43-45, 53, 55]). Of the ten effective interventions, eight used behaviour practice/rehearsal as a BCT.[14, 16, 38, 42, 45, 50, 52, 53] Five studies used social support (unspecified),[16, 35, 38, 45, 53] five used social comparison,[14, 16, 35, 45, 53] and five used framing/reframing.[14, 16, 38, 45, 53]

3.7.5 Anxiety meta-analysis

Thirteen studies were included in statistical meta-analysis.[14, 16, 41-47, 50, 52, 53, 55] One study[41] reported trait and state anxiety subscales; the state score was used in meta-analysis as it measures current anxiety levels. Meta-analysis included 958 participants (469 received the intervention), and resulted in a statistically significant medium effect, favouring the intervention (SMD -0.42; 95% CI: -0.70, -0.14; *P*=0.003). Heterogeneity was high (I^2=76%; chi-squared *P*<0.001) (**Figure 4**). Subgroup analysis was performed to examine the robustness of the findings. Overall, there was minimal change in SMD and heterogeneity when grouped by: minimum eight-week interventions (SMD -0.44; 95% CI -0.82, -0.07; *P*=0.02; I^2=86%; chi-squared *P*<0.001); in-person interventions (SMD -0.46; 95% CI -0.84, -0.07; *P*=0.02; I^2=84%; chi-squared *P*<0.001); waitlist control/usual care comparators (SMD -0.49; 95% CI -0.83, -0.15; *P*=0.005; I^2=80%; chi-squared *P*<0.001); and mindfulness interventions (SMD -0.54; 95% CI -1.02, -0.06; *P*=0.028; I^2=87%; chi-squared *P*<0.001).

3.7.6 Anxiety meta-regression

Studies with missing data on time since diagnosis,[14, 52] mean age,[14, 52] and percentage of females were excluded from meta-regression.[53] Minimum eight week intervention duration (*vs*. less than eight weeks) was the only statistically significant predictor of anxiety, with an inverse association (estimate -0.39, 95% CI -0.77, -0.01, *P*=0.048). This factor accounted for all variability in effect size estimates between studies (residual I^2 = 0%, adjusted R^2 =100%).

3.7.8 Quality of Life

Thirteen studies measured quality of life.[15, 16, 32, 40, 42, 47-51, 53, 55, 57] Relative to comparators, six studies reported significant improvements in quality of life scores.[16, 40, 49, 50, 53, 57] The most frequently used tool to measure quality of life was the Medical Outcomes Study 36-Item Short-Form Health Survey (SF-36; used in four studies[32, 49, 55, 57]). The most frequently used BCTs were: behavioural practice/rehearsal (12 studies[15, 16, 32, 40, 42, 47-51, 53, 55]); social comparison (nine studies[15, 16, 32, 40, 49, 51, 53, 55, 57]); credible source (eight studies[15, 16, 40, 42, 47, 49, 51, 57]); and goal setting (outcome) (eight studies[32, 40, 48-51, 53, 57]). Of the six effective interventions, five used behaviour practice/rehearsal[16, 40, 49, 50, 53] and five used goal setting (outcome)[40, 49, 50, 53, 57].

3.7.9 Quality of life meta-analysis

Twelve studies were included in statistical meta-analysis.[15, 16, 40, 42, 47-51, 53, 55, 57] One study[50] reported multiple measures of quality of life (the Hamburg Quality of Life Questionnaire in Multiple Sclerosis (HAQUAMS) and the Profile of Health-Related Quality of Life in Chronic Disorders): the HAQUAMS score was included in meta-analysis as this tool is specific to an MS population. Meta-analysis included 1121 participants (548 received the intervention), and resulted in a statistically significant small effect, favouring the intervention (SMD 0.28; 95% CI: 0.14-0.43; *P*<0.001). Heterogeneity was low-to-moderate (I^2=28%; chi-squared *P*=0.099) (**Figure 4**). Subgroup analysis was performed to examine the robustness of the findings. Heterogeneity was not statistically significant when grouped by minimum eight-week interventions (SMD 0.27; 95% CI 0.10, 0.43; *P*=0.001; I^2=28%; chi-squared *P*=0.11) and in-person interventions (SMD 0.30; 95% CI 0.15, 0.46; *P*<0.001; I^2=15%; chi-squared *P*=0.168). Heterogeneity increased to 'moderate' when studies were grouped by waitlist control/usual care comparators (SMD 0.30; 95%CI 0.09, 0.50; *P*=0.004; I^2=43%; chi-squared *P*=0.066) and mindfulness only (SMD 0.19; 95% CI -0.05, 0.44; *P*=0.125; I^2=48%; chi-squared *P*=0.096). Meta-regression analysis was not undertaken because heterogeneity was low-to-moderate.

3.7.10 Stress

Eight studies measured stress.[14, 15, 34, 45, 52, 54-56] Relative to comparators, six studies reported significant improvements in stress scores.[14, 15, 45, 52, 54, 56] The tools used to measure stress were the Depression, Anxiety and Stress Scales (five studies[14, 34, 45, 52, 56]) and the Perceived Stress Scale (three studies[15, 54, 55]). The most frequently used BCT was behavioural practice/rehearsal (seven studies[14, 15, 34, 45, 52, 54, 55]). Five studies used social comparison,[14, 15, 45, 54, 55] prompts/cues,[14, 15, 34, 54, 55] and reduce negative emotions.[14, 15, 34, 45, 55] Of the six effective interventions, behaviour practice/rehearsal was used in five.[14, 15, 45, 52, 54]

3.7.11 Stress meta-analysis

Seven studies were included in statistical meta-analysis.[14, 15, 45, 52, 54-56] Meta-analysis included 411 participants (207 received the interventions), and resulted in a statistically significant large effect, favouring the intervention (SMD -1.00; 95% CI -1.58, -0.43; *P*=0.001). Heterogeneity was high (I^2=87%; chi-squared *P*<0.001) (**Figure 4**). Due to the small number of studies, subgroup analysis and meta-regression were unable to be performed.

4. Discussion

4.1 Summary of findings

This systematic review and meta-analysis included 29 studies with 2323 participants, and investigated the effectiveness of emotional wellness programs on depression, anxiety, quality of life, and stress in adults with MS. Three-quarters of participants were female; consistent with the sex-distribution of the disease.[60] The mean age was 44 years, and participants had been diagnosed with MS for an average of nine years. The emotional wellness programs were based on several approaches, including mindfulness, self-management interventions, cognitive behavioural principles or other psychological therapies, adjustment to MS, health promotion/wellness, coping skills, and self-care instruction. The most common number of sessions was eight (conducted once a week or once a fortnight). The majority of studies compared the intervention group to a waitlist control group or a treatment as usual group. Sample sizes were generally small (intervention median=35; comparator median=31); the smallest study had 11 participants in the intervention group. At post-intervention, less than half of the studies were adequately powered to detect statistically significant effects.

Results from meta-analyses showed favourable effects of the interventions: decreasing stress (large effect); reducing depression and anxiety (medium effect); and improving quality of life (small effect). Many interventions lasted for eight weeks and were implemented in-person; however, subgroup analyses did not produce noteworthy changes in effect estimates compared with the main models. As such, there is insufficient evidence to make recommendations on optimal program duration or format. However, we acknowledge that the analyses may have been underpowered to detect significant changes given the small number of studies that were fewer than eight weeks in duration, and that were not conducted in-person.

The mean number of BCTs used across all interventions was eight. Behaviour practice/rehearsal was used in nearly all of the studies; social comparison and social support were both frequently used. Of the efficacious studies, behaviour practice/rehearsal was the most commonly used BCT. A large number of studies did not report an underlying behaviour change theory.

4.2 Comparison with existing literature

We found emotional wellness programs effective at improving depression, anxiety, quality of life, and stress in adults with MS. Consistent with our findings, a recent meta-analysis on psychosocial interventions for pwMS (minimum *n* intervention participants=20) reported statistically significant small effect sizes on depression and anxiety, and a greater effect size for health-related quality of life.[61] Likewise, Simpson and colleagues recently published a meta-analysis on mindfulness interventions for pwMS, reporting that mindfulness was moderately effective at treating depression (SMD 0.35; 95% CI 0.17-0.53), anxiety (SMD 0.35; 95% CI 0.15-0.55), and stress (SMD 0.55; 0.25-0.85).[62] Venasse and colleagues drew the same conclusion when examining mindfulness interventions for people with progressive MS (level B evidence; probably effective), but only three studies were included in their review.[20] Similarly, systematic reviews on self-management interventions (2017)[17] and stress-management interventions (2014)[23] both reported beneficial effects on mental health and quality of life outcomes for pwMS. However, both reviews included a small number of studies (10[17] and eight[23]), which varied considerably in quality.

The most commonly used BCTs in interventions that improved mental health outcomes were behaviour practice/rehearsal (participants were encouraged to practice the skills) and social comparison (participants were given the opportunity to discuss topics with peers). These findings provide some guidance for the design of future emotional wellness programs for pwMS. In previous reviews of self-management interventions for pwMS, goal setting was associated with improvements in depression and anxiety,[17] and general instruction, barrier identification practice, and social support were commonly used BCTs.[18] Differences in the commonly used BCTs in our findings and in the aforementioned reviews may be attributed to their specific focus on self-management interventions (empowering individuals to manage their symptoms, treatment, psychosocial, and lifestyle aspects of the disease), whereas the interventions in our review were broader in scope. Two reviews on physical activity behaviour in pwMS reported different BCTs compared with our study: goal setting was the most common in one study,[63] while social support was the most common in the other.[64] This highlights the variability in effective BCTs used in interventions for pwMS. Similar to our findings, a recent review on lifestyle behaviour change for preventing the progression of kidney disease found that social support and behaviour practice/rehearsal were frequently used in effective interventions.[65]

Few studies included in our review reported the use of specific behaviour change theories, despite describing behaviour change techniques. These results are consistent with two reviews (one on self-management interventions for pwMS[18] and the other on wellness interventions for pwMS[21]), which reported that studies were rarely based on behaviour change theories. The social cognitive theory and the transtheoretical model of change are two theories commonly used in the MS literature for wellness[21] and physical activity behaviour change.[63, 66] Given the complexities surrounding behaviour change, the use of appropriate theory-based interventions would strengthen research in this area.

4.3 Strengths and limitations of this review

This review was undertaken using a thorough search strategy that was developed in consultation with a Health Sciences librarian. The methods were guided by the JBI guidelines for systematic reviews of effectiveness[24] and the PRISMA checklist of items for reporting systematic reviews.[39] Studies included were RCTs and quasi-experimental trials with valid comparator groups, of which only three were excluded for poor methodological quality. The main limitations of this review pertain to the relatively small sample sizes of the included studies, the heterogeneous nature of the interventions, and potential publication bias. The number of studies in meta-analyses was less than 20, and the mean sample size was less than 80. As such, the I^2 index and the chi-squared *P* values should be interpreted with caution.[67] Furthermore, less than half of the studies were adequately powered to detect statistically significant changes in mental health outcomes post-intervention. Due to incomplete reporting, the effect of baseline mental health and disability status could not be investigated as potential covariates.

5. Conclusions

Despite the limitations pertaining to heterogeneity and sample size, there is evidence to support the effectiveness of emotional wellness programs for improving mental health outcomes in pwMS. While we cannot draw firm conclusions regarding optimal program characteristics, the majority of the included studies were conducted in group settings, in-person, and were run once a week or once a fortnight for eight sessions. Future studies would benefit from exploring adherence rates and follow-up data in order to assess the feasibility and long-term effectiveness of emotional wellness programs. Improved reporting

of BCTs in future studies would enable researchers to identify those that are most effective for pwMS.

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Appendices

Appendix A Search strategy for MEDLINE (Ovid) and CINAHL

MEDLINE (Ovid)	
Search number	Search terms
1	Exp Multiple Sclerosis or multiple sclerosis.mp.
2	deymyelinating disease.mp.
3	optic neuritis.mp.
4	demyelinating disorder.mp.
5	1 OR 2 OR 3 OR 4
6	exp Health Education/ or health education.mp.
7	exp Patient Participation or patient participation.mp.
8	education*.mp.
9	exp Health Promotion/ or health promotion.mp.
10	patient information.mp.
11	client information.mp.
12	Intervention.ab,ti.
13	Program*.ab,ti.
14	6 OR 7 OR 8 OR 9 OR 10 OR 11 OR 12 OR 13
15	exp Health Status/ or health status.mp.
16	well-being.mp. or wellbeing.mp.
17	exp "Quality of Life"/
18	exp Mindfulness/ or mindfulness.mp.
19	Mindfulness-based.mp.
20	exp Stress, Psychological/ or stress.mp.
21	exp Self Care/
22	(self care or self-care).mp.
23	cognitive health.mp.
24	wellness.mp.
25	exp Depression/ or depression.mp.
26	exp Anxiety/ or exp Anxiety Disorders/ or anxiety.mp.
27	coping.mp.
28	Resilienc*.mp/ or exp Resilience, Psychological/
29	Meditat*.mp. Or exp Meditation/
30	Cognitive training.mp.
31	Self-efficacy.mp. Or exp Self Efficacy/
32	15 OR 16 OR 17 OR 18 OR 19 OR 20 OR 21 OR 22 OR 23 OR 24 OR 25 OR 26 OR 27 OR 28 OR 29 OR 30 OR 31
33	5 AND 14 AND 32
34	limit 33 (English language and humans)

CINAHL	
Search number	Search terms
S1	(MH "Multiple Sclerosis+") OR "multiple sclerosis"
S2	"deymyelinating disease"
S3	"optic neuritis"
S4	"demyelinating disorder"
S5	S1 or S2 or S3 or S4
S6	(MH "Health Education+") OR "health education"
S7	(MH "Consumer Participation") OR "patient participation"
S8	(MH "Health Promotion+") OR "health promotion"
S9	"patient information"
S10	"client information"
S11	TI intervention* OR AB intervention*
S12	AB program* OR TI program*
S13	TI education OR AB education
S14	S6 or S7 or S8 or S9 or S10 or S11 or S12 or S13
S15	(MH "Health Status+") OR "health status"
S16	(MH "Psychological Well-Being") OR "well-being"
S17	"wellbeing"
S18	(MH "Quality of Life+")
S19	"mindfulness-based"
S20	(MH "Mindfulness") OR "mindfulness"
S21	(MH "Stress+") OR "stress" OR (MH "Stress, Psychological+")
S22	(MH "Self Care+") OR "self care" OR "self-care"
S23	"cognitive health"
S24	(MH "Wellness") OR "wellness"
S25	(MH "Depression+") OR "depression"
S26	(MH "Anxiety") OR "anxiety"
S27	(MH "Coping+") OR "coping"
S28	(MH "Hardiness:)
S29	"resilienc*"
S30	"meditat*" OR (MH "Meditation")
S31	"cognitive training"
S32	(MH "Self-Efficacy") OR "self-efficacy" OR "self efficacy"
S33	S15 or S16 or S17 or S18 or S19 or S20 or S21 or S22 or S23 or S24 or S25 or S26 or S27 or S28 or S29 or S30 or S31 or S32
S34	S5 and S12 and S31 (limiters - English Language)

Appendix B Tools used to measure depression, anxiety, quality of life, and stress

Outcome	Tool	Score range
Depression	Arthritis Impact Measurement Scales (AIMS)[1]	0 - 10
	Beck Depression Inventory (BDI)[1]	0 - 63
	BDI-II[1]	0 - 63
	Center for Epidemiologic Studies Depression Scale (CES-D)[1]	0 - 60
	Depression, Anxiety and Stress Scales (DASS-21)[1]	0 - 21
	Hospital Anxiety and Depression Scale (HADS)[1]	0 - 21
	Mental Health Inventory (MHI-18)[2]	0 - 100
	Patient Health Questionnaire (PHQ-9)[1]	0 - 27
	Patient-Reported Outcomes Measurement Information System (PROMIS)[1]	35.2 - 82.4
Anxiety	AIMS[1]	0 - 10
	DASS-2[1]	0 - 21
	HADS[1]	0 - 21
	MHI-18[2]	0 - 100
	PROMIS[1]	35.2 - 82.4
	State-Trait Anxiety Inventory (STAI)[1]	20 - 80
Quality of life	EuroQol (EQ-5D)[1]	0 - 1
	Hamburg Quality of Life Questionnaire in Multiple Sclerosis (HAQUAMS)[1]	1 - 5
	Life Satisfaction Questionnaire (LiSat-9)[1]	9 - 54
	Multiple Sclerosis Quality of Life-54 (MSQOL-54)[1]	0 - 24
	Profile of Health-Related Quality of Life in Chronic Disorders (PQOLC)[1]	0 - 24
	Medical Outcomes Study 8-Item Short-Form Health Survey (SF-8)[1]	0 - 100
	Medical Outcomes Study 12-Item Short-Form Health Survey (SF-12)[1]	0 - 100
	Medical Outcomes Study 36-Item Short-Form Health Survey (SF-36)[1]	0 - 100
Stress	DASS-21[1]	0 - 21
	Perceived Stress Scale (PSS)[1]	0 - 40

[1]Higher score indicates greater severity of outcome

[2]Higher score indicates lower severity of outcome

Appendix C Studies ineligible following review of full text (*n*=69)

Agland, S., Shaw, S., Lea, R., Mortimer-Jones, S., & Lechner-Scott, J. (2017). Does mindfulness, meditation and progressive muscle relaxation reduce stress in people with multiple sclerosis? Multiple Sclerosis Journal, 23(3), 963-964. https://doi.org/10.1177/1352458517731285
Reason for exclusion: Abstract or poster

Bermudez, M., Olivares, T., Moises, B., Hernandez, M. A., & Villar Van Weigaert, C. (2015). Cognitive behavioural therapy in multiple sclerosis: effectiveness in reducing depressive symptoms and cognitive impairments. Multiple Sclerosis, 21(11 SUPPL. 1), 230. https://doi.org/10.1177/1352458515602642
Reason for exclusion: Abstract or poster

Fischer, A., Schroder, J., Pottgen, J., Lau, S., Heesen, C., Moritz, S., & Gold, S. M. (2013). Effectiveness of an internet-based treatment programme for depression in multiple sclerosis: a randomized controlled trial. Multiple Sclerosis Journal, 19(11), 350-351.
Reason for exclusion: Abstract or poster

Franco, M., Barone, D., Barone, K., Foley, F., Pfohl, D. C., Rosenberg, J., . . . Treadaway, K. (2008). Patient education: using relaxation and guided imagery to lower anxiety associated with multiple sclerosis and injections. International Journal of MS Care, 10, 44-45.
Reason for exclusion: Abstract or poster

Goldoust, F., Ebadifard Azar, F., Solhi, M., & Ghorchiany, F. (2012). Planning and Evaluation of Stress Management Educational Program to Improve Behavior in Multiple Sclerosis Patients Based on Basnef Model. Journal of Urmia Nursing & Midwifery Faculty, 10(3), 1-9.
Reason for exclusion: Abstract or poster

Gonzalez-Suarez, I., Munoz-San Jose, A., Cebolla Lorenzo, S., Carrillo Notario, L., Lopez De Velasco, V., Orviz Garcia, A., . . . Oreja-Guevara, C. (2016). Benefits of a mindfulness-based intervention compared to psychoeducation among multiple sclerosis patients. Multiple Sclerosis, 22, 694-. https://doi.org/10.1177/1352458516663086
Reason for exclusion: Abstract or poster

Granmayeh, S. H., Besharat, M., Nabavi, S. M., Sadeghi, S., & Imani, A. (2012). The effects of Mindfulness-based Stress Reduction programme on physical symptoms, quality of life, and mental health in patients with multiple sclerosis. Journal of Neurology, 259, S154-S154.
Reason for exclusion: Abstract or poster

Kalina, J. (2016). Effects of a Program Designed to Improve Self-Efficacy and Subsequent Effects on Decreasing Loneliness and Depression Among People with Multiple Sclerosis. Neurology, 86.
Reason for exclusion: Abstract or poster

Landtblom, A. M., Guala, D., Hau, S., Jansson, L., Martin, C., & Fredrikson, S. (2017). RebiQoL: a telemedicine patient support program on health related quality of life and adherence in MS patients treated with Rebif. Multiple Sclerosis Journal, 23(3), 425-. https://doi.org/10.1177/1352458517731404
Reason for exclusion: Abstract or poster

Munoz San Jose, A., Cebolla Lorenzo, S., Carrillo, L., Gonzalez-Suarez, I., Sanz Velasco, N., Soto Lopez, T., . . . Oreja-Guevara, C. (2015). Mindfulness in multiple sclerosis patients. European Journal of Neurology, 22, 826. https://doi.org/10.1111/ene.12808
Reason for exclusion: Abstract or poster

Saeed, R., Evangelou, N., & Turner, A. (2014). A service evaluation of the Multiple Sclerosis Mindfulness Programme. Multiple Sclerosis Journal, 20(7), 991-991.
Reason for exclusion: Abstract or poster

Bombardier, C. H., Cunniffe, M., Wadhwani, R., Gibbons, L. E., Blake, K. D., & Kraft, G. H. (2008). The efficacy of telephone counseling for health promotion in people with multiple sclerosis: a randomized controlled trial. Archives of Physical Medicine & Rehabilitation, 89(10), 1849-1856
Reason for exclusion: Cannot extract emotional wellness program component

Burschka, J. M., Keune, P. M., van Oy, U. H., Oschmann, P., & Kuhn, P. (2014). Mindfulness-based interventions in multiple sclerosis: Beneficial effects of Tai Chi on balance, coordination, fatigue and depression. BMC Neurology, 14
Reason for exclusion: Cannot extract emotional wellness program component

Gilbertson, R. M., & Klatt, M. D. (2017). Mindfulness in Motion for People with Multiple Sclerosis: A Feasibility Study. International Journal of MS Care, 19(5), 225-231. https://doi.org/10.7224/1537-2073.2015-095
Reason for exclusion: Cannot extract emotional wellness program component

Hadgkiss, E. J., Jelinek, G. A., Taylor, K. L., Marck, C. H., van der Meer, D. M., Pereira, N. G., & Weiland, T. J. (2015). Engagement in a program promoting lifestyle modification is associated with better patient-reported outcomes for people with MS. Neurological Sciences, 36(6), 845-852.
Reason for exclusion: Cannot extract emotional wellness program component

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Reason for exclusion: Cannot extract emotional wellness program component

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Reason for exclusion: Cannot extract emotional wellness program component

Malec, C. A. (2002). The effect of a healthy lifestyle intervention on quality of life in the chronically ill: A Randomized Control Trial Ph.D. University of Calgary (Canada).
Reason for exclusion: Cannot extract emotional wellness program component

Marck, C. H., De Livera, A. M., Brown, C. R., Neate, S. L., Taylor, K. L., Weiland, T. J., . . . Jelinek, G. A. (2018). Health outcomes and adherence to a healthy lifestyle after a multimodal intervention in people with multiple sclerosis: Three year follow-up. PLoS ONE, 13(5), e0197759.
Reason for exclusion: Cannot extract emotional wellness program component

Ng, A., Kennedy, P., Hutchinson, B., Ingram, A., Vondrell, S., Goodman, T., & Miller, D. (2013). Self-efficacy and health status improve after a wellness program in persons with multiple sclerosis. Disability & Rehabilitation, 35(12), 1039-1044.
Reason for exclusion: Cannot extract emotional wellness program component

Plow, M. A. H. (2006). Comparing the effectiveness of a wellness intervention to prehabilitation in individuals with multiple sclerosis Ph.D. University of Minnesota.
Reason for exclusion: Cannot extract emotional wellness program component

Seifi, K., & Moghaddam, H. E. (2018). The Effectiveness of Self-care Program on the Life Quality of Patients with Multiple Sclerosis in 2015. Journal of the National Medical Association, 110(1), 65-72. https://doi.org/10.1016/j.jnma.2017.01.010
Reason for exclusion: Cannot extract emotional wellness program component

Tietjen, K. M., & Breitenstein, S. (2017). A Nurse-Led Telehealth Program to Improve Emotional Health in Individuals With Multiple Sclerosis. Journal of Psychosocial Nursing and Mental Health Services, 55(3), 31-37. https://doi.org/10.3928/02793695-20170301-04
Reason for exclusion: Cannot extract emotional wellness program component

Burleson Sullivan, A., Scheman, J., LoPresti, A., & Prayor-Patterson, H. (2012). Interdisciplinary Treatment of Patients with Multiple Sclerosis and Chronic Pain. International Journal of MS Care, 14(4), 216-220. https://doi.org/10.7224/1537-2073-14.4.216
Reason for exclusion: Disease or symptom focus

Feicke, J., Spörhase, U., Köhler, J., Busch, C., & Wirtz, M. (2014). A multicenter, prospective, quasi-experimental evaluation study of a patient education program to foster multiple sclerosis self-management competencies. Patient Education and Counseling, 97(3), 361-369. https://doi.org/10.1016/j.pec.2014.09.005
Reason for exclusion: Disease or symptom focus

Köpke, S., Kern, S., Ziemssen, T., Berghoff, M., Kleiter, I., Marziniak, M., . . . Heesen, C. (2014). Evidence-based patient information programme in early multiple sclerosis: a randomised controlled trial. Journal of Neurology, Neurosurgery & Psychiatry, 85(4), 411-418. https://doi.org/10.1136/jnnp-2013-306441
Reason for exclusion: Disease or symptom focus

Kos, D., Duportail, M., Meirte, J., Meeus, M., D'Hooghe, M. B., Nagels, G., . . . Nijs, J. (2016). The effectiveness of a self-management occupational therapy intervention on activity performance in individuals with multiple sclerosis-related fatigue: a randomized-controlled trial. International Journal of Rehabilitation Research, 39(3), 255-262. https://doi.org/10.1097/MRR.0000000000000178
Reason for exclusion: Disease or symptom focus

Thomas, S., Thomas, P. W., Kersten, P., Jones, R., Green, C., Nock, A., . . . et al. (2013). A pragmatic parallel arm multi-centre randomised controlled trial to assess the effectiveness and cost-effectiveness of a group-based fatigue management programme (FACETS) for people with multiple sclerosis. Journal of Neurology, Neurosurgery, and Psychiatry, 84(10), 1092-1099. https://doi.org/10.1136/jnnp-2012-303816
Reason for exclusion: Disease or symptom focus

Bogosian, A., Hughes, A., Norton, S., Silber, E., & Moss-Morris, R. (2016). Potential treatment mechanisms in a mindfulness-based intervention for people with progressive

multiple sclerosis. British Journal of Health Psychology, 21(4), 859-880. https://doi.org/10.1111/bjhp.12201
Reason for exclusion: Duplicate studies

Kalina, J. (2016). Effects of an educational socialization program designed to improve self-efficacy and subsequent effects on decreasing loneliness and depression among people with multiple sclerosis. Dissertation Abstracts International: Section B: The Sciences and Engineering, 77(3-B(E)).
Reason for exclusion: Duplicate studies

Cosio, D., Jin, L., Siddique, J., Mohr, D. C., Cosio, D., Jin, L., . . . Mohr, D. C. (2011). The effect of telephone-administered cognitive-behavioral therapy on quality of life among patients with multiple sclerosis. Annals of Behavioral Medicine, 41(2), 227-234. https://doi.org/10.1007/s12160-010-9236-y
Reason for exclusion: Individualised cognitive therapy

Fischer, A., Schroder, J., Vettorazzi, E., Wolf, O. T., Pottgen, J., Lau, S., . . . Gold, S. M. (2015). An online programme to reduce depression in patients with multiple sclerosis: a randomised controlled trial. Lancet Psychiatry, 2(3), 217-223. https://doi.org/10.1016/s2215-0366(14)00049-2
Reason for exclusion: Individualised cognitive therapy

Kiropoulos, L. A., Kilpatrick, T., Holmes, A., & Threader, J. (2016). A pilot randomized controlled trial of a tailored cognitive behavioural therapy based intervention for depressive symptoms in those newly diagnosed with multiple sclerosis. BMC Psychiatry, 16(1), 435.
Reason for exclusion: Individualised cognitive therapy

Mohr, D. C., Hart, S., & Vella, L. (2007). Reduction in disability in a randomized controlled trial of telephone-administered cognitive-behavioral therapy. Health Psychology, 26(5), 554-563.
Reason for exclusion: Individualised cognitive therapy

Anderson, J. K., Turner, A., & Clyne, W. (2017). Development and feasibility of the Help to Overcome Problems Effectively (HOPE) self-management intervention for people living with multiple sclerosis. Disability & Rehabilitation, 39(11), 1114-1121
Reason for exclusion: No comparator group

Becker, H., Stuifbergen, A. K., Schnyer, R. N., Morrison, J. D., & Henneghan, A. (2017). Integrating Acupuncture Within a Wellness Intervention for Women With Multiple Sclerosis. Journal of Holistic Nursing, 35(1), 86-96.
Reason for exclusion: No comparator group

Blankespoor, R. J., Schellekens, M. P., Vos, S. H., Speckens, A. E., & Jong, B. A. (2017). The effectiveness of mindfulness-based stress reduction on psychological distress and cognitive functioning in patients with multiple sclerosis: A pilot study. Mindfulness, 8(5), 1251-1258.
Reason for exclusion: No comparator group

Brittle, N., Brown, M., Mant, J., McManus, R., Riddoch, J., & Sackley, C. (2008). Short-term effects on mobility, activities of daily living and health-related quality of life of a Conductive Education programme for adults with multiple sclerosis. Clinical Rehabilitation, 22(4), 329-337.
Reason for exclusion: No comparator group

Calandri, E., Graziano, F., Borghi, M., & Bonino, S. (2017). Improving the quality of life and psychological well-being of recently diagnosed multiple sclerosis patients: preliminary evaluation of a group-based cognitive behavioral intervention. Disability & Rehabilitation, 39(15), 1474-1481
Reason for exclusion: No comparator group

Chruzander, C., Gottberg, K., Ytterberg, C., Backenroth, G., Fredrikson, S., Widén Holmqvist, L., & Johansson, S. (2016). A single-group pilot feasibility study of cognitive behavioural therapy in people with multiple sclerosis with depressive symptoms. Disability & Rehabilitation, 38(24), 2383-2391.
Reason for exclusion: No comparator group

Crawford, J. D., & McIvor, G. P. (1987). Stress management for multiple sclerosis patients. Psychological Reports, 61(2), 423-429.
Reason for exclusion: No comparator group

Hankin, V. M. (2010). Mindfulness based stress reduction in couples facing multiple sclerosis: Impact on self reported anxiety and uncertainty. Dissertation Abstracts International: Section B: The Sciences and Engineering, 70(10-B), 6551.
Reason for exclusion: No comparator group

Pakenham, K. I., Mawdsley, M., Brown, F. L., & Burton, N. W. (2018). Pilot evaluation of a resilience training program for people with multiple sclerosis. Rehabilitation Psychology, 63(1), 29-42.
Reason for exclusion: No comparator group

Pritchard, M., Elison-Bowers, P., & Birdsall, B. (2010). Impact of integrative restoration (iRest) meditation on perceived stress levels in multiple sclerosis and cancer outpatients. Journal of the International Society for the Investigation of Stress, 26(3), 233-237. https://doi.org/10.1002/smi.1290
Reason for exclusion: No comparator group

Sheppard, S. C., Forsyth, J. P., Hickling, E. J., & Bianchi, J. (2010). A novel application of acceptance and commitment therapy for psychosocial problems associated with multiple sclerosis: results from a half-day workshop intervention. International Journal of MS Care, 12(4), 200-206.
Reason for exclusion: No comparator group

Sinclair, V. G., & Scroggie, J. (2005). Effects of a cognitive-behavioral program for women with multiple sclerosis. Journal of Neuroscience Nursing, 37(5), 249-257, 276.
Reason for exclusion: No comparator group

Spitzer, E., & Pakenham, K. I. (2018). Evaluation of a brief community-based mindfulness intervention for people with multiple sclerosis: A pilot study. Clinical Psychologist, 22(2), 182-191. https://doi.org/10.1111/cp.12108
Reason for exclusion: No comparator group

Visschedijk, M. A., Collette, E. H., Pfennings, L. E., Polman, C. H., & Van Der Ploeg, H. M. (2004). Development of a Cognitive Behavioral Group Intervention Programme For Patients with Multiple Sclerosis: An Exploratory Study. Psychological Reports, 95(3,Part1), 735-746.
Reason for exclusion: No comparator group

Wingerson, N. W., & Wineman, N. (2000). The mental health, self-efficacy, and satisfaction outcomes of a community counseling demonstration project for multiple sclerosis patients. Journal of Applied Rehabilitation Counseling, 31(2), 11-17.
Reason for exclusion: No comparator group

Artemiadis, A. K., Vervainioti, A. A., Alexopoulos, E. C., Rombos, A., Anagnostouli, M. C., & Darviri, C. (2012). Stress management and multiple sclerosis: a randomized controlled trial. Archives of Clinical Neuropsychology, 27(4), 406-416.
Reason for exclusion: No education component

Beatus, J., O'Neill, J. K., Townsend, T., & Robrecht, K. (2002). The effect of a one-week retreat on self-esteem, quality of life, and functional ability for persons with multiple sclerosis. Neurology Report, 26(3), 154-159.
Reason for exclusion: No education component

Khan, F., Amatya, B., Elmalik, A., Lowe, M., Ng, L., Reid, I., & Galea, M. P. (2016). An Enriched Environmental Programme During Inpatient Neuro-Rehabilitation: A Randomized Controlled Trial. Journal of Rehabilitation Medicine, 48(5), 417-425. https://doi.org/10.2340/16501977-2081
Reason for exclusion: No education component

Lincoln, N., Dent, A., Harding, J., Weyman, N., Nicholl, C., Blumhardt, L., & Playford, E. (2002). Evaluation of cognitive assessment and cognitive intervention for people with multiple sclerosis. Journal of Neurology, Neurosurgery & Psychiatry, 72(1), 93-98
Reason for exclusion: No education component

Block, P., Vanner, E. A., Keys, C. B., Rimmer, J. H., & Skeels, S. E. (2010). Project Shake-It-Up: using health promotion, capacity building and a disability studies framework to increase self efficacy. Disability & Rehabilitation, 32(9), 741-754.
Reason for exclusion: No outcomes of interest

Kalina, J., Hinojosa, J., Strober, L., Bacon, J., Donnelly, S., & Goverover, Y. (2018). Randomized controlled trial to improve self-efficacy in people with multiple sclerosis: The Community Reintegration for Socially Isolated Patients (CRISP) program. American Journal of Occupational Therapy, 72(5), 1-8.
Reason for exclusion: No outcomes of interest

Liu, Y. J. (2017). A Hope-Based Group Therapy Program to Women with Multiple Sclerosis: Quality of Life. Neuroquantology, 15(4), 127-132. https://doi.org/10.14704/nq.2017.15.4.1135
Reason for exclusion: No outcomes of interest

Shevil, E. (2008). Developing and pilot testing a cognitive intervention program for persons with multiple sclerosis. Dissertation Abstracts International: Section B: The Sciences and Engineering, 69(5-B), 2954.
Reason for exclusion: No outcomes of interest

Shevil, E., & Finlayson, M. (2010). Pilot study of a cognitive intervention program for persons with multiple sclerosis. Health Education Research, 25(1), 41-53.
Reason for exclusion: No outcomes of interest

Stuifbergen, A., Becker, H., Rogers, S., Timmerman, G., & Kullberg, V. (1999). Promoting wellness for women with multiple sclerosis. Journal of Neuroscience Nursing, 31(2), 73-79.

Reason for exclusion: No outcomes of interest

Stuifbergen, A. K., Becker, H., Timmerman, G. M., & Kullberg, V. (2003). The use of individualized goal setting to facilitate behavior change in women with multiple sclerosis. Journal of Neuroscience Nursing, 35(2), 94-99, 106.
Reason for exclusion: No outcomes of interest

Dehghani, A., Kermanshahi, S., & Memarian, R. (2012). The effect of peer group educational program on multiple sclerosis patients ' level of stress.
Reason for exclusion: Not in English language

Boosman, H., Visser-Meily, J. M., Meijer, J.-W. G., Elsinga, A., & Post, M. W. (2011). Evaluation of change in fatigue, self-efficacy and health-related quality of life, after a group educational intervention programme for persons with neuromuscular diseases or multiple sclerosis: A pilot study. Disability and Rehabilitation: An International, Multidisciplinary Journal, 33(8), 690-696.
Reason for exclusion: Not exclusively MS participants (can't extract MS data)

Canade, R. F. (2014). Be here now: evaluating an adapted mindfulness-based intervention in a mixed population with acquired brain injury (ABI) and neurological conditions Ph.D. University of Hertfordshire (United Kingdom).
Reason for exclusion: Not exclusively MS participants (can't extract MS data)

Hughes, R. B., Robinson-Whelen, S., Taylor, H. B., & Hall, J. W. (2006). Stress self-management: an intervention for women with physical disabilities. Womens Health Issues, 16(6), 389-399.
Reason for exclusion: Not exclusively MS participants (can't extract MS data)

Mandel, A. R., & Keller, S. M. (1986). Stress management in rehabilitation. Archives of Physical Medicine & Rehabilitation, 67(6), 375-379
Reason for exclusion: Not exclusively MS participants (can't extract MS data)

Muller, R., Gertz, K. J., Molton, I. R., Terrill, A. L., Bombardier, C. H., Ehde, D. M., & Jensen, M. P. (2016). Effects of a Tailored Positive Psychology Intervention on Well-Being and Pain in Individuals With Chronic Pain and a Physical Disability: A Feasibility Trial. Clinical Journal of Pain, 32(1), 32-44.
Reason for exclusion: Not exclusively MS participants (can't extract MS data)

Classen, S. (2002). The long-term effectiveness of two occupational therapy interventions on the lives of people with MS: a randomized controlled trial Ph.D. Nova Southeastern University.
Reason for exclusion: Rehabilitation-focus

Egner, A., Phillips, V., Vora, R., & Wiggers, E. (2003). Depression, fatigue, and health-related quality of life among people with advanced multiple sclerosis: Results from an exploratory telerehabilitation study. NeuroRehabilitation, 18(2), 125-133.
Reason for exclusion: Rehabilitation-focus

Hanssen, K., Beiske, A., Landro, N., Hofoss, D., & Hessen, E. (2016). Cognitive rehabilitation in multiple sclerosis: A randomized controlled trial. Acta Neurologica Scandinavica, 133(1), 30-40.
Reason for exclusion: Rehabilitation-focus

Appendix D Studies excluded for scoring less than 50% on assessment of methodological quality

Haji-Adineh S, Farzanfar A, Salehi-Morekani S, et al. (2019). The Effectiveness of Mindfulness-Based Cognitive Therapy on Life Expectancy and Depression in Patients with Multiple Sclerosis. International Journal of Body, Mind, and Culture, 6, 79-89. https://doi.org/10.22122/ijbmc.v6i2.160.

Khayeri F, Rabiei L, Shamsalinia A, et al. (2016). Effect of Fordyce Happiness Model on depression, stress, anxiety, and fatigue in patients with multiple sclerosis. Complementary Therapies in Clinical Practice, 25, 130-135.

Rigby S, Thornton E and Young C. (2008). A randomized group intervention trial to enhance mood and self-efficacy in people with multiple sclerosis. British Journal of Health Psychology, 13, 619-631.

Appendix E Table of characteristics of included studies

Author, country, study design	MS type	Sample size (n)	Age y; mean (SD), Female (%)	Disease duration y; mean (SD)	Intervention description; delivery method	Intervention duration; frequency	Comparator	Primary outcomes of the study	Emotional wellness outcome (tool): main findings between IG and CG	Behaviour change theory used
Alschuler *et al.,* 2018. USA, RCT	All	IG: 12 CG: 16	59.8 (7.7), 83% 59.8 (6.5), 100%	18.6 (16.3) 21.0 (12.2)	"Everyday Matters"; aging-focussed resilience; group, tele-conference	90 min; 6 sessions over 6 weeks	Waitlist control	Resilience	Depression (PROMIS): no significant difference (P = 0.09) Anxiety (PROMIS): no significant difference (P > 0.05)	NR
Amiri *et al.,* 2016. Iran, RCT	NR	IG: 20 CG: 20	25.2 (4.5), 48%	NR	Mindfulness; group, in-person	2 hr; 8 sessions over 8 weeks	Usual care	Anxiety Depression Executive Function	Depression (BDI-II): significant improvement in IG (P < 0.01) Anxiety (STAI): significant improvement in IG (P < 0.01)	NR
Bahrani *et al.,* 2017. Iran, RCT	NR	IG: 23 CG: 24	36.8 (6.1), 100% 36.0 (7.1), 100%	7.3 (3.5) 6.7 (3.2)	Mindfulness-integrated cognitive behaviour therapy; group, in-person	2 hr; 8 sessions over 8 weeks	Usual care	Anxiety Depression Stress	Depression (DASS-21): significant improvement in IG (P < 0.001) Anxiety (DASS-21): significant improvement in IG (P < 0.001) Stress (DASS-21): significant improvement in IG (P < 0.001)	NR
Barlow J, *et al.,* 2009. UK, RCT	NR	IG: 78 CG: 64	48.2 (10.1), 73% 50.7 (11.7) 69%	9.6 (8.3) 12.1 (7.4)	Chronic Disease Self-Management Course; group, in-person	2 hr; 6 sessions over 6 weeks	Waitlist control	Depression Self-efficacy	Depression (HADS): IG trend towards improvement (P = 0.051) Anxiety (HADS): no significant difference (P > 0.05)	Self-efficacy theory
Bogosian *et al.,* 2015. UK, RCT	Progressive	IG: 19 CG: 21	53.4 (8.3), 47% 50.9 (9.9), 62%	16.2 (10.1) 12.6 (8.6)	Mindfulness; group, videoconference	1 hr; 8 sessions over 8 weeks	Waitlist control	Distress	Depression (HADS): significant improvement in IG (P = 0.017) Anxiety (HADS): no significant difference at post (P = 0.099) QoL (EQ-5D): no significant difference (P > 0.05)	NR

Calandri *et al.,* 2016. Italy, Quasi-controlled trial	All	IG: 54 CG: 31	38.0 (12.5), 61% 34.8 (11.9), 55%	1.5 (0.7) 1.8 (0.8)	Cognitive behavioural program; group, in-person	2 hr; 5 sessions over 8 weeks, and 1 session at 6 month follow-up	Waitlist control	Depression Optimism Psychological well-being Quality of life	Depression (CES-D): no significant difference (P = 0.258) QoL (SF-12): significant improvement in IG (P = 0.036)	NR
Cavalera *et al.,* 2019. Italy, RCT	RR and SP	IG: 69 CG: 70	42.3 (8.4), 67% 43.2 (9.0), 62%	11.2 (8.0) 12.2 (7.3)	Mindfulness; group, online	2 hr; 8 sessions over 8 weeks	Online psychoeducational group	Quality of life	Depression (HADS): significant improvement in IG (P = 0.020) Anxiety (HADS): significant improvement in IG (P = 0.049) QoL (MSQOL-54): significant improvement in IG (P = 0.033)	NR
Crescentini *et al.,* 2018. Italy, Quasi-controlled trial	All	IG: 15 CG: 13	47.8 (9.3), 80% 49.1 (10.6), 77%	13.1 (10.7) 14.5 (7.7)	Mindfulness-oriented meditation; group, in-person	2 hr; 8 sessions over 8 weeks	Usual care	Temperament and character	Depression (BDI): no significance difference (P < 0.05) Anxiety (STAI-trait): significant improvement in IG (P = 0.04) Anxiety (STAI-state): no significant difference (P > 0.08)	NR
das Nair *et al.,* 2016. UK, RCT	All	IG: 11 CG: 10	48.9 (10.4), 73% 48.0 (11.2), 70%	9.3 (6.8) 8.9 (6.4)	Modified group program for adjustment to MS, based on cognitive and psycho-educational framework; individual, in-person	1 hr; 6 sessions over 12 weeks	Group adjustment program	Feasibility Mood	Depression (BDI-II and HADS): no significant difference (HADS P = 0.13, BDI-II P = 0.57) Anxiety (HADS): no significant difference (P = 0.16)	NR
Ehde *et al.,* 2015. USA, RCT	RR and Progressive	IG: 75 CG: 88	51.0 (10.1), 89.3% 53.2 (10), 85.2%	<5 y 28%; 5-9 y 3%; 10-19 y 39%; 20+ y 11% <5 y 24%; 5-9 y 28%; 10-19 y 30%; 20+ y 18%	Self-management intervention (skill-building) for chronic conditions; individual, telephone-delivered	45-60 min; 6 sessions over 6 weeks	Education program; individual, telephone-delivered	Fatigue impact Pain interference Depression	Depression (PHQ-9): no significant difference (P > 0.05) QoL (SF-8): no significant difference (P > 0.05)	NR

Ennis *et al.,* 2006. UK, RCT	All	IG: 32 CG: 30	45.0 (9), 63% 46.0 (8), 63%	7.0 (5) 8.0 (6)	'OPTIMSE' health promotion education intervention; group, in-person	3 hr; 8 sessions over 8 weeks	Waitlist	Health Promoting Lifestyle Profile	QoL (SF-36, mental health): significant improvement in IG (P < 0.01)	NR
Forman & Lincoln, 2010. UK, RCT	All	IG: 20 CG: 20	47.3 (10.3), 80% 47.7 (9.8), 80%	7.3 (5.4) 12.4 (11.4)	Adjustment to MS program; group, in-person	2 hr; 6 sessions over 12 weeks	Waitlist	Mood	Depression (HADS): significant improvement in IG (area under curve P = 0.02; includes 6 month follow-up) Anxiety (HADS): no significant difference (area under curve P = 0.89; includes 6 month follow-up) QoL (SF-36, psychological): no significant difference (area under curve P = 0.90, includes 6 month follow-up)	Cognitive behavioral therapy principles
Graziano *et al.,* 2014. Italy, RCT	All	IG: 41 CG: 41	42.3 (5.2), 66% 38.3 (10.1), 60%	8.6 (5.2) 7.2 (5.3)	Cognitive behavioural program; group, in-person	2 hr; 4 sessions over 8 weeks, and 1 session at 6 month follow-up	Information sessions; group, in-person	Depression Psychological wellbeing QoL	Depression (CES-D): no significant difference (P = 0.224) QoL (MSQOL-54): no significant difference (P > 0.05)	NR
Grossman *et al.,* 2010. Switzerland, RCT	RR and SP	IG: 76 CG: 74	45.9 (10.0), 78% 48.7 (10.6), 81%	7.7 (0.9) 9.7 (0.9)	Mindfulness-based intervention (MBI), based on mindfulness-based stress reduction; group, in-person	2.5 hr; 8 sessions over 8 weeks, and one 7-hr session at week 6	Usual care	Depression Fatigue Quality of Life	Depression (CES-D): significant improvement in IG (P < 0.001) Anxiety (STAI): significant improvement in IG (P < 0.001) QoL (HAQUAMS and PQOLC): significant improvement in IG (HAQUAMS P < 0.001; PQOLC P < 0.001)	NR
Haji-Adineh *et al.,* 2019. Iran, RCT	NR	IG: 15 CG: 15	33.1 (9.1), 53% 31.5 (12.5), 53%	Minimum 1 y[1]	Mindfulness-based cognitive therapy; group, in-person	90 min; 8 sessions over 8 weeks	Usual care	Depression Life expectancy	Depression (BDI): significant improvement in IG (P < 0.001)	NR

Hoogerwerf *et al.,* 2017. Netherlands, Quasi-controlled trial	RR and SP	IG: 55 CG: 59	48.0 (8.5), 83%[1]	11.0 (8.2)[1]	Modified mindfulness-based cognitive therapy; group, in-person	2.5 hr; 8 sessions over 10 weeks	Waitlist control[2]	Fatigue	Depression (HADS): significant improvement in IG ($P < 0.001$) Anxiety (HADS): significant improvement in IG ($P < 0.001$) QoL (LiSat-9): no significant difference ($P = 0.220$)	NR
Khayeri *et al.,* 2016. Iran, RCT	NR	IG: 70 CG: 70	49.3 (6.8), 57.6%[1]	NR	Fordyce Happiness Model; group, in-person	1.5-2 hr; 8 sessions over 4 weeks	Usual care	Anxiety Depression Stress	Depression (DASS-21): significant improvement in IG ($P = 0.04$) Anxiety (DASS-21): no significant difference ($P = 0.07$) Stress (DASS-21): no significant difference ($P = 0.09$)	NR
Kolahkaj & Zargar, 2015. Iran, RCT	NR	IG: 24 CG: 24	5.8 (25.7), 100% 2.4 (24.8), 100%	NR	Mindfulness-based stress reduction; group, in-person	2 hr; 8 sessions over 8 weeks	Usual care	Anxiety Depression Stress	Depression (DASS-21): significant improvement in IG ($P < 0.001$) Anxiety (DASS-21): significant improvement in IG ($P < 0.001$) Stress (DASS-21): significant improvement in IG ($P < 0.001$)	NR
Lincoln, 2011. UK, RCT	All	IG: 72 CG: 79	44.5 (11.1), NR 47.5 (10.5), NR	9.2 (7.8) 10.5 (8.0)	Adjustment to MS program; group, in-person	2 hr; 6 sessions over 12 weeks	Waitlist	Mood	Depression (BDI-II and HADS): significant improvement in IG (BDI-II $P = 0.001$; HADS $P = 0.008$) Anxiety (HADS): significant improvement in IG ($P = 0.028$) QoL (EQ-5D): significant improvement in IG ($P = 0.041$)	Cognitive behavioral therapy principles
Nordin & Rorsman, 2012. Sweden, RCT	RR and SP	IG: 11 CG: 10	43.0 (9)[3], 73% 48.5 (7)[3], 80%	5 (10)[3] 9 (16)[3]	Acceptance and commitment therapy; group, in-person	NR; 5 sessions over 15 weeks	Relaxation training	Anxiety Depression	Depression (BDI and HADS): significant improvement in CG for HADS ($P < 0.05$). No significant difference for BDI ($P > 0.05$) Anxiety (HADS): no significant difference ($P > 0.05$)	NR

Pahlavanzadeh *et al.,* 2017. Iran, RCT	NR	IG: 35 CG: 35	NR, 100%[1]	NR	Cognitive behavioural therapy; group, in-person	90 min; 8 sessions over 8 weeks	Usual care	Anxiety Depression Stress	Depression (DASS-21): significant improvement in IG ($P < 0.001$) Anxiety (DASS-21): significant improvement in IG ($P < 0.001$) Stress (DASS-21): significant improvement in IG ($P < 0.001$)	NR
Rigby *et al.,* 2008. UK, RCT	NR	IG: 44 CG1: 42 CG2: 52	44 (9.6), 63%[1]	9 (7.5)[1]	Brief psychosocial intervention plus information booklet; group, in-person	90 min; 3 sessions over 3 weeks	CG1: Social discussion group plus information booklet CG2: Information booklet only	Mood Self-efficacy	Depression (HADS): no significant difference (area under curve $P = 0.153$, includes 12 month follow-up) Anxiety (HADS): No significant difference between IG and CG1 ($P < 0.05$). Significant improvement in IG compared to CG2 (area under curve $P < 0.01$, includes 12 month follow-up)	NR
Sanaeinasab *et al.,* 2017. Iran, RCT	NR	IG: 40 CG: 40	29.4 (7.5), 100% 32.0 (5.9), 100%	4.8 (3.5)[1]	Lazaraus and Folkman's transactional model of stress and coping program; group, in-person	1 hr; 6 sessions over 6 weeks	Usual care	Coping Stress	Stress (PSS): significant improvement in IG ($P < 0.001$)	NR
Schwartz, 1999. USA, RCT	RR and Progressive	IG: 64 CG: 68	43.0 (9.0), 73%[1]	7.3 (6.8) 8.6 (6.4)	Coping skills group plus monthly peer phone-calls; group, in-person	2 hr; 8 sessions over 8 weeks, plus monthly phone-calls for 10 additional months	Peer telephone support, monthly for 12 months (15 min duration)	Coping skills	Depression (AIMS): no significant difference ($P > 0.05$) Anxiety (AIMS): no significant difference ($P > 0.05$)	NR
Senders *et al.,* 2018. USA, RCT	All	IG: 33	53.2 (10.7), 85%	14.6 (10.1)	Mindfulness-based stress reduction; group, in-person	2 hr; 8 sessions over 8 weeks, plus	MS Education program; 2-hr classes over 8	Feasibility	Depression (PROMIS): no significant difference ($P = 0.18$) Anxiety (PROMIS): no significant difference ($P = 0.13$)	NR

		CG: 29	52.6 (12.3), 69%	17.9 (11.2)		a 6-hr retreat at week 6	weeks, plus a 6-hr retreat at week 6		QoL (SF-36, emotional well-being): no significant difference (P = 0.15) Stress (PSS): no significant difference (P = 0.30)	
Shahdadi *et al.,* 2017. Iran, RCT	NR	IG: 39 CG: 39	34.1 (8.2), 79% 35.6 (8.4), 67%	4.9 (5.7) 3.6 (4.8)	Self-care program based on Orem's self-care model; NR	45 min; 9 sessions over 2 weeks	Usual care	Stress	Stress (DASS-21): significant improvement in IG (P < 0.001)	NR
Simpson *et al.,* 2017. UK, RCT	All	IG: 25 CG: 25	43.6 (10.7), 92% 46.3 (11.1), 88%	8.9 (8.5) 9.6 (9.4)	Mindfulness-based stress reduction; group, in-person	2.5 hr; 8 sessions over 8 weeks	Waitlist control	Feasibility Stress QoL	Depression (MHI): significant improvement in IG (P < 0.05) Anxiety (MHI-18): borderline significant improvement in IG (P = 0.05) QoL (EQ-5D): no significant difference (P = 0.48) Stress (PSS): significant improvement in IG (P < 0.05)	NR
Stuifbergen *et al.,* 2003. USA, RCT	All	IG: 56 CG: 57	45.8 (10.1), 100%[1]	10.8 (6.9)[1]	Wellness program; group, in-person	1.5 hr; 8 sessions over 8 weeks, or, 3 hr; 4 sessions over 8 weeks. Plus bimonthly phone-call	Waitlist control	Self-efficacy for health behaviours Health promotion behaviours QoL	QoL (SF-36, mental health): significant improvements in IG points (combined 8 month follow-up, P < 0.05)	Health belief model, Pedner model of health promotion, and self-efficacy theory
Tesar *et al.,* 2003. Austria, Quasi-controlled trial	NR	IG: 14 CG: 15	38.2 (3.2), 86% 35.7 (9.9), 87%	5.1 (3.2) 4.2 (3.2)	Psychological therapy program; group, in-person	90 min,; 7 sessions over 7 weeks	Usual care	Anxiety Coping Depression on body image	Depression (BDI): no significant difference (P < 0.05) Anxiety (STAI): no significant difference (P < 0.05)	NR

[1]total study sample data reported (intervention and control not reported separately)
[2]control group enrolled into intervention after serving a waiting period

[3]median (interquartile range)
AIMS, Arthritis Impact Measurement Scales; BDI, Beck Depression Inventory; BDI-II, Beck Depression Inventory II; CES-D, Center for Epidemiologic Studies Depression Scale; CG, comparator group; DASS-21, Depression, Anxiety and Stress Scales; EQ-5D, EuroQol; HADS, Hospital Anxiety and Depression Scale; HAQUAMS, Hamburg Quality of Life Questionnaire in Multiple Sclerosis; HPLP-II, Health-Promoting Lifestyle Profile-II; IG, intervention group; LiSat-9, Life Satisfaction Questionnaire; MHI-18, Mental Health Inventory; MS, multiple sclerosis; MSQOL-54, Multiple Sclerosis Quality of Life-54; NR, none reported; PHQ-9, Patient Health Questionnaire; PQOLC, Profile of Health-Related Quality of Life in Chronic Disorders; PROMIS, Patient-Reported Outcomes Measurement Information System; PSS, Perceived Stress Scale; QoL, Quality of life; RCT, randomized controlled trial; RR, relapsing-remitting; SD, standard deviation; SF-8, Medical Outcomes Study 8-Item Short-Form Health Survey; SF-12, Medical Outcomes Study 12-Item Short-Form Health Survey; SF-36, Medical Outcomes Study 36-Item Short-Form Health Survey; SP, secondary progressive; STAI, State-Trait Anxiety Inventory.

Chapter 5. The perceived role of neurologists in providing dietary advice to people with multiple sclerosis

Thesis objective addressed in this chapter:

Objective 3: To explore the perceptions and experiences of neurologists in giving dietary advice and of pwMS in receiving dietary advice.

The content of this chapter is covered by Publication 3:

Russell, R. D., Black, L. J., & Begley, A. The unresolved role of the neurologist in providing dietary advice to people with multiple sclerosis. *Mult Scler Relat Disord*. 2020;44:102304. https://doi.org/10.1016/j.msard.2020.102304

The version that appears in this thesis is of an article that has been through peer-review with *Multiple Sclerosis and Related Disorders* but has not been through the copyediting process.

The contribution of co-authors, A/Prof Andrea Begley and A/Prof Lucinda Black are detailed in the author attribution statements in Appendix A: Attribution statements.

See Appendix E for the study consent form, participant information statement, and demographic questionnaire.

The unresolved role of the neurologist in providing dietary advice to people with multiple sclerosis

R. D. Russell[a], L. J. Black[a], A. Begley[a]

Affiliations:

[a]School of Public Health, Curtin University, Perth, Australia

Corresponding Author:

Andrea Begley

School of Public Health, Kent St, Bentley WA 6102

a.begley@curtin.edu.au

+61 8 9266 2773

Declaration of Conflicting Interests

The authors declare that there is no conflict of interest.

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Author contributions

R. D. Russell: Conceptualization, Formal analysis, Investigation, Writing - Original Draft, Visualization, Project administration.

L. J. Black: Conceptualization, Supervision, Writing - Review & Editing, Validation.

A. Begley: Conceptualization, Supervision, Writing - Review & Editing, Validation.

Highlights

- Dietary advice provided to patients ranged from none to counselling about specific diets
- Neurologists felt they had little influence over their patients' dietary habits
- Neurologists could direct patients to evidence-based resources (national dietary guidelines)

Abstract

Background: People with MS often make dietary changes after diagnosis with the aim of slowing disease progression. Although people with MS commonly use the internet for information on diet and MS, neurologists are their preferred source of information. However, little is known about what dietary advice is provided by neurologists.

Objectives: To explore the perceptions of neurologists about diet and MS, and to identify the type of dietary advice they provide to their patients with MS.

Methods: In this exploratory qualitative study, 11 semi-structured interviews were conducted with neurologists in Western Australia. Audio files were transcribed verbatim, and transcripts were thematically analysed using a general inductive approach.

Results: Four themes emerged: 1) juggling the evidence on the role of diet in MS; 2) acknowledging the risks and benefits of specific diets; 3) distancing from the diet 'gurus'; and 4) the unresolved role of the neurologist in providing dietary advice.

Conclusion: Neurologists could meet their patients' expectations by providing evidence-based dietary advice, such as promoting the benefits of diets that adhere to national dietary guidelines, and being prepared to explain potential risks of restrictive diets. Information about healthy eating needs to be targeted to people with MS.

Keywords: Diet; dietary advice; dietary guidelines; multiple sclerosis; nutrition; neurologists

1. Introduction

Despite the lack of scientific evidence for any therapeutic diet(1), approximately 40% of people with MS (pwMS) report making dietary modifications after being diagnosed(2-4), often with the aim of slowing disease progression(5). For pwMS, the internet is the most common source of information on diet(6, 7), where a number of specific diets are promoted. Many of these diets are restrictive, i.e. they do not satisfy the minimum nutrient requirements according to national dietary guidelines. A recent scoping review highlighted the wide range of non-evidenced-based online dietary advice promoted for pwMS, much of which was contradictory(7). Popular restrictive diets vary markedly in composition and include the Swank Diet (saturated fat restricted to <15 g/day; unsaturated fat restricted to <20-50 g/day; limited red meat)(8), the Overcoming MS Recovery Program (low in saturated fat; moderate in seafood, avocado and nuts; no meat, dairy, egg yolks, or refined foods)(9), and the Paleo diet (high in meat, vegetables, and fruits; no dairy, legumes, eggs, or grains)(1). PwMS find it difficult to judge the credibility of online dietary information, but generally trust professionals with reputable titles, such as doctors (even when the diets are not evidence-based)(5). Although the internet is commonly used for information on diet, neurologists are the preferred and most trusted source of health information for pwMS(10, 11) and, therefore, should be prepared to offer evidence-based dietary advice, and/or refer to a dietitian where required.

In Australia, people newly diagnosed with MS have reported that their neurologist provided either no information, or very little information, about diet(5). When dietary advice was provided, it was likely to be general and not tailored to MS(5); however, pwMS consider generic health information to be of little use(12). PwMS want neurologists to proactively provide information about all aspects of the disease(13), since accurate information can help with decision-making and self-management of the disease(14). Given that little is known about the dietary advice provided by neurologists to pwMS, we aimed to explore the perceptions of neurologists about diet and MS, and to identify the type of dietary advice they provide to their patients with MS.

2. Methods

We conducted an exploratory qualitative study, with methods guided by a general inductive approach (where raw data was used to derive themes)(15). The study was conducted in Perth, Western Australia (WA), between June 2019 and March 2020, and

was approved by the Human Research Ethics Committee at Curtin University (approval number HRE2019-0179). Prior to commencing interviews, participants were given written study information (containing information on the expected duration, study aims, and anonymity) and provided written, informed consent. We adhered to the Standards for Reporting Qualitative Research (SRQR)(16).

2.1 Participant recruitment

We estimated that there were 40-50 neurologists working in WA at the time of recruitment(17). Neurologists who were diagnosing and/or treating pwMS in WA were eligible. There were no exclusion criteria. We used purposive and snowball sampling methods(18). After discussions with MS Western Australia (MSWA) and the project stakeholder advisory group, we created an initial list of potential participants from both public and private health sectors. Additional participants were identified by using an online search engine (Google), and participants were asked at the conclusion of the interview for names of other neurologists to contact. Neurologists were contacted by email or phone, and invited to participate in a single interview. We encouraged neurologists to participate even if they provided little or no dietary advice to their MS patients.

2.2 Data collection

The interview topic guide (Table 1) was developed with input from both the research team and from relevant literature(19, 20). Given the time constraints for the neurologists, we aimed to complete the interviews within 30 minutes. Two authors (R.D.R., nutritionist and PhD student, and A.B., dietitian and qualitative researcher) piloted the topic guide with one neurologist for interview duration and suitability of the questions. The topic guide was not altered after piloting; hence, the transcript of the pilot interview was included in the analysis. At the beginning of the interviews, the study aims were discussed and demographic information was collected using a short questionnaire. Interviews were audio-recorded, transcribed verbatim, and rechecked for accuracy by R.D.R. The interviewer (R.D.R.) made notes after each interview, outlining emotional responses, key phrases, and topics to probe in subsequent interviews. For reflexivity(18), R.D.R. reflected on her assumptions. To ensure that comments reflected the practices and opinions of the neurologists, transcripts were posted to participants for member checking(18). No amended transcripts were returned. Recruitment continued until thematic saturation was reached (i.e. no new themes emerged from the data)(21).

Table 1 Interview topic guide

Topics	Discussion guide
Diets for MS	In your clinical experience, what is your take on the role of diet and MS? (Probe: to elaborate further; why/why not?) Can you tell me about any diets that you have heard about or read about for MS? There are some people with medical training, including in neurology, that promote specific diets for MS. What are your views on that?
Provision of dietary advice and information	When you are diagnosing a patient with MS, what emphasis do you place on diet; where does dietary information sit? Is there anything you routinely discuss about diet with your patients? (Probe why/why not?) How often would you say that your patients have questions for you about diet? • What (if anything) do they typically ask of you? What information do you provide about diet, and why? (If none, then why not?) • Are there any resources you give out, or direct patients to? • What [additional] resources would you like to be able to refer client to? I.e. where should your patients get nutrition information from? If a patient were to come to you, and insist on following a specific diet for their MS, and choosing to do a diet over taking medication, how would you typically respond to that? (Follow-up: how would you respond if that patient agreed to take the recommended medication, but was still insistent on following the specific diet?) • What do you perceive to be some of the benefits to them following a specific diet if they are continuing to take their recommended medication? • What do you perceive to be some of the risks to them following a specific diet if they are continuing to take their recommended medication? As their neurologist, what degree of influence do you think you have on your patients' dietary habits if you were to make suggestions or recommendations?
Conclusion	Anything else you would like to add that I have not covered?

2.3 Analysis

Commencing after the first interview, the interviewer (R.D.R.) used inductive thematic analysis methods(15) to analyse the transcripts. The transcripts were read in detail to identify sections of text that related to the objectives, creating 39 categories in the initial coding stage. We used literal (direct observations from the data) and interpretive (inferred from the data) coding techniques(22). For the second stage of coding, categories were reduced and refined by grouping those with similar meanings, resulting in 15 categories. Final revision involved reducing redundant categories and creating relevant subcategories. This resulted in four overarching themes.

The research team discussed emerging themes throughout the analysis process as a means of peer debriefing(18). This encouraged new perspectives from those who were external to the data collection and not immersed in the data, ensuring credibility of the findings. A.B. (an experienced qualitative researcher) listened to all audio recordings, and L.J.B. acted as an external auditor (was not involved in data collection or preliminary analysis). Final themes were confirmed by the research team. NVivo software (version 12.6.0, QSR International Pty Ltd) was used for qualitative data management. Stata software (version 16.0, StataCorp LP, College Station, TX, USA) was used to analyse participant characteristics.

3. Results

3.1 Participants

We interviewed 11 neurologists: eight interviews were conducted in person and three were conducted using Skype (version 7.58, Microsoft Corp., Luxembourg). The mean interview duration was 26 minutes (range, 18-42 minutes). Participant characteristics are presented in Table 2. The majority were male (64%), with equal representation across the public and private sectors (36% public; 36% private; 27% a combination of both). The median number of years since qualifying was 12 years (range, 8-27 years). The proportion of pwMS treated by neurologists in the previous three working days ranged from zero to 50% (median, 5%).

Table 2 Characteristics of the participants

Sex, *n* (%)	
Male	*7* (63.6%)
Female	*4* (36.4%)
Health sector, *n* (%)	
Public	*4* (36.4%)
Private	*4* (36.4%)
Both	*3* (27.3%)
Location of practice	
Perth[1] metropolitan only	*6* (54.5%)
Perth[1] metropolitan and regional Western Australia	*5* (45.5%)
Country of medical training[2], n (%)	
Australia	*9* (81.8%)
Other	*6* (54.5%)
Number of years practicing medicine	
Median (IQR, range)	22 (14, 19-38)
Number of years as a qualified neurologist	
Median (IQR, range)	12 (13, 8-27)
Percentage of MS patients seen in previous three working days	
Median (range)	5% (0-50%)

IQR, interquartile range
[1]Capital city of Western Australia
[2]Participants could record multiple countries

3.2 Themes

Inductive thematic analysis resulted in four overarching themes: 1) juggling the evidence on the role of diet in MS; 2) acknowledging the risks and benefits of specific diets; 3) distancing from the diet 'gurus'; and 4) the unresolved role of the neurologist in providing dietary advice. Quotes to support themes are presented in Table 3.

3.2.1 Theme 1 Juggling the evidence on the role of diet in MS

In most cases, the dietary advice given by neurologists to pwMS was informed by credible sources, such as peer-reviewed journals, MS conferences, and dietitians. However, when healthy eating was mentioned, the descriptions were vague; *"I just tell them to eat well"* (N7), and there was no reference to an evidence-based resource, such as the *Australian Dietary Guidelines*. The advice was inconsistent with no apparent clinical consensus.

Some neurologists relied on anecdotes from other patients, or on their own personal food preferences, to inform the dietary advice they provided.

The viewpoints of the neurologists on the role of diet ranged from sceptics (did not see a role for diet in general) through to those who counselled patients with information about specific diets (e.g. Mediterranean, vegan, or the Overcoming MS Recovery Program diet). Some neurologists were *"deliberately vague"* (N1) when giving dietary advice. Reasons for providing little or no dietary advice included limited knowledge about diet/nutrition or being unsure what the right advice was, given the lack of evidence. Neurologists wanted to see more robust evidence in the field of diet and MS before they could be confident enough to make any dietary recommendations or to counsel for/against specific restrictive diets. Neurologists who emphasised the importance of diet cited general health, weight management, vascular health, and a sense of wellbeing, as reasons for giving dietary advice.

There was very little discussion on the role of diet in managing the symptoms of MS, and any such discussion was limited to the symptoms of constipation and fatigue. A few neurologists avoided discussing diet if their patients indicated they were already following a restrictive diet. Contradicting patients with information that the diet was not evidence-based was considered confronting, and was avoided. Rarely, neurologists were openly supportive of their patients trying specific diets, and spent time helping them make an informed choice.

Table 3 Participant quotes to support themes

Theme 1 Juggling the evidence on the role of diet in MS
"I might say, 'You know, I've had some patients actually come off treatment and done really well on that [particular diet]'." N3
"Until there is some concrete evidence about [a] specific diet, um, I, I would, I'd be a sceptic." N5
"A lot of people do go on to specific diets and they ask me, you know, what I recommend. And I'm deliberately vague, because I don't really know for sure. There's not enough evidence to be confident of what diet is, is appropriate." N1
"[When] the patients come to me, I never really give them much information on diet. I just tell them to eat well." N7
"People don't like to be contradicted. So if someone thinks a low-fat diet will cure my MS, there's no point in me saying that's a pile of crap." N10
"I'd have to look at the diet, see what's known about the diet, and then discuss it with the patient. So I am, I am open minded to this." N4
Theme 2 Acknowledging the risks and benefits of specific diets
"General cardiovascular health, general cerebrovascular health, […] not being overweight, ah, just general health issues. Which actually translates into a good immune system, and looking after yourself." N2
"If I think they're very limiting, so some go on like a vegan one, ah, I just ensure that they get all the vitamins. I prescribe them multivitamins or get them to buy them." N8
"Suddenly someone who can't touch or eat anything at anyone's place, you're changing how you present to your friends, to your family, to your children. Ah, and I think that's a problem. You want a diet that integrates with society." N2
"'Well must be your fault. It must be that you didn't stick to the diet'. Which can become damaging for peoples ah, you know, mental health, if they think the MS getting worse is their fault." N11
"Yeah, I mean, the thing with neurological conditions, including MS is, things feel like they're out of control OK. You're losing control of your limb or your vision or whatever. So, so sometimes diet is a form of control." N8
"Some patients that are overweight with MS can, can lose weight on those diets, and obesity is a risk factor for um, poor prognosis in MS." N11
Theme 3 Distancing from the diet 'gurus'
"[People] strongly believe what they're told by, you know, by certain, you know Terry Wahls[1] or George Jelinek[2] because they are very, you know, they're very sort of um, concrete. And 'this is this is right, and correct', and people like that certainty." N1
"It all has to be science-based. It can't be religion-based, it can't be faith-based. You know, we've got to collect evidence, find the evidence." N10
"I think it's very common for people to be caught up in the zeitgeist, and be, 'Okay, I'm gonna go vegan, dairy-free, gluten-free', even though they don't have intolerance to those things." N1
"Dr Terry Wahls in North America, same idea, slightly guru paradigm, someone comes along and says I've got MS and I eat bananas, and you know, cured MS." N10
Theme 4 The unresolved role of the neurologist in providing dietary advice

"I don't know that I have that much influence over what people do in terms of their diet." N11

"Usually unless they ask directly about it [diet], I don't address it. There's just not enough time in the day." N9

"They might have the intent to stick to a very restrictive diet, but mostly they can't […] they self-protect in a way." N8

"Honestly, I would wait for the patient to bring it up, um, except for vitamin D, I don't bring up anything else about diet." N5

"[If] it gets to a point where I think that they need like really good dietary advice, I will send them to a dietitian." N4

"[If patients ask] whether any particular type of diet would be helpful or not […] my usual suggestion is that you can try that and you can see if it works." N5

N[x], neurologist ID code
[1]Dr Wahls promotes the Wahls Paleo Protocol
[2]Professor Jelinek promotes the Overcoming MS Recovery Program

3.2.2 Theme 2 Acknowledging the risks and benefits of specific diets

Nutrient deficiencies and unintended weight loss were the most commonly mentioned risks associated with specific diets, although probing was needed to extract this topic of discussion. For patients where nutrient deficiencies were a concern, half of the neurologists said they would recommend a dietary supplement. Patients who were committed to following a restrictive diet were rarely referred to a dietitian. Only one neurologist mentioned ordering blood tests to ensure their patients on restrictive diets were not suffering from nutrient deficiencies. Any potential deleterious effects on mental health were rarely discussed. A few neurologists considered restrictive diets to be socially isolating, and two discussed the guilt felt by pwMS if they strayed from the diet. Those neurologists noted that patients often blamed their relapses on poor adherence to their chosen diet.

Most neurologists acknowledged that certain diets could be beneficial, including low-fat, Mediterranean, vegan, and restrictive (e.g. Paleo) diets, as they were likely to be healthier than the patient's previous diet. Commonly cited benefits of such diets included increased energy levels, improved heart and/or brain health, weight loss, having a sense of control, and generally feeling better.

3.2.3 Theme 3 Distancing from the diet 'gurus'

Given the lack of evidence for altering the disease course, most neurologists were dismissive of the restrictive diets. However, they thought that their patients considered the dietary advice from the *"gurus"* (N10) as clear, concise, and trustworthy. The language used when referring to restrictive diets and their promotors was distancing and dismissive, with references to pwMS having faith and belief in the 'gurus'.

The neurologists were more likely to discuss restrictive diets than to discuss what constitutes a healthy diet. However, since non-evidence-based restrictive diets were seen to reflect the *"zeitgeist"* (N1) of the time, they were disregarded by neurologists as fads that come and go. As a result, not all neurologists engaged in discussions about diet with their patients, and some were unaware of which restrictive diets were popular with pwMS.

3.2.4 Theme 4 The unresolved role of the neurologist in providing dietary advice

The role of the neurologist in providing dietary advice was uncertain. More than two-thirds believed they had very little influence, or even no influence, when it came to providing dietary advice to their patients. In contrast, they believed the diet 'gurus' to be very influential. Time constraint was a common reason for not discussing diet in any detail.

Neurologists recognised the importance of autonomy for their patients, and acknowledged that diet was a way for pwMS to have a sense of control. It was generally accepted that their patients' compliance to restrictive diets would wane over time. As such, the neurologists dismissed any potential risks. Hence, discussing the pros and cons of restrictive diets was not considered a priority.

Many neurologists felt it was not their role to provide any dietary advice other than general healthy eating information, but even so they rarely referred to the *Australian Dietary Guidelines*. Some neurologists did not provide any dietary guidance. Unless a patient specifically enquired about diet, it was often not mentioned. The tension surrounding the role of the neurologist in providing dietary advice was further amplified by the limited, and often conflicting, evidence for the role of diet in MS. In contrast to disease-modifying

therapies, there are very few randomised controlled trials assessing diet and MS progression, making it hard for neurologists to judge what impact diet has on MS. Although providing dietary advice was not considered part of their role, only a few neurologists directed patients to MS organisations or dietitians for dietary assessment and counselling.

4. Discussion

This is the first study to explore the perceptions of neurologists about diet and MS, and identify the type of dietary advice they provide to their patients. A strength of the study was that we interviewed neurologists with a range of experience, who were consulting in both the metropolitan and regional areas of WA, and in both public and private health sectors. In summary, we found that the dietary advice provided to patients was inconsistent, with the neurologists juggling the conflicting evidence around diet and MS (theme 1). A range of risks and benefits associated with the specific diets promoted for pwMS were acknowledged (theme 2). By using dismissive language and religious expressions, the neurologists distanced themselves from the ‘gurus’ who promote restrictive diets (theme 3). In light of the first three themes, we found that the role of the neurologist in providing dietary advice was ultimately unresolved (theme 4).

The neurologists in our study did not always follow an evidence-based approach when providing dietary advice. Given the conflicting evidence around diet and MS, there was no clear consensus in the dietary advice provided by neurologists, despite MS organisations encouraging a diet in line with national dietary guidelines(23). This is consistent with a study of family physicians(24), where the physicians deviated from evidence-based medicine when the preferences of their patients conflicted with the evidence. In addition, when there is no clear consensus in the medical literature, patient perspectives are critical for enabling physicians to make decisions without restricting patient autonomy(24, 25).

Discussions about diet focussed on specific diets: some enabled pwMS to meet their nutrient requirements (e.g. Mediterranean diet), while others did not and were considered restrictive (e.g. Paleo diet). The *Australian Dietary Guidelines* were not mentioned as an example of a healthy diet, and were not considered to be a diet that could benefit pwMS. Given that less than 4% of Australians meet the recommendations of the *Australian Dietary Guidelines,* it is likely that following these guidelines would be a significant dietary change for pwMS(26). For pwMS, potential benefits of following the *Australian Dietary*

Guidelines include reduced risk of co-morbidities (such as type 2 diabetes and cardiovascular disease, which increase the risk of disability progression for pwMS(27)), weight management, less constipation, and better general health(28). Such healthy eating could promote a sense of control for pwMS, while avoiding the risks associated with restrictive diets (e.g. nutrient deficiencies(29) and financial difficulties, since such diets have been described as “expensive”(5)). The neurologists did not consider nutrient deficiencies to be an issue, as they perceived compliance to such diets to be low. Nevertheless, doctors should actively monitor the nutritional status of pwMS who follow diets that are known to be restrictive and potentially limiting in specific nutrients(30).

Few neurologists in our study discussed dietary modifications as a way for their patients to potentially manage their symptoms. Given that pwMS experiment with diet as a means of symptom management(5, 31), not addressing the potential for diet to manage MS symptoms may be incompatible with patients’ expectations. In addition, the potentially detrimental effects on mental health from following restrictive diets were rarely discussed. This may also be misaligned with patient expectations, since pwMS have reported social isolation from strict adherence to restrictive diets, and a fear of failure (worsening symptoms) if they stray from the diet(5).

Neurologists felt they had little influence over the dietary choices of their patients. This is contrary to previous literature on the influential nature of neurologists: pwMS value the perspectives of their neurologist on a range of information (beyond medical advice), and view them as a source of hope for therapeutic advances(13). Neurologists are the most important, reliable, and preferred source of MS information(11, 13, 32). Our findings suggest that neurologists may underestimate their influence regarding diet, since pwMS want information on all aspects of their disease management, including diet(11).

Overall, neurologists considered the provision of dietary advice (beyond “eat well”) to be outside their role. This is consistent with oncologists, who also think providing dietary advice is outside their role(33). This viewpoint does not match the perceptions of their patients: insufficient dietary advice provided by neurologists may be considered by pwMS as a lack of knowledge about diet(5), and a lack of reliable resources can negatively affect patient engagement(34). The neurologists were dismissive of the so-called diet ‘gurus’

(where ‘guru’ was used in a disparaging way, as a person with knowledge who advocates for their particular theory(35) and has a financial interest). Nevertheless, they thought ‘gurus’ were influential, since they provided definitive dietary advice that gave hope to pwMS. Neurologists should not underestimate their degree of influence when providing consistent, evidence-based dietary advice. Clear advice could enable neurologists to be as influential as the ‘gurus’. This would maintain their authority as the experts, while giving their patients hope.

We applied a number of techniques to ensure rigour in this study(36); however, there are some limitations. There is the potential for self-selection bias in the participant sample, where those with an interest in diet may have been more willing to participate. We attempted to overcome this by encouraging participation from a range of neurologists, regardless of whether they provided dietary advice or not. Our findings may not be generalizable outside of Australia, since other countries may have specific clinical practice guidelines with respect to diet and MS.

5. Conclusion

PwMS prefer to receive information about MS from their neurologists than from any other source. Neurologists could meet the expectations of their patients with MS by providing evidence-based dietary advice. They could direct patients to resources such as the *Australian Dietary Guidelines* and highlight the ways that this diet may be beneficial, while discussing the potential risks of restrictive diets and referring patients to dietitians. Future resources could be developed to help neurologists illustrate the potential benefits of the *Australian Dietary Guidelines* for pwMS. Further research is needed to elucidate how neurologists could provide dietary advice within their current time constraints, and improve their self-perceived influence regarding giving dietary advice.

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Chapter 6. How people with multiple sclerosis navigate dietary advice

Thesis objective addressed in this chapter:

Objective 3: To explore the perceptions and experiences of neurologists in giving dietary advice and of pwMS in receiving dietary advice.

The content of this chapter is covered by Publication 4:

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The contribution of co-authors, A/Prof Andrea Begley and A/Prof Lucinda Black are detailed in the author attribution statements in Appendix A: Attribution statements.

See Appendix F for the study consent form, participant information statement, demographic questionnaire, and program preference questionnaire.

Original article: *Health Expectations*

Navigating dietary advice for multiple sclerosis

Short running title: Navigating diet and multiple sclerosis

Rebecca. D. Russell[1], Lucinda. J. Black[1], Andrea Begley[1]

Affiliations: [1]School of Public Health, Curtin University, Perth, Australia

Corresponding Author

Andrea Begley

a.begley@curtin.edu.au

+61 8 9266 2773

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Conflict of interest

The authors declare no conflicts of interest.

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Abstract

Background: Multiple sclerosis (MS) is an inflammatory demyelinating disease with no known cure. Numerous diets are promoted to reduce symptoms or even cure MS, despite insufficient evidence for any therapeutic diet. There are few qualitative studies exploring the experiences of people with MS in relation to diet, and no use of theory to explain the findings.

Purpose: To explore the experiences of adults with MS when navigating dietary advice, their attitudes when making dietary decisions, and their needs regarding dietary resources and education.

Methods: In this qualitative study, we conducted six focus groups with people with MS (n=33 plus one spouse without MS). Groups were audio-recorded, transcribed verbatim. Primary analysis used a general inductive approach with thematic analysis. Secondary analysis aligned themes with the constructs of the self-determination theory.

Results: Six themes emerged: 1) confusion about where to seek dietary advice; 2) scepticism towards national dietary guidelines; 3) personalised approaches to dietary change; 4) barriers to dietary changes; 5) judging if dietary changes work; and 6) wanting dietary guidelines for MS.

Conclusion: People with MS are highly motivated to make dietary changes and improve their health. The self-determination theory explained why people with MS make dietary modifications, and the varying levels of motivation. MS-specific dietary resources and nutrition education need to incorporate ways to increase autonomous forms of motivation. Future dietary intervention studies could use the self-determination theory as a framework to improve long-term adherence to healthier diets.

Keywords: autonomy; diet; dietary modifications; focus groups; self-determination theory; motivation; multiple sclerosis; qualitative

Patient or Public Contribution

Members of a stakeholder advisory group, which included a neurologist, MS counsellor, MS nurse, psychologist, dietitians, and MS consumers, provided input during the development of the question guide and methods for participant recruitment. One MS consumer provided feedback after the pilot focus group session.

Introduction

Multiple sclerosis (MS) is an inflammatory demyelinating disease of the central nervous system with no known cure.[1] Immune-mediated attacks cause inflammation and damage to the myelin sheaths, interrupting nerve signal transmission.[2] Any of the sensory, visual, or motor systems can be affected, causing symptoms that vary widely between individuals and over time.[3] MS affects more than 25,000 Australians, and approximately 2.3 million people worldwide; three-quarters of those are female.3 The most common form of MS is relapsing-remitting MS, where periods of neurological decline are followed by periods of remission.[4] Over time this may progress to secondary progressive MS (half of cases over 10 years5), where deterioration is ongoing. Less common is primary progressive MS (occurring in 10-15% of cases), where deterioration is from the onset, and there are no remissions.[4]

Although diet has been proposed as a potential modifiable risk factor to reduce MS symptom severity and/or slow disability progression,[6] there is insufficient evidence to support any specific therapeutic diets.[7] As such, the dietary advice for people with MS (pwMS) is to follow Government-issued national dietary guidelines. This is to reduce the risk of comorbid diseases (such as cardiovascular disease and type 2 diabetes) and ensure optimum nutritional status. This is imperative, as vascular co-morbidities have been associated with increased disability progression,[8] and some nutrient deficiencies can exacerbate symptoms and accelerate demyelination.[9] The food group and nutrient intake recommendations in the Australian Dietary Guidelines (an example of national dietary guidelines) can be achieved with a range of dietary patterns, including vegetarian, vegan, and Mediterranean diets.[10] Unfortunately, less than 4% of Australians achieve these food group and nutrient recommendations.[11] To our knowledge, there is no literature reporting how many pwMS follow national dietary guidelines.

There are numerous non-evidence-based diets promoted online, claiming to reduce MS symptoms, slow MS progression, or cure MS.[12] This creates a challenge for pwMS when deciding what foods to eat, given that the diets are often contradictory[12] and restrictive, i.e. they don’t meet the minimum nutrient requirements outlined in national dietary guidelines. There is an opportunity to provide tailored education to assist pwMS in decision-making

and meal planning in order to improve dietary intakes. Dietary education for pwMS needs to take into account factors such as food preferences, budgets, and food literacy skills.

Quantitative studies show that more than 80% of pwMS consider diet to be important,[13] and around 40% report making dietary modifications after their diagnosis.[13-16] Reducing symptoms or number of relapses, losing weight, having a sense of control, slowing disease progression, and curing themselves of MS are common reasons why pwMS make dietary modifications.[13,17] The most common dietary changes described are adopting a low-fat[14-16] or low-carbohydrate diet,[16] modifying fatty acid intake,[13] eliminating meat intake,[13] decreasing sugar intake,[15] and increasing fruit and/or vegetable consumption.[14] Such modifications are not always evidence-based, or in line with national dietary guidelines. While there is literature capturing what specific dietary changes are made by pwMS, little is known about why pwMS make non-evidence-based dietary modifications, or what would motivate them to increase adherence to a healthy diet.

Only two qualitative studies have explored the rationale behind the dietary modifications made by pwMS. Fatigue and limited mobility have been reported as barriers to engaging in healthy dietary behaviours.[18] In a previous study, people recently diagnosed with MS expressed that a lack of dietary advice from neurologists was incompatible with the seriousness of the disease, and experimented with dietary modifications to control or cure their MS.[17] There has been little theoretical explanation as to why pwMS make and adhere to any type of dietary modification. There are very few information provision interventions for pwMS that have been based on theoretical frameworks,[19] despite this being recommended as best practice.[20] Theoretical models are useful for understanding behaviour change and maintenance, and for developing interventions and strategies for behaviour change.[21] In the field of physical activity and MS, the concepts of self-determination theory (SDT), focusing on types of motivations, have been applied to better understand physical activity behaviours and adherence in pwMS,[22,23] and social cognitive theory has been used to develop a physical activity intervention.[24]

Exploring the motivations and barriers for healthy dietary behaviours in pwMS would aid in developing evidence-based dietary resources and interventions for pwMS. These should aim to help pwMS achieve national dietary guideline recommendations, thus reducing the

risk of co-morbidities and potentially improving quality of life.[25] The aims of this research were to explore the experiences of adults with MS when navigating dietary advice, their attitudes when making dietary decisions, and their needs regarding dietary resources and education.

Methods

This study was approved by the Human Research Ethics Committee at Curtin University (approval HRE2019-0179). Given the paucity of qualitative literature in the field of diet and MS, we used a general inductive approach to guide this research, where themes were derived from interpretations of the raw data26 and reviewed for connections to theoretical frameworks.[27] This allowed the analysis to be guided by the objectives, and ensured participant responses were not influenced by predetermined hypotheses.[28] Focus groups were conducted between July 2019 and March 2020 in Western Australia. The research information statement (outlining the study aims, expected duration, and anonymity) was provided to participants before the focus groups commenced. Participants provided written, informed consent. We adhered to the Consolidated Criteria for Reporting Qualitative Research (COREQ).[29]

Participants and recruitment

Participants were eligible for inclusion if they were English-speaking adults (age ≥ 18 years) and had been diagnosed with MS. There were no exclusion criteria. We used purposive sampling to recruit participants from a local MS organisation (MS Western Australia (MSWA)) and networks of the project stakeholder advisory group (which included two MS consumer representatives). The study was advertised by MSWA via emails to the member database and social media postings. Potential participants were invited to take part in a single focus group. Participants were given an AUD$20 department store voucher as remuneration. No participants withdrew from the study after attending a focus group.

Data collection

We aimed to conduct 5-6 focus groups with 5-8 participants per group. The focus groups were facilitated by RDR (BSc(Hons)), with one of either AB (DrPH) or LJB (PhD) as co-facilitators. The topic guide (Table 1) was developed with input from both the relevant literature[17,30,31] and the research team, and was piloted to test the suitability of the questions by AB (dietitian and qualitative researcher) and RDR (nutritionist and PhD student). As a result of piloting, the topic guide was considered suitable and therefore the

transcripts from the pilot group were included in analysis. Participants were asked to arrive 30 minutes before the start time to establish rapport with other participants and the researchers, since the researchers did not have existing relationships with the participants. During the focus groups, probing was used to clarify information or seek further details. Demographic information (sex, age, type of MS, and duration of MS) and nutrition program preferences (delivery mode, topics of interest, and frequency, duration, and number of sessions) were collected using two short questionnaires developed by the research team. To maintain a reflexive stance,[32] the facilitators discussed and made notes after each session to reflect on their assumptions and biases, and how their role as researchers influenced the group discussions. Memos documenting key phrases, states of mind, emotional responses, and/or questions to probe in subsequent groups were written after each group. The focus groups were audio-recorded and transcribed verbatim. Transcripts were posted to participants for member checking,[32] confirming that data represented the group discussions. Focus groups were conducted until thematic saturation was reached (i.e. no new codes emerged).[33]

Table 1 Focus group topic guide

Topics	Discussion guide
Introduction	Welcome, purpose of the research, ground rules, format, anonymity reminder. Are there any questions before we begin?
Icebreaker	Thinking about the last week, has your MS impacted on what you are eating or what you've chosen to eat?
Barriers and facilitators	Can you tell me about any ways that you may find your MS affects the way you eat? [Probe: shopping and preparing food, use of utensils or equipment, cooking methods, side effects from medications, fatigue] What sort of things do you do that make it easier for you to eat well?
Dietary information or advice	What (if any) dietary information have you asked a health professional about? Whose role is it to give out information about diet/foods for MS?
Dietary education program preferences	What would you have liked to have known about food or diet when you first found out you had MS? What topics would you like covered in an MS nutrition program? What types of things would need to happen for you to know you had made improvements, and what improvements are important to you? Have you been to any seminars related to MS? If so, what did you attend, and what did you like and not like about those events?
Wrap-up	Is there anything else about diet and MS that you want to talk about that we have not discussed?

Analysis

Transcripts were managed with NVivo (version 12.6.0, QSR International Pty Ltd). The first author used a general inductive approach[26] to thematically analyse all transcripts. Analysis commenced after the first focus group. The initial coding stage involved two authors (RDR and AB) reading the transcripts in detail and labelling text relating to each of the objectives. RDR then labelled behaviours, strategies, and states of mind using literal (direct observations) and interpretive (inferred from the data) coding techniques,[34] which included text unrelated to the objectives. This resulted in 31 initial categories. In the second stage of coding, 15 categories resulted from grouping those with similar meanings. Final revision of the data involved further grouping of categories with similar meanings and

collapsing redundant categories. RDR and AB discussed the categories and emerging themes several times during the analysis as a form of peer debriefing. This produced six main themes[32] which were confirmed by the research team. A secondary analysis was conducted by RDR, where the lens of the self-determination theory (using the constructs of autonomy, competence and relatedness) was applied to explain the themes.

Results

Participants

Thirty-four participants (33 pwMS; one spouse) attended one of six focus groups. The mean number of participants per group was six (range, four to eight). Focus group duration was between 50 and 68 minutes (mean, 60 minutes). The majority of the participants were female (82%), and the mean (SD) age was 50.2 (12.4) years. The median time since diagnosis was 6 years (range, 0.5-37 years), and the most common type of MS was relapsing-remitting (68%). See Table 2 for participant characteristics.

Table 2 Participant characteristics (*n*=34)

Sex, *n* (%)	
Female	*28* (82.4%)
Male	*6* (17.6%)
Age (years)	
Mean (SD, range)	50.2 (12.4, 27-79)
Time since diagnosis (years)†	
Median (IQR, range)	6.0 (13.5, 0.5-37)
Type of MS† *n* (%)	
Relapsing-remitting	*23* (67.6%)
Secondary progressive	*1* (2.9%)
Primary progressive	*4* (11.8%)
Unsure or other	*5* (14.7%)
Country of birth,‡ *n* (%)	
Australia	*21* (61.8%)
Overseas	*11* (32.4%)
Employment status, *n* (%)	
Employed	*17* (50%)
Disability pension	*5* (14.7%)
Retired	*6* (17.6%)
Other§	*6* (17.6%)

IQR, interquartile range; MS, multiple sclerosis; SD, standard deviation
†Participants with MS, *n*=33 (one spouse attended one focus group)
‡Missing data, *n*=2
§Other included home duties, looking for work, not working, and volunteering

Themes

Six themes emerged: 1) confusion about where to seek dietary advice; 2) scepticism towards national dietary guidelines; 3) personalised approaches to dietary change; 4) barriers to dietary changes; 5) judging if dietary changes work; and 6) wanting dietary guidelines for MS. Participant number, focus group number, and time since diagnosis are detailed after each quote.

Theme 1: Confusion about where to seek dietary advice

Participants discussed accessing dietary information from a wide range of sources: friends, family, healthcare professionals, websites, documentaries, and books. Dietitians and MS organisations were rarely mentioned. The conflicting information about diets for MS meant there was confusion about what were appropriate foods and diets. It was difficult for some participants to judge reliability; causing angst when deciding which foods include/exclude, or which specific diet to follow.

> "There are so many different diet plans and people having their two cents' worth on the internet, and it's like a minefield trying to get information that's relevant and correct." (P15, FG2, 6 years)

> "Should I go on Keto? Should I go on low-fibre? Should I do this? Can I eat a low-GI bread? Can I eat gluten-free bread? […] I just have no idea." (P31, FG6, 20 years)

Some participants were afraid that their dietary decisions may cause a relapse, and were anxious when deciding what to eat.

> "It can create a lot of anxiety because you're so frightened of, on one hand, of having a relapse. Which way do I go when there's no, um, official guidance." (P20, FG4, 2 years)

> Participants indicated an interest in what other pwMS were doing with diet, seeking confirmation from their peers about their dietary modifications.

> "Can I ask, do you do gluten-free? So I've always had this question mark over this, is this something? Why do you guys do gluten-free? (P11, FG2, 6 years)

It was discussed that neurologists and other MS health professionals did not promote any specific diets for MS. Participants thought that neurologists generally had inadequate

knowledge and/or training to give dietary advice, and their focus was on treating the disease with medication. Some participants were alarmed that neurologists were not interested in dietary modification as "preventative medicine" (P24, FG4, 10 years), which is how some restrictive MS diets are promoted. There were some comments that were conspiratorial in nature: neurologists and other MS professionals were keeping something from them and could be sued if they recommended diets other than the national dietary guidelines.

> "I said surely diet's gotta be- play a big role in this sort of thing, right? And they- it was almost like they [neurologists] were barred from saying yes." (P10, FG2, 6 years)

> "They've [neurologists] got the guidelines, and they can't sway from it, otherwise, they get sued and all sorts." (P18, FG3, 6 months)

Despite the perceived lack of training and/or interest in diet by neurologists, the participants agreed that they wanted to receive dietary advice from their neurologists.

Theme 2: Scepticism towards national dietary guidelines

Participants were sceptical as to whether the "national guidelines" (P33, FG6, 4 years) or the "healthy food pyramid" (P6, FG2, 2.5 years) were suitable for pwMS. In light of the information about diet and MS that participants were accessing online and in books, national dietary guidelines were perceived as not good enough. There were misconceptions about what was recommended within those guidelines, e.g. participants thought it was necessary to consume all foods in the guidelines, including meat, dairy, and grains. Vegetarian and vegan diets were not considered to be compliant with national dietary guidelines.

> "On that food pyramid is dairy. Well, should we be eating dairy? Or should be substituting the dairy section?" (P28, FG6, 17 years)

Some participants were frustrated and angry in response to being given the "national guidelines" or "food pyramid" as dietary advice. In some cases, there was scepticism about the suitability of the "national guidelines" for the general population, as well as for pwMS.

> "It's not a healthy diet, even though you're eating your five pieces of wholemeal bread a day, and you know, your two cups of pasta, or whatever. […] The powers

that be realised they made mistakes 40-50 years ago when they came up with the National Dietary Guidelines." (P15, FG3, 6 years)

"Absolutely. That pyramid is an absolute load of crap." (P18, FG3, 6 months)

Theme 3: Personalised approaches to dietary change

The general discussion in the groups demonstrated that most participants were highly motivated to learn about potentially beneficial dietary modifications. Some participants mentioned that they were very strict when adhering to their dietary changes, many adopted a moderation approach, and a few did not make any dietary changes. During the discussions, it became evident to the research team that part of the reason for attending was to discover what other pwMS were doing with diet, and that modifying their diets was a way for pwMS to feel in control of their disease. Some participants were convinced it would slow disease progression and help to avoid disability.

"It's something you feel you've got- that you can control […]. You can't control your MS, you know, but, you can control your diet." (P8, FG2, 37 years)

Sometimes participants were very persistent about the dietary approach they were taking and were open to sharing what had and had not worked for them. A wide range of dietary modifications were described, from small or targeted dietary changes (e.g. eating more fruits and vegetables, eliminating sugar, reducing fat intake, and/or eliminating food groups), to total dietary changes (e.g. adopting a specific diet such as the Wahls Protocol diet,[35] the Overcoming MS Recovery Program diet,[36] the ketogenic diet,[7] or the Swank diet[7]).

"My diet's changed in ways of being more aware […] Instead of going to KFC, you'll go and have a Subway because it's got salad and vegetables, and all that sort of stuff. Or you know, if you have takeaway stuff I'll have a kebab because it's got meat, it's got vegetables." (P25, FG5, 1.5 years)

"I've been dairy-free, sugar-free, gluten-free, eating nine cups of vegetables every day, sourcing you know, good quality veggies and good quality meats. Before that, I was just a regular person eatin' anything I wanted." (P18, FG3, 6 months)

Participants had different opinions about how strictly they thought they should adhere to their chosen diet, and about their capacity to sustain the changes. There was conversation about continuing to eat all foods “in moderation” (P25, FG5, 1.5 years), predominantly from those who were more recently diagnosed.

> “I am just going to try and live my life right now, and get into a least some kind of healthy pattern. I'm not gonna cut out dairy, I'm not gonna cut out those things out. I'm just gonna be more realistic about the amounts […] and what's possible for me.” (P4, FG1, 2 years)

For participants without many MS symptoms, food or diet was considered a low priority. They stated that they assumed that their neurologist would have informed them if a specific diet or dietary modification was important. Maintaining current dietary habits was a way of upholding some normality and, for some, represented a degree of denial about the perceived need to change and/or about their diagnosis.

> “I don't read about it [diet] […] I've just kind of ignored it. I'm a bit blasé about. [...] Has anybody's neurologist even given them any advice on diet? It's not something I've looked into or thought about to be honest.” (P29, FG6, 20 years)

> “I don't see any difference in my MS, so I don't- I haven't done a lot of research [about diet]. Like maybe there's still some blinders up.” (P26, FG5, 1.5 years)

At the other extreme, some participants were very serious about their chosen dietary modifications to slow their disease progression or keep their symptoms at bay. Those participants believed that following a specific diet was of the highest priority, which required a lot of time and mental effort. The choice between strictly adhering to a specific diet or not was likened to choosing between continuing to be able to walk (not ending up in a wheelchair) or eating McDonalds (fast-food).

> “Your future, it's everything. Like, if you wanna be- if you wanna, you know, eat McDonalds, or do you wanna walk? That’s kinda like the choices I made.” (P11, FG2, 6 years)

It appeared that the participants committed to personalising their diet plans in an attempt to recognise the individuality of the disease, and to cope with the conflicting dietary information. Some dietary modifications described by participants were an amalgamation of diets, creating a so-called “flexitarian” (P20, FG4, 2 years) diet. As they discussed their eating habits in the groups, it was apparent that even those claiming to follow one specific diet were incorporating aspects from other diets. There did not appear to be any practical reasoning in the decision-making process; rather a lucky dip as to what might work. As they listened to what others in the group were doing with diet, some participants were confused about which specific diet they were adhering to: “So it is really a keto [ketogenic] or Mediterranean [diet].” (P34, FG6, 6 years).

> “It’s a matter of taking a bit of that, and a bit of that, and a bit of that, and a bit of that, and like- and just piling it all into one [diet].” (P7, FG2, 22 years)

> “You really just got to find something that suits you and I’m the same. I’m well-Paleo-ish. I know about Wahls Protocol as well, but gluten and dairy are the main things.” (P15, FG2, 6 years)

Theme 4: Barriers to making dietary changes

Even when diet was a high priority, it was not always easy to achieve or maintain the desired dietary modifications. Since the majority were working, time to prepare ingredients and cook meals was limited by long days at work.

> “I live on my own as well, so to try and do all those things, and work full time, and get home […]. I'm generally pretty tired by the time I get home at 6:30 anyway. But that kind of impacts my food choices.” (P4, FG1, 2 years)

The participants’ living situations dictated the capacity to strictly adhere to a specific diet. Many did not want to cook two meals at each eating occasion (i.e. one meal for themselves, and one for their partner and/or the rest of the family). Rarely, participants described putting in the effort to cook separate meals.

> “I've got a family, so we can't [afford], for me to have a different diet.” (P31, FG6, 20 years)

It was common for MS symptoms to present as a barrier to sticking to planned dietary modifications. Fatigue, feeling unwell, and having a relapse, typically caused participants to waver from their dietary plans.

> "My fatigue is so high that if I'm at home on my own […] I can't be bothered cooking. […] I don't have the energy to get up and cook." (P13, FG3, 15 years)

Some of the specific diets required special ingredients that were expensive and only available at specialty stores. There was discussion on how managing a specific diet was "hard work" (P10, FG2, 6 years), and required mental and physical effort every day.

> "Suddenly you have this whole obscure list of ingredients [ha], like where do I find this stuff anyway? Um, and then it costs like $22 or something for it, rather than paying, I don't know, $2.99 for cereal you're paying $14 for something different." (P4, FG1, 2 years)

The participants described ways in which they overcame some of the barriers to making dietary changes. Strategies to overcome fatigue included prepping raw ingredients during the day, cooking large batches of food to freeze, and using kitchen appliances such as slow cookers, mandolin slicers, and food processors.

> "I've got a Thermomix, which I love, and it chops. It just does make my coleslaw in like seven seconds and all I have to do is you know just chop it into chunks and chuck it in." (P13, FG3, 15 years)

Assistance from family or MS support workers with shopping, preparing ingredients, and/or cooking reduced the effort and fatigue for some pwMS.

> "I'm so lucky to have MS helpers come […]. A food prep person that comes in once a week, and she makes a huge chicken broth that lasts a week." (P10, FG2, 6 years)

Theme 5: Judging if dietary changes work

The presence or absence of MS symptoms were discussed as ways to judge the impact of dietary modifications. Changes in energy levels, limb strength or dexterity, cognitive clarity ("brain fog" (P2, FG1, 6 years)), skin condition (e.g. pimples, hives, or itchy skin), bladder and bowel functioning, and the presence of migraines were perceived to be the direct

result of dietary modifications. Seeing an improvement in their symptoms motivated participants to continue with the dietary changes.

> “I think [it’s] how we feel, like literally. You know if the fasting's going to help you with your symptoms or feeling um- I really gauge on how I feel.” (P10, FG2, 6 years)

> “I find when I was eating bread and all that crap, that's when I had really bad issues with my bladder and bowel. So that's why I stopped.” (P33, FG6, 4 years)

There was uncertainty about whether fluctuations in energy or mood were a result of dietary modifications, or if they were simply due to MS. Despite this doubt, participants were hesitant to revert back to old dietary habits.

> “I think I did actually feel better, but again I don't know if that's because I was just having a period of time that I felt better, or whether the diet changes made me feel better.” (P4, FG1, 2 years)

> “[My] neurologist said 100% [that] the diet hasn’t had any benefits to my MS at all, uhm, but like I’ve said to my husband, he can’t prove to me that I wouldn’t be worse if I wasn’t eating healthier […] He can’t tell me I wouldn’t be ten times worse if I wasn’t implementing the diet.” (P21, FG4, 3 years)

There was some discussion that objective measures of MS progression, such as lesion activity evident from magnetic resonance imaging scans, gave definitive answers about the effectiveness of dietary modifications. This eliminated the need to make judgement calls based on feelings or symptoms. In those situations, it was not clear how the effects of dietary modification were differentiated from either the natural disease progression or benefits from disease-modifying therapies.

> “My diet significantly changed […]. As a result, I haven’t had an attack, no more new lesions.” (P18, FG3, 6 months)

One participant described experiencing a relapse after making a dietary modification. This was considered to be evidence that diet does influence disease progression.

> “The time that I slipped was when um, I got introduced to coconut oil as a good fat, and I ended up relapsing.” (P11, FG2, 6 years)

Theme 6: Wanting dietary guidelines for MS

Despite being sceptical of the national dietary guidelines and personalising specific diets, participants overwhelmingly wanted to be told what to eat for their MS. While they accepted that guaranteed benefits were unlikely, participants wanted to know what dietary modifications may be beneficial. The desire for clear MS-specific dietary advice contradicted the discussions about personalising diet plans. While participants agreed that they wanted to be told what to eat, the individuality of MS meant that "a one-size fits-all" (P29, FG6, 20 years) answer for diet was unlikely.

> "You just want someone to say-" (P4, FG1, 2 years) "-eat this, or do this, and this will make it, make your life better. […] (P5, FG1, 6 years)
>
> "I think you go, okay, 'dairy-free, gives you these benefits because it affects, I don't know, your gut, or your acidity, or your inflammation, or your fatigue.' […] 'If you go dairy-free then this is the benefits you should feel because we've researched it.'" (P28, FG6, 17 years)
>
> "I don't think you could tell somebody that this is the diet for MS, because we're all so different." (P34, FG6, 6 years)

There was discussion about the desire for MS-specific dietary guidelines: a "pyramid chart to show you what's accurate for MS" (P10, FG2, 6 years). The participants wanted well-researched "baseline" (P30, FG6, 16 years) guidelines, which could be adapted to suit their own personal experiences with diet. The national dietary guidelines were not seen to fit this need.

> "We all understand that whole triangle [food pyramid]. But on that triangle are things like dairy, wheat, pasta, rice. […] Should our food pyramid be substituted with 'okay, instead of eating this, eat this.' This is our food pyramid because science, food science, tells us that for our gut we don't eat the dairy, we don't eat the wheat." (P28, FG6, 17 years)

Participants wanted simple instructions about suitable dietary modifications for MS, including a "list of foods to avoid [and a] list of foods to eat" (P15, FG2, 6 years). Access to evidence-based MS-specific dietary guidelines would provide relief from having to sift through the "minefield" (P15, FG2, 6 years) of information on the internet. MS-specific dietary guidelines would give participants the confidence and motivation to adhere to

dietary changes, since they would be sure that they were meeting their nutritional requirements. Participants agreed that an MS dietary education program would be ideal to learn about MS-specific dietary guidelines, and that it should be facilitated by a credible health professional (nutritionist or dietitian).

Discussion

This qualitative study provides insight into the experiences of pwMS when navigating dietary advice, their attitudes when making dietary decisions, and their needs regarding dietary resources and education. PwMS were confused about where to seek dietary advice. The majority of participants thought that neurologists were not allowed to counsel on specific diets outside of the national dietary guidelines, and were sceptical about the suitability of the guidelines for pwMS. Most pwMS were highly motivated to make dietary modifications, and they wanted MS-specific dietary guidelines.

The SDT is a theory of human motivation, development, and health, focusing on the types of motivators as predictors of personal and well-being outcomes.[37] Why pwMS make dietary modifications - and their varying levels of motivation - can be explained by the central tenants of SDT.[37] There are three fluid types of motivation on a continuous scale. At one end is amotivation, where there is a lack of motivation to change. For some pwMS, not getting MS-specific dietary advice from their neurologist led to greater amotivation. Further along the scale is controlled or external motivation, where behaviour change is shaped by extrinsic factors, such as obligation or coercion. Health professionals could play a role in motivating patients by providing this type of motivation. For pwMS, the perceived physical benefits for disease management (an external motivation) from physical activity has been reported as a positive predictor of physical activity participation.[22] At the other end, the highest form of motivation is autonomous motivation, which occurs when the values of an activity have been integrated into one's own values.[37] This type of motivation has been associated with improved physical and psychological outcomes.[38] The desire to achieve intrinsic goals (e.g. improving health through dietary behaviours, and the accompanying sense of accomplishment) meets the needs for autonomy and competence in pwMS to drive behaviour change.[37] A nutrition education program could provide information and skills that also meet those needs.

Seeking information and maintaining dietary behaviour change were driven by external motivation. In the group discussions, the participants were eager to know what their peers were doing with diet, even if they had not made dietary changes themselves and/or were encouraging each other to consider dietary modifications that seemingly worked for themselves. For some, the fear of worsening symptoms (which causes worry, and disrupts valued activities and everyday routines of pwMS[39]) or relapse due to dietary behaviours was a form of external motivation to continue with their dietary modifications. Similarly, external motivation has been associated with adherence to dietary recommendations in people newly diagnosed with type 2 diabetes.[40] When discussing where to seek dietary advice, the participants in our study were confused and found it difficult to determine what information was credible. They wanted dietary advice from their neurologists (external motivation). These findings are in line with a previous study of people newly diagnosed with MS, who found it difficult to judge the credibility of dietary information, and wanted input from their neurologists.[17]

Most of the participants in our study were highly motivated to make dietary modifications, and their behaviours were autonomously motivated. The participants' goals to manage their symptoms and improve their health were intrinsic, since diet was something they could change to self-manage their disease. This autonomous motivation was driven by the apparent ability to make and adhere to dietary modifications, and the perceived effectiveness of those changes (i.e. improvement in MS symptoms). When autonomous or internalised motivations drive behaviour change, the outcomes are more sustained.[37] Making dietary modifications is a way that pwMS can feel in control of their disease,[17,41] and provides a sense of hope since they are able to take action.[42] Newly-diagnosed pwMS prioritise their health (motivations are autonomously-driven);[41] hence, this is an ideal time to help pwMS build competence and improve their diets to meet the recommendations of the national dietary guidelines.

Focus group discussions revealed that participants wanted to be told what to eat for their MS. MS-specific dietary advice from neurologists (the first MS health professional encountered after diagnosis), appropriate referral to dietitians, and access to an evidence-based nutrition education program could give pwMS the confidence and motivation to make and adhere to dietary modifications. Credible and evidence-based dietary advice

from a neurologist could start to build intrinsic motivation for pwMS and empower them in their decision-making,[43] and provide an additional source of external motivation. An MS nutrition education program could contribute to building self-esteem and autonomy in making dietary decisions, and provide pwMS with the tailored education they seek; as opposed to the generic advice that they report receiving from their health-care providers.[39] Over time, this could transition motivation from external to autonomous, as the drivers for dietary behaviour change become more intrinsic. This could lead to pwMS making healthier choices, since autonomous forms of motivation are central to the adoption and maintenance of healthy diets.[44]

A strength of this study was that we included participants who had MS for varying lengths of time, and the proportion of females compared to males was similar to the sex distribution of the disease. Our study has some limitations. The participants who chose to participate may have been more motivated to make dietary modifications than the typical population of pwMS, resulting in selection bias. People who had low English proficiency, such as culturally and linguistically diverse groups, may not have participated. The views expressed were representative of the participants, but may not be generalizable to the wider MS community.[32] There is the potential for social desirability bias, where participants may have conformed to the general group consensus, instead of expressing their authentic views.

Conclusion

PwMS want to take action in the self-management of their disease, and they are reconsidering their lifestyle choices after diagnosis. Evidence-based MS-specific dietary resources need to be available from dietitians and neurologists. Such resources should highlight the potential benefits from adhering to national dietary guidelines, e.g. avoiding nutrient deficiencies that may exacerbate MS symptoms. Given the suitability of the SDT for explaining how pwMS make dietary decisions, and the different degrees of motivation for dietary change, future dietary change interventions could use the SDT as a framework for design. Our finding that people tend to personalise specific diets promoted for pwMS is informative for future quantitative research: surveys need to provide participants with the opportunity to detail the dietary changes they are adhering to, including any variations to specific diets promoted for pwMS.

Data Availability Statement

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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Chapter 7. Using co-design principles to develop an online nutrition education program for people with multiple sclerosis

Thesis objectives addressed in this chapter:

Objective 4: To develop an evidence-based nutrition education program for people with MS and explore the acceptability and ease of comprehension of one draft module.

The content of this chapter is covered by Publication 5 (under review):

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The version that appears in this thesis is of an article that is currently under peer-review. Phases 1, 2, and 3 in this manuscript are referred to as “stages” in the other sections of this thesis.

The contribution of co-authors, A/Prof Andrea Begley, Ms Justine Purdue, Dr Alison Daly, and A/Prof Lucinda Black are detailed in the author attribution statements in Appendix A: Attribution statements.

See Appendix G for the study consent forms, participant information statements, discussion guides, and demographic questionnaire.

A collaborative approach to designing an online nutrition education program for people with multiple sclerosis

Rebecca D Russell[a], Lucinda J Black[a,b], Justine Purdue[a], Alison Daly[a], and Andrea Begley[a]*

[a]Curtin School of Population Health, Curtin University, Perth, Australia; [b]Curtin Health Innovation Research Institute, Curtin University, Perth, Australia

*corresponding author: Andrea Begley, a.begley@curtin.edu.au +61 8 9266 2773

Implications for Rehabilitation

- Co-designed nutrition education programs can help people achieve high-quality diets in line with recommendations, but very few programs exist for people with MS, and none were co-designed
- One module of a co-designed nutrition program was deemed acceptable and easy to comprehend by people with MS
- Co-design can ensure that the language is appropriate for the target audience, and positive language appeared to improve motivation in people with MS to engage with the online nutrition education program
- Health professionals should collaborate with MS consumers when developing resources, and use positive, empowering language

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Chapter 8. Discussion

This thesis has achieved the aim of using a collaborative approach to develop a nutrition education program for people with multiple sclerosis (pwMS). The thesis used a collaborative, mixed-methods approach to research, design, develop, and test one module of a novel nutrition education program created solely for pwMS. It has filled the knowledge gaps identified in Chapter 1 by achieving the four defined objectives:

1. To identify what nutrition education programs have been implemented for adults with neurological diseases, and the characteristics of those programs
2. To determine the characteristics of effective education programs for pwMS, namely emotional wellness programs
3. To explore the perceptions and experiences of neurologists in giving dietary advice and of pwMS in receiving dietary advice
4. To develop an evidence-based nutrition education program for pwMS and explore the acceptability and ease of comprehension of one draft module

This discussion chapter presents a summary of the main findings from each of the four phases that addressed the objectives, then explores the significance of the findings to the thesis overall, including recommendations for future research to further collaborate with pwMS to continue developing and testing the nutrition education program.

8.1 Nutrition education programs for adults with neurological diseases (phase 1)

A scoping review is presented in Chapter 3 to address objective 1 and answer the research question “What nutrition education programs currently exist for pwMS?” The review mapped out existing nutrition education programs for adults with neurological diseases, including MS, and thoroughly examined the characteristics of each programs[94] to inform the development of a nutrition education program.

Dietary behaviour change is complex and requires support, particularly for pwMS.[95] Hence, it is recommended as best practice to have suitable theories underpinning nutrition education interventions to support dietary change and to guide the constructs of the BCTs used.[12] Of the four MS programs identified in the review, only one in the used an

underlying behaviour change theory (the Health Action Process Approach[96]). Other theories have been widely used in nutrition education programs, such as the Transtheoretical Model, Social Cognitive Theory, the Health Belief Model, and the Theory of Planned Behavior,[12, 97] but they may not be suitable for the MS population, given the complex cognitive and behavioural factors that characterise MS.[98] Therefore, it is necessary to identify a suitable theory, or combined aspects of multiple theories to underpin a nutrition education program for pwMS.

There was little evidence of co-design, with only one MS program demonstrating some indication of co-design in the form of participatory research (an online survey was used to inform the topics covered in the subsequent two-hour, single-session nutrition education program[80]). The variation in program characteristics and poor reporting did not provide a clear insight into suitable program characteristics for a nutrition education program for pwMS. However, I was able to extract the common characteristics (including behaviour change techniques (BCTs)) as potentially useful for informing a nutrition education program dedicated to pwMS.

In the context of this thesis, the review provided insights into common characteristics and potential BCTs, but no insight into a suitable theory to support dietary behaviour change in pwMS. Chapter 3 filled the knowledge gap pertaining to what nutrition education programs exist for pwMS and highlights the scarcity of such programs – and further, none adhere to best practice principles for nutrition education programs. These findings justify the need to first identify characteristics of education programs that are appropriate for the MS population (Chapter 4) and to develop a nutrition education program for pwMS that aligns with best practice principles, including the use of co-design to collaborate with pwMS while developing the program.

8.2 Effective emotional wellness programs for adults with multiple sclerosis (phase 2)

Chapter 4 aimed to answer the research question “What are the characteristics of effective nutrition education programs for pwMS?”; however, due to the scarcity of nutrition education programs for pwMS, a meta-analysis was not possible. This chapter addressed objective 2 through a systematic review and meta-analysis of emotional wellness programs for pwMS[99] and examined the characteristics of effective programs.

This chapter provided insights into some of the potential characteristics for a nutrition education program for pwMS, including BCTs that were used in effective interventions. The findings were consistent with some of the BCTs used in physical activity interventions for pwMS and adults with disabilities[100, 101] and self-management interventions for pwMS[102, 103]; however, these reviews also identified other BCTs, such as barrier identification/problem solving, general instruction, and self-monitoring.[101, 103] Given that the constructs of BCTs are guided by the overarching theoretical framework, the differences in BCTs between the types of interventions further supports the need to identify a suitable theoretical framework to support pwMS in making dietary behaviour changes.

The findings from Chapters 3 and 4 identified the common characteristics and BCTs that may be suitable for the developing nutrition education program for pwMS. The potential characteristics were sessions held in-person, weekly, within a group setting, six-to-eight sessions, and up to nine hours of delivery (i.e., 1-1.5 hours per session). This intervention dosage is consistent with the literature regarding sufficient time for dietary behaviour change.[104]

8.3 The perceived role of neurologists in providing dietary advice to people with multiple sclerosis (phase 3, study 1)

A qualitative study was used to answer the research question “What dietary advice is provided to pwMS by their neurologists and how do neurologists perceive the role of diet in MS?” in Chapter 5. This was one of two qualitative studies that addressed objective 3. This chapter explored the perceptions of neurologists about diet and MS through semi-structured interviews and identified the type of dietary advice that the neurologists provided to their patients with MS.[105]

In the context of this thesis, these interviews provided an understanding of what dietary advice pwMS may receive from their neurologist, including the rationale underpinning such advice. Overall, there was very little discussion about diet or nutrition, and the advice that the neurologists interviewed gave to their patients with MS was not always evidence based. This deviation from evidence-based advice is not unusual when the preferences of the patient either conflicts with the evidence, when the medical literature does not provide a clear consensus, and/or when physicians focus on facilitating the autonomy of their patients.[106, 107] This chapter highlights the need for an easily accessible and evidence-based source of nutrition information that neurologists could refer their patients to, such as

a nutrition education program. This would enable neurologists to provide consistent advice about dietary changes that could benefit pwMS. This is vital for pwMS, since neurologists are perceived as a reliable and preferred source of MS health information.[75, 76, 82]

In general, the neurologists very rarely acknowledged any potential benefits of dietary changes on MS symptoms or diet-related co-morbidities, despite pwMS experimenting with diet to manage their symptoms.[74, 80] The role of co-morbidities and disability progression, quality of life, and hospital readmission in pwMS[41-43, 45] (as presented in Chapter 1) was also rarely considered in the dietary advice that they provided. The lack of focus on the interactions between diet, symptoms, and co-morbidities that are important for pwMS highlighted an interconnected area that could be addressed in the nutrition education program. This would address the gap pertaining to services offered by neurologists for pwMS.

The neurologists expressed that they had little influence, time, and knowledge when it came to dietary recommendations or advice for their patients with MS. Instead of referring their patients to dietitians, they gave non-specific healthy eating advice, but none referred to the Australian Dietary Guidelines as a guide for healthy eating. This aligns with a 2022 study of neurology residents treating patients with stroke, where a lack of perceived influence, time, and training were cited as barriers to providing dietary advice, and the vast majority did not refer their patients to dietitians.[108] It is possible that the vague advice and lack of emphasis for dietitians related to the role with which they viewed diet and the inconsistent evidence for diet in reducing MS disease progression. However, it is unclear why the neurologists did not refer patients to dietitians as standard practice and as such, warrants future investigation. Moreover, the provision of generic advice from health providers, as opposed to personally tailored advice, has been widely reported by pwMS[109]. In the absence of neurologists providing referrals to dietitians as standard practice, this chapter highlights the importance of including information in the nutrition education program for pwMS regarding how to find a dietitian.

8.4 How people with multiple sclerosis navigate dietary advice (phase 3, study 2)

Chapter 6 details the second qualitative study that addressed objective 3 and answered the research question “How do pwMS respond to the nutrition information that they receive?”. The study explored the experiences of pwMS when navigating dietary advice,

including their attitudes when making dietary decisions, and their nutrition education needs using by focus groups.[110]

In the context of this PhD project, the overarching themes from Chapter 6 were incorporated into the developing nutrition education program, namely, confusion about where to seek dietary advice (and how to judge the credibility of information), wanting dietary guidelines that are tailored for MS, scepticism towards national dietary guidelines, and barriers to making dietary changes (such as fatigue and other symptoms, time, living with someone else and not wanting to prepare separate meals, as examples). Participants shared ways that they overcome these barriers; exemplars which were also incorporated into the nutrition education program. The findings from this chapter highlights the importance of addressing the suitability of the Australian Dietary Guidelines with targeted messages (i.e., specifying why they are important for pwMS to improve diet quality and how various dietary patterns that pwMS may experiment to evaluate potential benefits for their symptoms *can* fit within the guidelines if approached carefully, e.g., vegetarian and vegan diets). These findings are supported by a 2021 study exploring the desired resources for supporting dietary changes in pwMS.[95] The study participants reported the need for MS-specific dietary information, including information about how diet may impact their symptoms, as well as dietary guidelines for MS that were provided to them by a reliable source.[95]

Recent literature has also noted the importance of addressing the underlying challenges associated with dietary behaviour change in pwMS by using behavioural supports such as self-monitoring, tangible resources including recipes and food lists, and motivation.[95] This is supported by the information provided by participants who took part in this study. Most of the pwMS in the focus groups were motivated to make dietary changes, and the central tenants of the self-determination theory explained the varying degrees of motivation.[111] This theory has also been used to understand physical activity behaviours and adherence to recommendations in pwMS[112, 113] and was used as the underlying theoretical basis for the developing nutrition education program in this PhD project. Given the psychological, cognitive, and physical complexities surrounding the initiation and maintenance of behaviour change for pwMS, the CEOS theory was also used as a theoretical basis to guide the development of the nutrition education program.[98]

Building on the potential characteristics and BCTs identified in Chapters 3 and 4, the qualitative findings from Chapters 5 and 6 were used to develop a framework for a nutrition education program. The program framework outlined key topics, overarching messages,

and barriers to dietary change. The framework included elements that could potentially influence external and internal motivation based on the self-determination theory,[111] such as a printed activity book with weekly activities based on the content (external motivator), and goal setting and re-evaluating activities to facilitate internal/autonomous motivation.

8.5 Using co-design principles to develop an online nutrition education program for people with multiple sclerosis (phase 4)

The final research chapter of this thesis, Chapter 7, addressed objective 4. This mixed-methods chapter ties together the findings from the previous chapters used to develop the framework for the draft nutrition education program. The chapter details the collaborative process involving pwMS and MS health professionals to further develop the program framework to produce a prototype nutrition program and test one module of the online program. The use of mixed-methods for this chapter enabled me to gain a more complete perspective[87] into the nutrition education needs of pwMS by facilitating accessibility for range of participants to share their preferences via an online survey, and in-depth exploration of key factors that influenced acceptability of an online module using cognitive interviews.

PwMS are interested in online nutrition resources,[95] and this chapter reinforces the importance of using co-design principles to ensure the nutrition education program meets their needs, considers their beliefs and values, and that the theories, BCTs, visuals, language, and experiential activities are appropriate.[12, 16, 17] By meeting the needs of the end-users, and therefore creating interventions that are accepted, co-design can prevent research funds and resources from being wasted.[15] While published evidence of co-designed programs and research partnership involving pwMS is limited,[114] a co-designed web-based mindfulness program was reported as relatable and acceptable by pwMS[115] and online resources that were developed in collaboration with pwMS were rated as useful, aesthetically pleasing and easy to understand by pwMS.[116, 117] The use of co-design in nutrition interventions has increased in recent years[118] and co-designed nutrition interventions show the most promise for positive health behaviour change.[14] However, a 2022 scoping review has concluded that there is a need to ensure the lived experiences of consumers are incorporated to produce interventions that meet their needs, as opposed to ‘tokenistic’ engagement.[118] Therefore, future nutrition education programs should be developed using co-design to ensure that the overall program is acceptable to end-users.

The cognitive interviews at the full prototype stage of one module provided vital information that could impact the acceptability of the final program, and warrants consideration for future online education programs for pwMS. The language and visual design appeared to influence engagement with the online module and may ultimately influence the motivation of pwMS to make evidence-based, healthy dietary changes. The preference for a positive tone was not surprising, given that pwMS make dietary changes as a way to feel in control of their disease,[74] and the expert recommendations state that nutrition messages should be practical, motivating, and positive.[119, 120] Previous research has also demonstrated the importance of involving pwMS to produce online resources to ensure the wording and language used is tailored and appropriate for pwMS.[121] Given the reported effectiveness of online interventions to support lifestyle (diet and physical activity) behaviour change in non-clinical adults[122] and dietary change in adults with type 2 diabetes,[123] this chapter contributes important insights for future online nutrition education programs for pwMS. Through ongoing engagement with pwMS throughout this study, the nutrition education program modules were driven by, and developed with, those who will potentially gain the most benefit, due to the recognised need to use the lens of pwMS throughout all stages of the program development.

8.6 Significance

This thesis has made a significant new contribution to new knowledge by filling the gaps in the literature defined in Chapter 1, specifically: 1) mapping out what nutrition education programs exist for pwMS and the characteristics of those programs, including evidence of consumer involvement in the development; 2) defining the characteristics of effective emotional wellness programs for pwMS and the BCTs used; 3) understanding how neurologists perceive the role of diet in MS and what dietary advice they give to their patients with MS; and 4) understanding how pwMS respond to the nutrition information they receive, including their attitudes when making dietary decisions and their nutrition education needs. Filling these knowledge gaps uncovered the need to develop an online nutrition education program specifically for pwMS. This thesis also identified and justified the self-determination theory as suitable for underpinning nutrition education programs for pwMS, as well as a range of BCTs that may be useful to support dietary behaviour change in this population.

Given the abundance of conflicting information about diet on the internet, it was evident from the findings in this thesis that pwMS wanted and needed an online nutrition education program that included guidance on how to interpret what is evidence-based advice, and how to apply such dietary advice to their food preferences and living situations. Online programs are positively received by pwMS and provide greater accessibility than in-person programs, enabling pwMS to overcome physical and geographic barriers to facilitate participation.[124] Furthermore, these interventions can be more cost effective compared to usual care, educational information controls, and face-to-face traditional interventions.[125] The program prototype developed in this thesis project was designed to not only meet the expectations of pwMS concerning the information that they wanted to learn about, including how diet may affect symptoms of MS, but also with the potential to alleviate the uncertainty surrounding the role of the neurologist in providing dietary advice. Neurologists could refer their patients to such an evidence-based resource to learn about appropriate dietary modifications, which would further meet the expectations of pwMS with their neurologists providing dietary guidance and alleviate the barriers identified by neurologists with regards to lack of time and expertise.

This thesis also highlighted the significance of using principles of co-design to develop the nutrition education program prototype. The collaborative processes involving pwMS and MS health professionals, supported by the project stakeholder advisory group, ensured that the values, needs, language preferences, and visual design preferences of pwMS, and their potential barriers to dietary change, were considered. These elements were important to identify and consider, as they appeared to contribute to the overall acceptability and willingness to engage with the draft module. This thesis has demonstrated that pwMS want to feel empowered to make healthy dietary changes, and that they are motivated by information about dietary changes that could improve their health and help to manage some of their symptoms, as opposed to highlighting the lack of evidence to support one specific diet for pwMS. Overall, the prototype nutrition education program could give pwMS the skills and confidence that they need to make beneficial dietary changes with sufficient behaviour change support, and potentially empower them to help manage their symptoms and provide a sense of control over their disease.

8.7 Strengths and limitations

The major strength of this thesis that informed the development of a prototype nutrition education program for pwMS was the systematic collection of data that included systematic literature reviews, qualitative interviews and focus groups, and a quantitative survey. The use of qualitative methods enabled me to understand the participants' subjective experiences and to explore the topics more deeply as a result, compared to the use of quantitative methods alone.[126] The use co-design principles throughout this thesis enabled the insights of pwMS to direct the content, characteristics, design, and language used in the prototype program. I also used several recognised techniques to ensure rigour in the qualitative studies in this thesis.[85]

The limitations of the studies in this thesis are subject to the common limitations of nutrition-based studies. Firstly, self-selection bias, meaning that neurologists and pwMS who are interested in diet were more likely to participate in the research. Secondly, social desirability bias, or, put simply, where participants may have answered based on what they assumed that I wanted to hear or what was considered to be socially acceptable, as opposed to their actual opinions. However, during all three qualitative studies in this thesis, I emphasised that all answers were the participants' own experiences and opinions and that there was no right or wrong answer. All three studies involved participants who indicated that they were either indifferent or unsure about how they felt about diet. Furthermore, the qualitative studies (interviews with neurologists and focus groups and interviews with pwMS) were all conducted in Western Australia (WA); therefore, it is possible that the findings may not be generalisable outside of WA and/or Australia. Lastly, while I attempted to interview a range of neurologists working across both the public and private sectors and in both the metropolitan and regional areas of WA, the sample of neurologists may not be representative of all 40-50 neurologists working in WA at the time of the interviews.

8.8 Implications for future research

The overall aim of this thesis was to use a collaborative approach to develop a nutrition education program for pwMS. The next stage of this research project is to amend the draft program prototype based on the findings from the cognitive interviews (phase 4) and test the complete program. With the support of a 2021-2022 MS Australia Incubator Grant (#21-1-072; Russell RD (CIA), Begley A, Black LJ, and Daly A), our research team has

made the necessary amendments to the draft program prototype and produced a full program prototype with program logic model ready for feasibility testing (Australian New Zealand Clinical Trials Registry ACTRN12622000276752). The feasibility outcomes for the trial have been guided by the United States National Cancer Institute framework for feasibility studies.[127] These outcomes are demand (participant recruitment: if the target of 75 participants is recruited within six weeks); practicality (proportion of participants that complete each module, overall participant retention, and completion of baseline and post-program questionnaires); and acceptability (assessed using the interest/enjoyment and value/usefulness subscales of the Intrinsic Motivation Inventory[128]).

Future research should involve further development of the nutrition education program based on findings derived from this feasibility study. Researchers should continue to use principles of co-design by involving pwMS to identify and prioritise changes from the feasibility study outcomes and exit surveys from pwMS who completed the program and pwMS who began the program but did not complete it. This would support the identification of potential barriers to participation, which could be addressed in the next version of the program. Future research should also involve a large randomised controlled trial to evaluate efficacy regarding dietary behaviour change and to examine the characteristics of effective nutrition education programs for pwMS, including the BCTs.

This thesis explored the perspectives of neurologists and what dietary advice they give to their patients with MS. It is not known if other MS health professionals provide dietary advice to their patients and/or clients with MS, and if so, what the nature of that advice is. Future research would benefit from exploring the attitudes regarding diet of other MS health professionals, such as MS nurses and counsellors, and to determine if they give dietary advice to their patients and/or clients with MS. Additionally, future research should explore the perceived need for nutrition education resources or nutrition education programs that are targeted to MS health professionals.

8.9 Conclusion

This PhD thesis has produced fundamental knowledge regarding the need and characteristics of a nutrition education program for pwMS in Australia and reinforces the importance of using co-design for end-user acceptability. Nutrition education programs can support pwMS in making healthy dietary changes that could reduce their risk of co-morbidities and alleviate some of their MS symptoms. A nutrition education program that

meets the needs and expectations of pwMS could alleviate the uncertainty of neurologists in this space by giving them an evidence-based resource to direct their patients. However, very few nutrition education programs exist for pwMS, and those that do exist do not align with the best practice principles nor do they have consistent characteristics. Evidence of effective program characteristics is lacking. The publications in this thesis have identified content, language, design, and format considerations of an online nutrition education program that may ultimately improve the motivation of pwMS to make evidence-based, healthy dietary changes. This thesis has also identified a theoretical framework and BCTs that may be useful for supporting dietary behaviour change among pwMS and should be considered in the development of future nutrition education programs. One module of an online nutrition education program that was developed in collaboration with pwMS and MS health professionals was deemed acceptable and easy to comprehend by pwMS. Future nutrition education programs should be developed using co-design principles to ensure that the program is acceptable, engaging, and sufficiently supports pwMS to make healthy dietary changes.

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Appendix A: Attribution statements

Nutrition education programs for adults with neurological diseases are lacking: a scoping review

	Conception and Design	Acquisition of Data and Method	Data Conditioning and Organisation	Analysis and Statistical Method	Interpretation and Discussion	Confirming interpretation and discussion	Final Approval
Co-author 1 Mrs Rebecca Russell	X	X	X	X	X		X
I acknowledge that these represent my contribution to the above research output Signed: [signature redacted] Date: 25th August 2022							
Co-author 2 A/Prof Lucinda Black	X					X	X
I acknowledge that these represent my contribution to the above research output Signed: [signature redacted] Date: 19th September 2022							
Co-author 3 A/Prof Andrea Begley	X			X BCT coding		X	X
I acknowledge that these represent my contribution to the above research output Signed: [signature redacted] Date: 19th September 2022							

BCT, behaviour change technique

The effectiveness of emotional wellness programs on mental health outcomes for adults with multiple sclerosis: a systematic review and meta-analysis

	Conception and Design	Acquisition of Data and Method	Data Conditioning and Organisation	Analysis and Statistical Method	Interpretation and Discussion	Confirming interpretation and discussion	Final Approval
Co-author 1 Mrs Rebecca Russell	X	X	X	X	X		X
I acknowledge that these represent my contribution to the above research output Signed: [signature redacted] Date: 25th August 2022							
Co-author 2 A/Prof Lucinda Black	X					X	X
I acknowledge that these represent my contribution to the above research output Signed: [signature redacted] Date: 19th September 2022							
Co-author 3 Dr Minh Pham				X Statistical support			X
I acknowledge that these represent my contribution to the above research output Signed: [signature redacted] Date: 25th August 2022							
Co-author 4 A/Prof Andrea Begley	X			X BCT coding		X	X
I acknowledge that these represent my contribution to the above research output Signed: [signature redacted] Date: 19th September 2022							

BCT, behaviour change technique

The unresolved role of the neurologist in providing dietary advice to people with multiple sclerosis

	Conception and Design	Recruitment and Data Collection	Data Management	Qualitative Analysis	Interpretation and Discussion	Confirming interpretation and discussion	Final Approval
Co-author 1 Mrs Rebecca Russell	X	X	X	X	X		X
I acknowledge that these represent my contribution to the above research output Signed: [signature redacted] Date: 25th August 2022							
Co-author 2 A/Prof Lucinda Black	X					X	X
I acknowledge that these represent my contribution to the above research output Signed: [signature redacted] Date: 19th September 2022							
Co-author 3 A/Prof Andrea Begley	X			X Confirmed qualitative themes		X	X
I acknowledge that these represent my contribution to the above research output Signed: [signature redacted] Date: 19th September 2022							

Navigating dietary advice for multiple sclerosis

	Conception and Design	Recruitment and Data Collection	Data Management	Qualitative Analysis	Interpretation and Discussion	Confirming interpretation and discussion	Final Approval
Co-author 1 Mrs Rebecca Russell	X	X	X	X	X		X
I acknowledge that these represent my contribution to the above research output Signed: [signature redacted] Date: 25th August 2022							
Co-author 2 A/Prof Lucinda Black	X	X Co-facilitated focus groups				X	X
I acknowledge that these represent my contribution to the above research output Signed: [signature redacted] Date: 19th September 2022							
Co-author 3 A/Prof Andrea Begley	X	X Co-facilitated focus groups		X Confirmed qualitative themes		X	X
I acknowledge that these represent my contribution to the above research output Signed: [signature redacted] Date: 19th September 2022							

Using co-design principles to develop an online nutrition education program for people with multiple sclerosis

	Conception and Design	Recruitment and Data Collection	Data Management	Data Analysis	Interpretation and Discussion	Confirming interpretation and discussion	Final Approval
Co-author 1 Mrs Rebecca Russell	X	X	X	X	X		X
I acknowledge that these represent my contribution to the above research output Signed: [signature redacted] Date: 25th August 2022							
Co-author 2 A/Prof Lucinda Black	X					X	X
I acknowledge that these represent my contribution to the above research output Signed: [signature redacted] Date: 19th September 2022							
Co-author 3 Ms Justine Purdue	X Phase 1 only	X Phase 1 only	X Phase 1 only	X Phase 1 only			X
I acknowledge that these represent my contribution to the above research output Signed: [signature redacted] Date: 19th September 2022							
Co-author 4 Dr Alison Daly				X Statistical support			X
I acknowledge that these represent my contribution to the above research output Signed: [signature redacted] Date: 27th September 2022							
Co-author 5 A/Prof Andrea Begley	X			X Confirmed qualitative themes		X	X
I acknowledge that these represent my contribution to the above research output Signed: [signature redacted] Date: 19th September 2022							

Phase 1: quantitative survey

Appendix B: Publications and presentations not central to this thesis

The following publications, presentations, awards, and prizes were attributed to work that was related, but not central, to this thesis.

Peer-reviewed publications (100% Q1)

1. **Russell RD**, Langer-Gould A, Gonzales EG, Smith JB, Brennan V, Pereira G, Lucas RM, Begley A, Black LJ. (2019). Obesity, dieting, and multiple sclerosis. *Multiple Sclerosis and Related Disorders* doi:10.1016/j.msard.2019.101889.(1 citation, Scopus)

Conference presentations (oral communication)

1. **Russell RD**, Langer-Gould A, Gonzales EG, Smith JB, Brennan V, Pereira G, Lucas RM, Begley A, Black LJ. (2019). Asian Congress of Nutrition; Denpasar, Indonesia. “A wide variety of different diets are adopted after symptom onset for multiple sclerosis”

B.1 Published paper

The content of this appendix is covered by:

Russell, R. D., Langer-Gould, A., Gonzales, E. G., Smith, J. B., Brennan, V., Pereira, G., Lucas, R. M., Begley, A., & Black, L. J. (2020). Obesity, dieting, and multiple sclerosis. Multiple Sclerosis and Related Disorders, 39, 101889. https://doi.org/10.1016/j.msard.2019.101889

The contribution of all co-authors is detailed in the author attribution statement in Appendix B.2.

The version that appears in this thesis appendix is of an article that has been through peer review with *Multiple Sclerosis and Related Disorders* but has not been through the copyediting process.

Obesity, dieting, and multiple sclerosis

R. D. Russell[1], A. Langer-Gould[2], E. G. Gonzales[3], J. B. Smith[3], V. Brennan[1], G. Pereira[1], R. M. Lucas[4,5], A. Begley[1], L. J. Black[1]

Affiliations:

[1] School of Public Health, Curtin University, Perth, Australia
[2] Neurology Department, Los Angeles Medical Center, Southern California Permanente Medical Group/Kaiser Permanente, Los Angeles, USA
[3] Department of Research and Evaluation, Kaiser Permanente, Pasadena, USA
[4] National Centre for Epidemiology and Population Health, Research School of Population Health, Australian National University, Canberra, Australia
[5] Centre for Ophthalmology and Visual Science, University of Western Australia, Perth, Australia

Corresponding Author:

Lucinda J Black
School of Public Health, Kent St, Bentley WA 6102
Lucinda.black@curtin.edu.au
+61 8 9266 2523

Highlights

- Thirty-seven percent of participants were obese
- People with multiple sclerosis were no more likely to adopt a diet than controls
- Being obese, younger, female, or non-Hispanic were associated with dieting

Lay summary

We investigated obesity and dieting in a study comparing 470 people with multiple sclerosis (MS) and 519 people without MS in Southern California. Despite the risks associated with overweight and obesity in MS, people with MS were no more likely to adopt a diet for nutrition or weight loss purposes than people without MS. Those who were obese, younger, female, or non-Hispanic were more likely to adopt a diet, with Weight Watchers being the most frequently reported diet. Nutrition education for people with MS is needed to promote healthy weight loss in those who are overweight or obese.

Abstract

Background

Obesity is common in the United States and is associated with a higher risk of relapse and comorbidities, and increased disease progression, in people with MS.

Methods

We examined the prevalence of overweight and obesity in the MS Sunshine Study, a matched case-control study of multiple sclerosis in Southern California (470 cases, 519 controls). We reported the proportion of participants who adopted a specific diet for nutrition or weight loss purposes, and identified independent predictors of dieting.

Results

In the total population, 32% and 37% were overweight and obese, respectively. Case participants were no more likely to adopt a specific diet for nutrition or weight loss purposes than control participants (10% and 11%, respectively). Being obese, younger, female or non-Hispanic were independently associated with dieting.

Conclusion

Despite the evidence that obesity can worsen MS prognosis, and the high prevalence of overweight/obesity, case participants were no more likely to adopt a specific diet than control participants. Improved nutrition education may help people with MS make healthy dietary changes for nutrition or weight loss purposes.

Keywords

Dietary behavior; dietary changes; MS Sunshine Study; multiple sclerosis; nutrition

1.0 Introduction

In the United States (US), 40% of adults are obese, a trend that is increasing (Hales, Carroll, Fryar, & Ogden, 2017). Overweight/obesity in people with MS (pwMS) has been associated with greater neuroinflammation (Stampanoni Bassi et al., 2019), relapse risk and disability progression (Tettey et al., 2017). There is little research exploring the diets adopted by pwMS for nutrition or weight loss purposes. Using data from the MS Sunshine Study, a multi-ethnic matched case-control study in Southern California examining risk factors for MS (Langer-Gould et al., 2018), we aimed to describe the number and proportion of pwMS adopting specific diets after MS symptom onset, and to identify predictors of adopting a specific diet. We hypothesized that the onset of MS symptoms would increase motivation among overweight or obese individuals to change their diet.

2.0 Material and Methods

Participants of the 2011-2015 MS Sunshine Study were recruited from the Kaiser Permanente Southern California (KPSC) database of >4 million members; detailed methods are described elsewhere (Langer-Gould et al., 2018). In brief, adult members (≥18 years) diagnosed with MS or clinically isolated syndrome within the past 18 months, or those with symptom onset within the past three years, were eligible. Control participants from the KPSC population were matched on age, sex, race/ethnicity, and home KPSC facility (a surrogate measure for socioeconomic status). After written informed consent was obtained, data were collected from structured in-person interviews (race/ethnicity, education), self-administered questionnaire (diets), and the complete electronic health record (BMI at date of symptom onset). The study was conducted in accordance with the Declaration of Helsinki. The protocol was approved by the KPSC Institutional Review Board (IRB 5962).

Body mass index (BMI) was categorised as: normal/underweight (<25 kg/m^2); overweight (25-<30 kg/m^2); obese class I (30-<35 kg/m^2), or obese class II (≥35 kg/m^2). Participants reported start and end dates of specific diets they followed for “nutrition or weight loss purposes”, selecting from nine predefined diets (Paleo, South Beach, Perricone, Jenny Craig, Weight Watchers, 17 Day Diet, Jillian Michael’s, The Mommy Diet, Nutrisystem), and an open text field for “Other”. Participants could select multiple options.

Control participants were considered to have adopted a diet if it occurred between date of symptom onset and interview date of their matched case. Analyses were conducted for the three major racial/ethnic groups (whites, blacks and Hispanics). Predictors of adopting a specific diet (case/control status, sex, age at symptom onset, education, race, BMI category, smoking history) were investigated using logistic regression models (unadjusted and adjusted). We tested for an interaction between case/control status and BMI using an interaction term in the adjusted model. Data were analysed using Stata Software version 14 (StataCorp, College Station, TX, USA). Statistical significance was defined as $P<0.05$.

3.0 Results

Of the 1193 white, black, and Hispanic participants, 989 (83%) had complete data on diets and potential predictors. BMI distribution was similar for cases and controls, with a median of 28 kg/m^2 (interquartile range (IQR) 9 kg/m^2). Most participants were overweight or obese (Table 1), and the prevalence was similar for cases and controls (overweight 31% and 33%; obese 36% and 38%, respectively). Cases and controls had a similar education level (58% and 53%, respectively, had not completed college). The median time from symptom onset to questionnaire completion was 278 months (IQR, 30 months).

Table 1 Participant characteristics at index date[1]

	Cases (*n*=470)	Controls (*n*=519)
Sex, % *(n)*		
Male	26.8% *(126)*	27.6% *(143)*
Female	73.2% *(344)*	72.5% *(376)*
Age, y, mean (SD)	37.5 (12.6)	37.0 (12.7)
Race/ethnicity, % *(n)*		
White	48.1% *(226)*	45.3% *(235)*
Black	21.5% *(101)*	21.0% *(109)*
Hispanic	30.4% *(143)*	33.7% *(175)*
BMI (kg/m^2), median (IQR)	28.1 (24.0-33.0)	27.9 (24.1-33.5)
BMI category (kg/m^2), % *(n)*		
Normal/underweight (<25)	32.6% *(153)*	29.1% *(151)*
Overweight (25-<30)	31.1% *(146)*	33.0% *(171)*
Obese class I (30-<35)	17.9% *(84)*	17.2% *(89)*
Obese class II (≥35)	18.5% *(87)*	20.8% *(108)*
Education, % *(n)*		
Some college or less	57.7% *(271)*	53.4% *(277)*
College or graduate school	42.3% *(199)*	46.6% *(242)*
Smoking history (ever smoked), % *(n)*		
No	66.8% *(314)*	74.0% *(384)*
Yes	33.2% *(156)*	26.0% *(135)*

[1]Date of MS symptom onset (or matched time frame for controls)

IQR, interquartile range; SD, standard deviation

A total of 10% (*n*=46) of case participants reported adopting a specific diet for nutrition or weight loss purposes after symptom onset, while 11% (*n*=56) of controls did so within the same time frame. There was no independent association between MS status and adopting a specific diet (Table 2). Being overweight/obese, female or younger were independently associated with significantly increased odds of adopting a specific diet. Hispanics were 46% and blacks 44% less likely to adopt a specific diet compared to whites, even after controlling for BMI, sex, age, education and smoking, although this finding did not reach statistical significance in blacks. There was no statistically significant interaction between MS status and BMI category (*P*>0.05).

Table 2 Unadjusted and adjusted logistic regression models showing participant characteristics and odds of adopting a specific diet after MS symptom onset (*n*=989; cases, *n*=470; controls, *n*=519)

	Model 1: unadjusted		Model 1: adjusted	
	OR (95% CI)	***P***	**aOR (95% CI)[1]**	***P***
Age at symptom onset (years)[2]	0.97 (0.95, 0.99)	<0.001	0.96 (0.94, 0.98)	<0.001
MS status				
Control	Reference		Reference	
Case	0.90 (0.59, 1.35)	0.605	0.94 (0.61, 1.44)	0.759
Sex				
Male	Reference		Reference	
Female	4.29 (2.13, 8.62)	<0.001	4.48 (2.20, 9.12)	<0.001
Race/ethnicity				
White	Reference		Reference	
Black	0.98 (0.58, 1.68)	0.952	0.66 (0.38, 1.16)	0.150
Hispanic	0.91 (0.57, 1.46)	0.691	0.54 (0.31, 0.92)	0.023
BMI category (kg/m^2)				
Normal/underweight (<25)	Reference		Reference	
Overweight (25-<30)	1.23 (0.66, 2.29)	0.518	1.71 (0.90, 3.25)	0.103
Obese class I (30-<35)	2.65 (1.42, 4.95)	0.002	3.76 (1.95, 7.23)	<0.001
Obese class II (≥35)	3.06 (1.68, 5.55)	<0.001	3.93 (2.11, 7.35)	<0.001
Education				
Some college or less	Reference		Reference	
College or graduate school	1.07 (0.71, 1.61)	0.750	1.12 (0.72, 1.75)	0.604
Smoking history (ever smoked)				
No	Reference		Reference	
Yes	0.72 (0.44, 1.15)	0.170	0.80 (0.48, 1.35)	0.400

OR: odds ratio; aOR: adjusted odds ratio; CI: confidence interval

[1]All variables included in a single model

[2]Odds ratio is per one-year increase in age

The proportion of participants within each BMI category who adopted a specific diet was similar for cases and controls (Figure 1). The most frequently reported diet was Weight Watchers (cases, *n*=16; controls *n*=18).

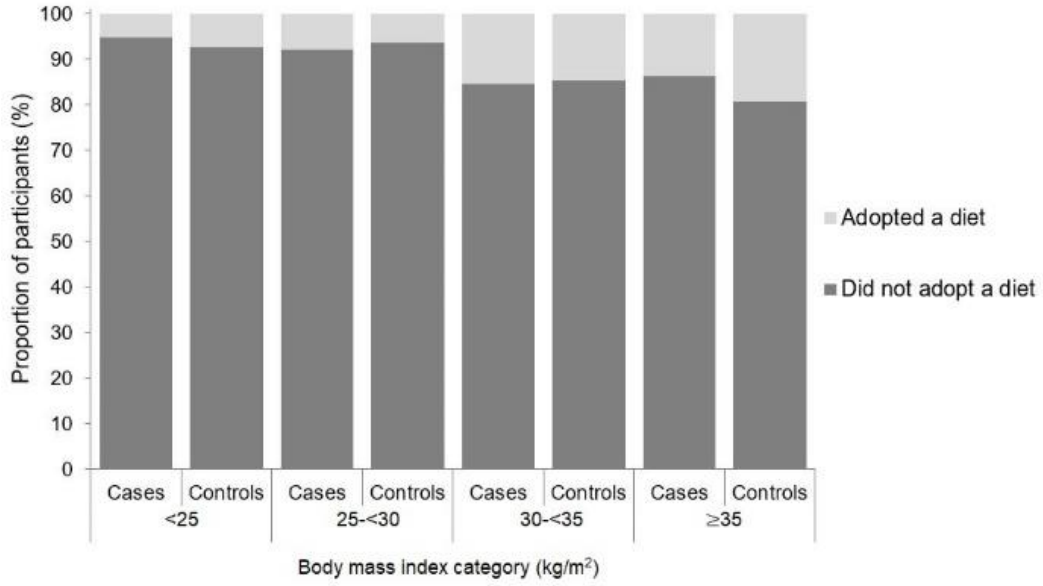

Figure 1 Proportion of participants who did and did not adopt a specific diet for nutrition or weight loss purposes since MS symptom onset (or matched time frame for controls)

4.0 Discussion

In the MS Sunshine Study, case participants with recent onset of MS symptoms or diagnosis of MS were no more likely to adopt a specific diet for weight loss or nutrition purposes than control participants, despite a high proportion being overweight or obese. Achieving and maintaining a healthy weight is particularly important for pwMS, since obesity is associated with a higher risk of relapse (Tettey et al., 2017), comorbidities (e.g. diabetes, hypertension, depression), and greater disease progression of MS (Marrie, 2017).

The proportion of pwMS making dietary modifications in other studies is higher than in our study, ranging from 17% (Brenton & Goldman, 2016) to approximately 40% (Fitzgerald et al., 2018; Riemann-Lorenz et al., 2016; Russell et al., 2018). This is likely due to differences in study population, design and dietary assessment. Previous studies included greater representation of white participants (>90%) (Fitzgerald et al., 2018; Russell et al., 2018) and prevalent cases (mean disease duration >7 years) (Brenton & Goldman, 2016; Riemann-Lorenz et al., 2016), and inquired about any dietary change (Russell et al., 2018), whereas we focused on specific diets for weight loss or nutrition purposes.

Our finding that females were more likely to adopt a weight loss diet than males is consistent with the general population (Martin, Herrick, Sarafrazi, & Ogden, 2018) and a large survey of prevalent MS cases (*n*=6989) (Fitzgerald et al., 2018). Likewise, our findings that low-calorie and low-carbohydrate diets were the most popular weight loss diets are consistent with a large survey of pwMS (Fitzgerald et al., 2018).

A limitation of our study is that we did not capture other dietary weight loss efforts participants may have been making after symptom onset or diagnosis, such as reducing the consumption of unhealthy foods. We also cannot exclude the possibility that, with longer duration of follow-up, case participants may have been more likely to engage in specific diets than control participants. Furthermore, we did not examine the relationship between the likelihood of adopting a diet and other variables that may influence dieting, such as MS phenotype, disability status, physical activity, alcohol consumption, comorbidities, and family history of obesity.

5.0 Conclusions

Our findings support the need for MS-focussed nutrition education, with an emphasis on healthy approaches to weight loss for those who are overweight or obese. Only one study has reported the development and feasibility of a dietary education program for pwMS (Riemann-Lorenz et al., 2016), but it had overall low participant satisfaction and was a largely white study population. Furthermore, there is no evidence regarding the effective elements of dietary education to elicit behavior change or weight loss in pwMS. Such programs, particularly ones targeted to ethnic diversity and males, should be developed and evaluated in randomized controlled trials.

Acknowledgements

We thank the participants of the MS Sunshine Study.

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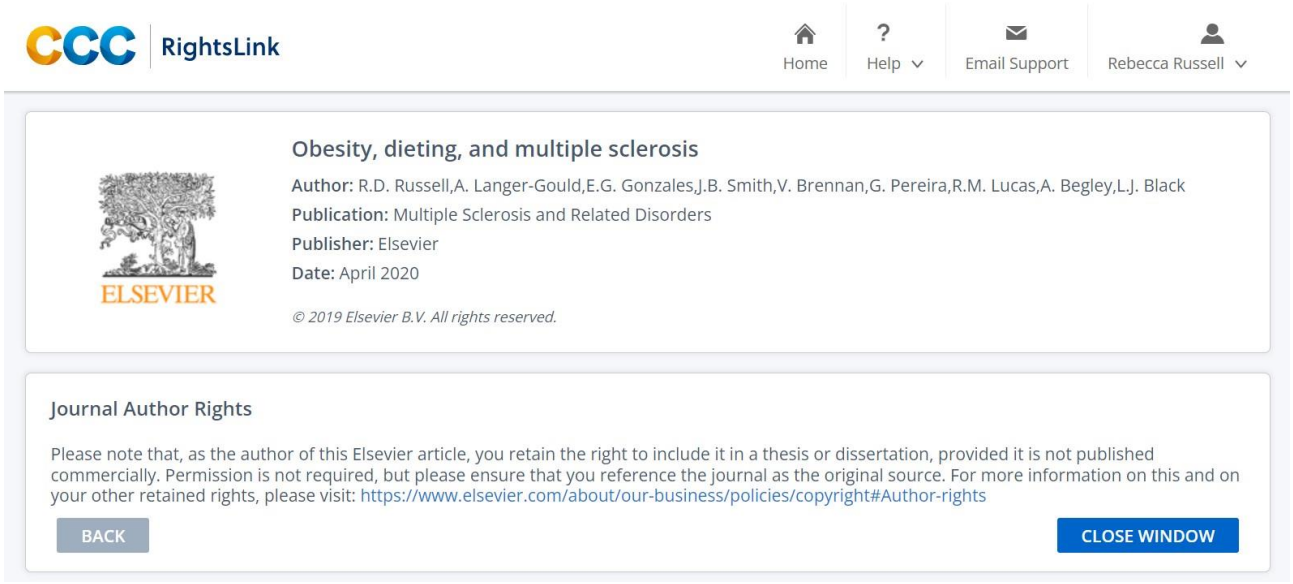
B.2 Attribution statement

Obesity, dieting, and multiple sclerosis

	Conception and Design	Recruitment and Data Collection	Data Management	Data Analysis	Interpretation and Discussion	Final Approval
Co-author 1 Mrs Rebecca Russell				X	X	X
I acknowledge that these represent my contribution to the above research output Signed: [redacted] Date: 25th August 2022						
Co-author 2 Dr Annette Langer-Gould	X		X		X	X
I acknowledge that these represent my contribution to the above research output Signed: [redacted] Date:						
Co-author 3 Ms Edlin Gonzales		X			X	X
I acknowledge that these represent my contribution to the above research output Signed: [redacted] Date:						
Co-author 4 Ms Jessica Smith			X		X	X
I acknowledge that these represent my contribution to the above research output Signed: [redacted] Date:						
Co-author 5 Ms Vanessa Brennan				X		X
I acknowledge that these represent my contribution to the above research output Signe [redacted] Date:						
Co-author 6 Prof Gavin Pereira				X Statistical support		X
I acknowledge that these represent my contribution to the above research output Signe [redacted] Date: 26th August 2022						
Co-author 7 Prof Robyn Lucas						X
I acknowledge that these represent my contribution to the above research output Signe [redacted] Date: 7 September 2022						
Co-author 8 A/Prof Andrea Begley					X	X
I acknowledge that these represent my contribution to the above research output Signed: [redacted] Date: 19th September 2022						
Co-author 9 A/Prof Lucinda Black	X				X	X
I acknowledge that these represent my contribution to the above research output Signe [redacted] Date: 19th September 2022						

B.3 Copyright record

Obesity, dieting, and multiple sclerosis



Appendix C: Copyright records

Figure 1.1: Three main phenotypes of multiple sclerosis, graphically represented by disability accumulation over time, relapses, and new magnetic resonance imaging (MRI) activity. Images from the National MS Society[28] (data sourced from Lublin et al., 2014[26]).

Re: FW: National Contact Form

C Chatafterhours <Chatafterhours@nmss.org>
To Rebecca Russell

Reply | Reply All | Forward

Tue 20/09/2022 3:35 AM

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Warm regards,

Tara Hensley, MSCIR

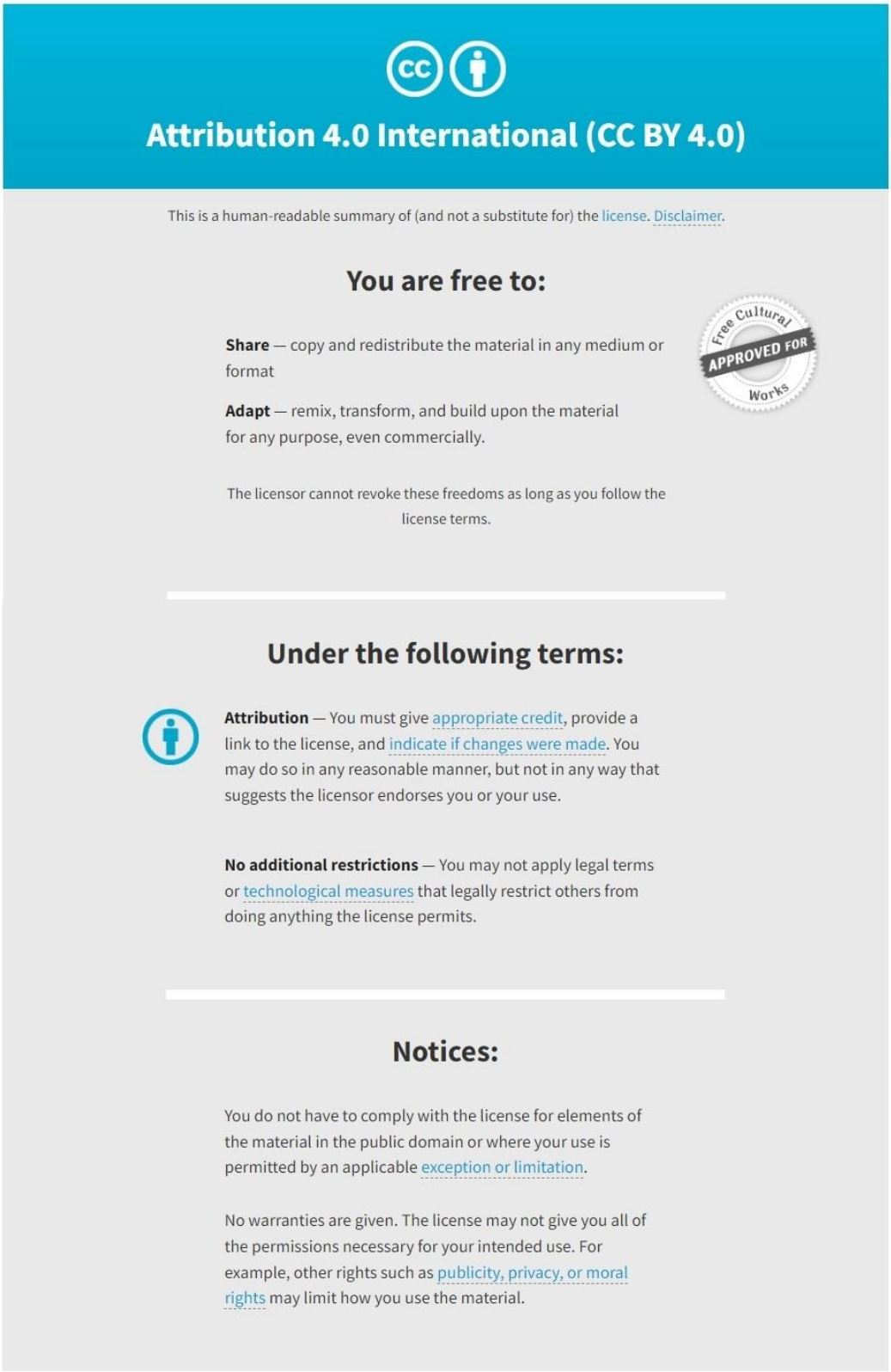
Manager, MS Navigator Services Delivery

MS Navigator Experience Team

Pronouns: She/Her

National Multiple Sclerosis Society

Nutrition education programs for adults with neurological diseases are lacking: a scoping review



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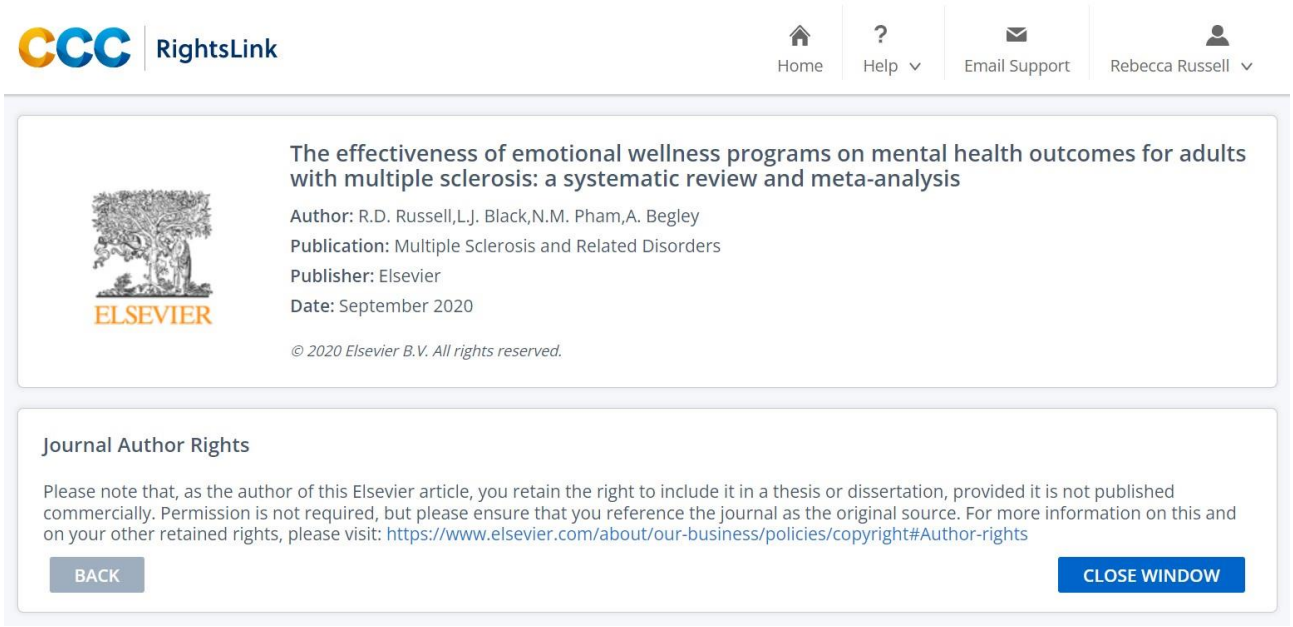
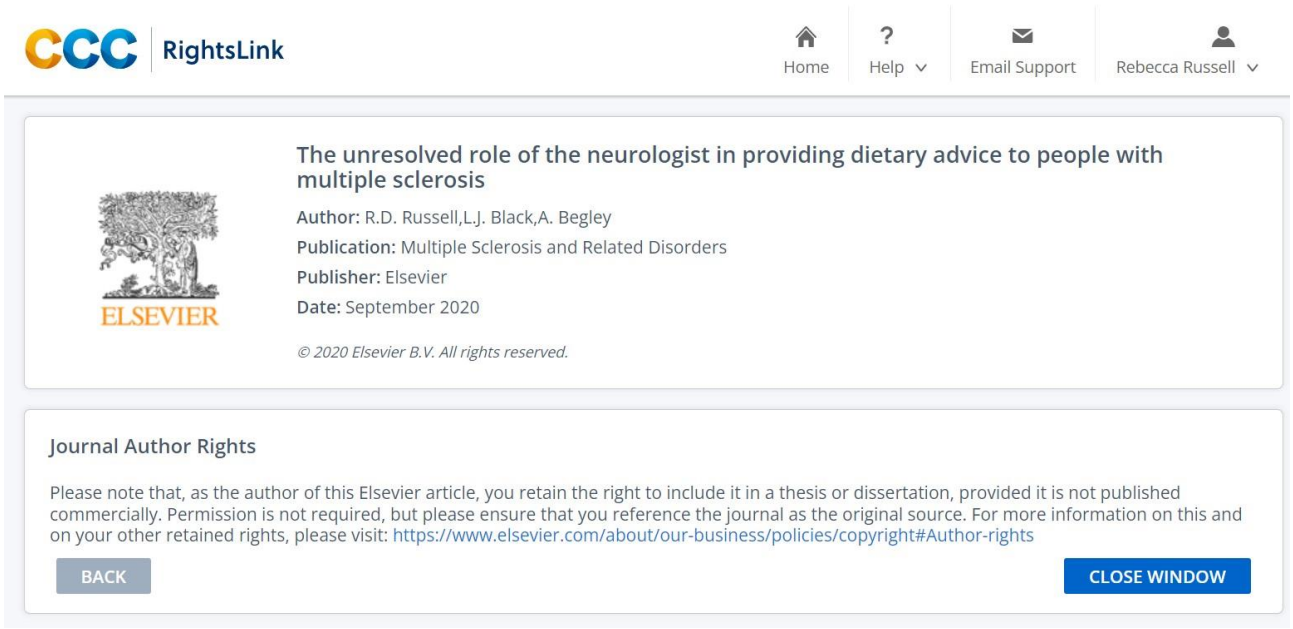
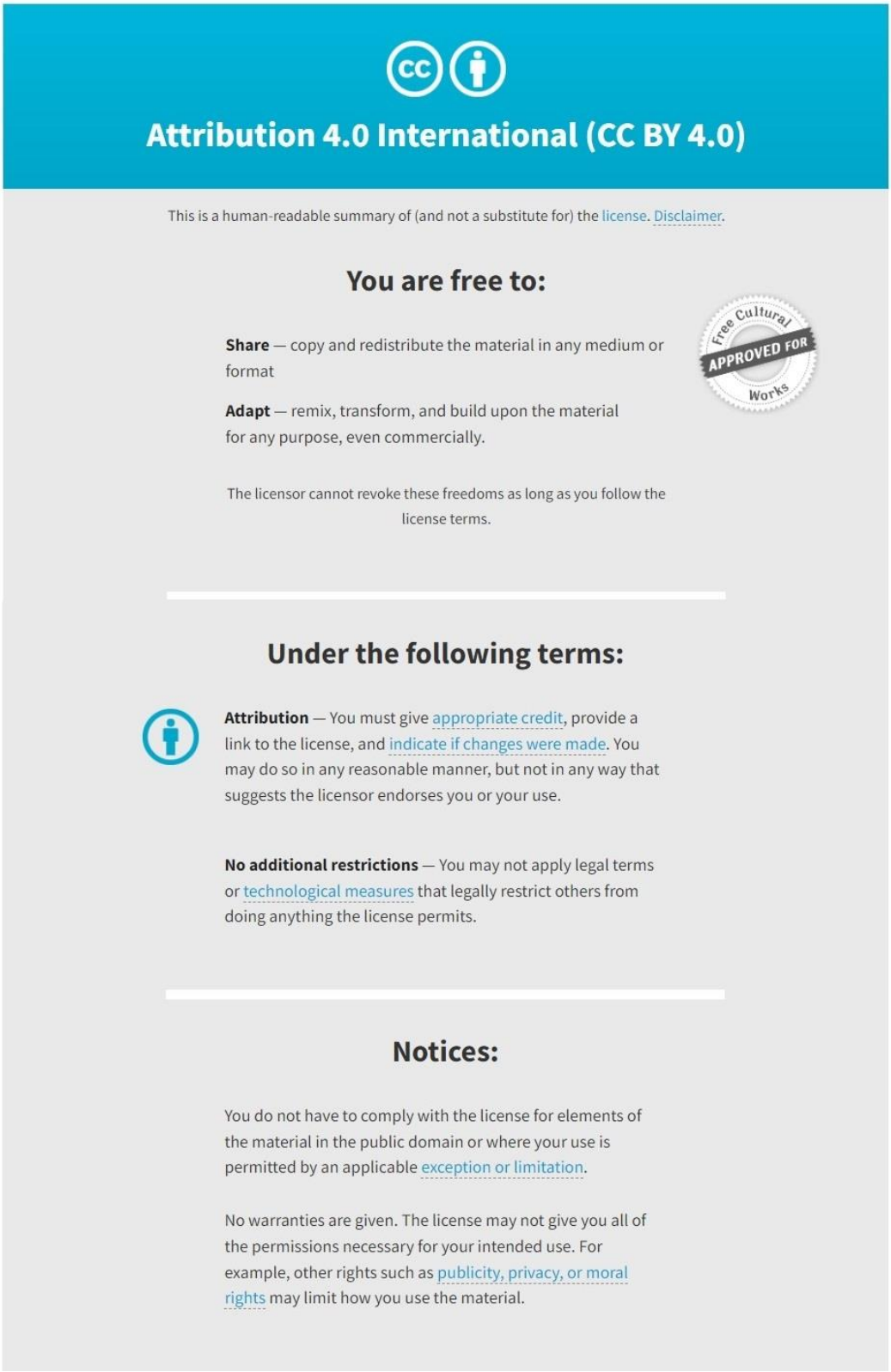
The effectiveness of emotional wellness programs on mental health outcomes for adults with multiple sclerosis: a systematic review and meta-analysis



The unresolved role of the neurologist in providing dietary advice to people with multiple sclerosis





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Appendix D: Published scoping review protocol

Appendix D.1 Published paper

The content of this appendix is covered by:

Russell, R. D., Black, L. J., & Begley, A. (2020). Dietary education programs for adults with neurological diseases: a scoping review protocol. JBI Evid Synth, 19(1), 170-176. https://doi.org/10.11124/JBISRIR-D-19-00394

The contribution of co-authors, A/Prof Andrea Begley and A/Prof Lucinda Black are detailed in the author attribution statements in Appendix D.2

The version that appears in this thesis appendix is of an article that has been through peer review with *JBI Evidence Synthesis* but has not been through the copyediting process.

Dietary education programs for adults with neurological diseases: a scoping review protocol

Rebecca D Russell[1]
Lucinda J Black[1]
Andrea Begley[1]

1. School of Public Health, Curtin University, Perth, Australia

Corresponding author:
Dr Andrea Begley
a.begley@curtin.edu.au

Acknowledgements

This review will partially fulfil the requirements for the degree, Doctor of Philosophy in the School of Public Health, Curtin University, for RDR. The reviewers would like to acknowledge the assistance of Diana Blackwood, Health Sciences Faculty Librarian (Curtin University), for her assistance in developing the search strategy.

Funding

RDR is supported by an Australian Government Research Training Program Scholarship and a Multiple Sclerosis Western Australia PhD Top-Up Scholarship. LJB is supported by an MS Research Australia Postdoctoral Fellowship and a Curtin University Research Fellowship. Funders had no role in the conceptualization or creation of this protocol.

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D.2 Attribution statement

Dietary education programs for adults with neurological disease: a scoping review protocol

	Conception and Design	Drafting protocol	Editing	Final Approval
Co-author 1 Mrs Rebecca Russell	X	X	X	X
I acknowledge that these represent my contribution to the above research output Signed: [signature redacted] Date: 25th August 2022				
Co-author 2 A/Prof Lucinda Black	X		X	X
I acknowledge that these represent my contribution to the above research output Signed: [signature redacted] Date: 19th September 2022				
Co-author 3 A/Prof Andrea Begley	X		X	X
I acknowledge that these represent my contribution to the above research output Signed: [signature redacted] Date: 19th September 2022				

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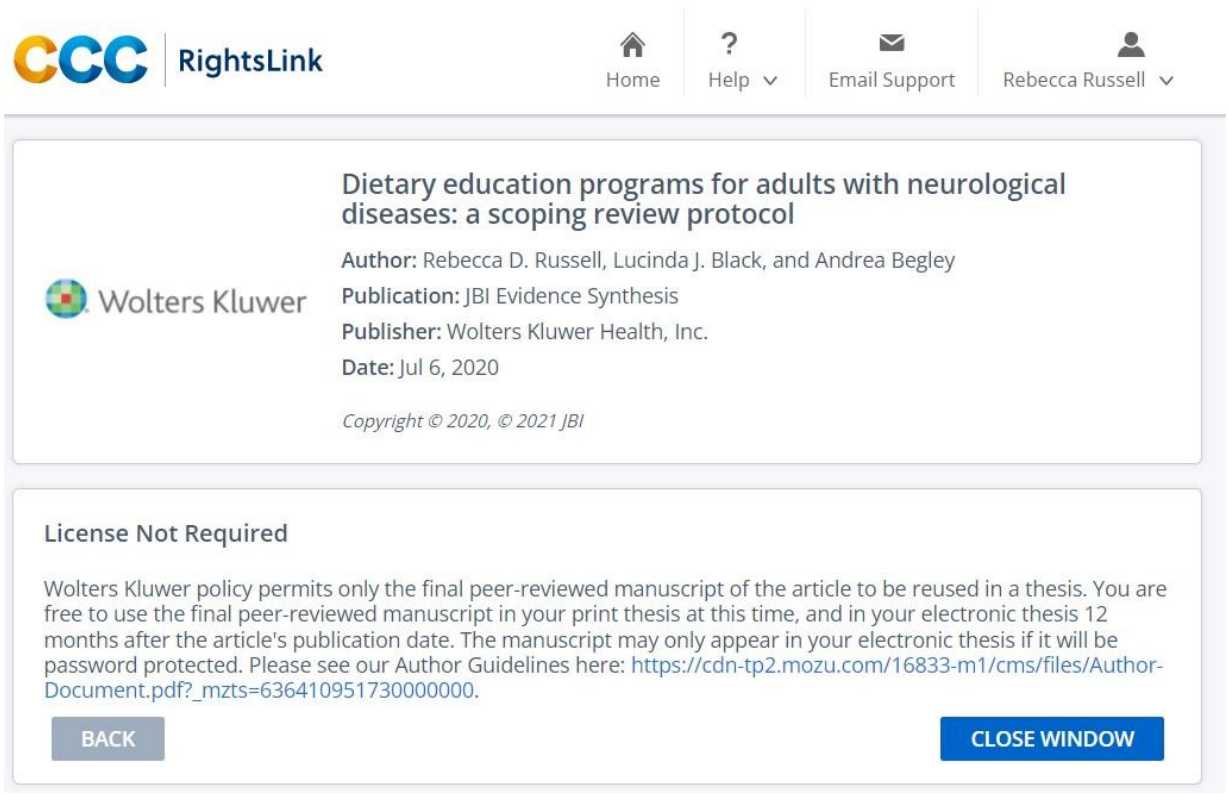

Appendix E: Documents from Chapter 4: The perceived role of neurologists in providing dietary advice to people with multiple sclerosis

E.1 Consent form

CONSENT FORM

HREC Project Number:	HRE2019-0179
Project Title:	Developing a dietary education program for people with multiple sclerosis
Chief Investigator:	Dr Andrea Begley, Senior Lecturer
Student researcher:	Rebecca Russell
Version Number:	V1
Version Date:	08/03/2019

- I have read the information statement version listed above and I understand its contents.
- I believe I understand the purpose, extent and possible risks of my involvement in this project.
- I voluntarily consent to take part in this research project.
- I have had an opportunity to ask questions and I am satisfied with the answers I have received.
- I understand that this project has been approved by Curtin University Human Research Ethics Committee and will be carried out in line with the National Statement on Ethical Conduct in Human Research (2007).
- I understand I will receive a copy of this Information Statement and Consent Form.

☐ I do	☐ I do not	consent to being audio-recorded
☐ I do	☐ I do not	consent to be contacted about future research projects that are related to this project
☐ I do	☐ I do not	consent to the storage and use of my information in future ethically-approved research projects related to this (project/disease)
☐ I do	☐ I do not	consent to you using any data I provided before withdrawing from the study, if relevant

Participant Name	
Participant Signature	
Date	

Declaration by researcher: I have supplied an Information Letter and Consent Form to the participant who has signed above, and believe that they understand the purpose, extent and possible risks of their involvement in this project.

Researcher Name	
Researcher Signature	
Date	

Note: All parties signing the Consent Form must date their own signature.

E.2 Participant information statement

Curtin University

A dietary education program for MS

PARTICIPANT INFORMATION STATEMENT

HREC Project Number:	HRE2019-0179
Project Title:	Developing a dietary education program for people with multiple sclerosis
Chief Investigator:	Dr Andrea Begley, Senior Lecturer
Student researcher:	Rebecca Russell
Version Number:	V2
Version Date:	29/03/2019

What is the Project About?

- We know that dietary education has the potential to improve the health, wellbeing, and sense of control in people with multiple sclerosis (MS), but there has been very little research done in this area. There is lots of information about diets available online, but it's not always based on current evidence, so it can be confusing trying to work out what is reliable information.
- Currently, there are no dietary education programs being run by MS organisations in Australia, and it is not clear what dietary advice is provided to people with MS when they are diagnosed.
- With this project, we aim to find out what information is provided to people with MS about diet.
- This research is important so that we can develop a dietary education program that fills in the gaps in this field.
- We will interview 10-12 neurologists.

Who is doing the Research?

- The project is being conducted by PhD candidate Rebecca Russell, and her supervisors Dr Andrea Begley and Dr Lucinda Black.
- The results of this research project will be used by Rebecca Russell to obtain a Doctor of Philosophy at Curtin University and is funded by the University.
- There will be no costs to you and you will not be paid for participating in this project.

Why am I being asked to take part and what will I have to do?

- You have been asked to take part because you are a neurologist in Western Australia who diagnoses people with MS

- Your participation will involve having a one-to-one casual interview with Rebecca, at a time that best suits you. The interview will last for up to half an hour, and you will be asked to fill out a short questionnaire before the interview starts, with some standard questions about your age, sex etc.
- The study will take place at a mutually convenient location, or via a computer-based web conferencing program, such as WebEx or Skype.
- We will ask you questions about dietary advice for people with MS.

- There will be no cost to you for taking part in this research and you will not be paid for taking part.
- We will make a digital audio recording so we can concentrate on what you have to say and not distract ourselves with taking notes. After the interview we will make a full written copy of the recording.
  - Optional Consent Future Research: We would like you to consider allowing us to send you information about future research projects. Once you receive the information it is your choice if you decide to take part or not.
  - Optional Consent: We would like you to consider letting us share the information we collect during this research with other researchers working in this area. The information will be de-identified then re-identifiable, meaning your name will be changed.

Are there any benefits' to being in the research project?

- There may be no direct benefit to you from participating in this research, but sometimes, people appreciate the opportunity to discuss their opinions.
- We hope the results of this research will allow us to:
  - Develop a dietary education program for people with MS that is tailor-made for them.

Are there any risks, side-effects, discomforts or inconveniences from being in the research project?

- We have been careful to make sure that the questions in the interview do not cause you any distress. But, if you feel anxious about any of the questions you do not need to answer them. If the questions cause any concerns or upset you, we can refer you to a counsellor.
- Apart from giving up your time, we do not expect that there will be any risks or inconveniences associated with taking part in this study.

Who will have access to my information?

- The information collected in this research will be re-identifiable (coded). This means that we will collect data that can identify you, but will then remove identifying information on any data or sample and replace it with a code when we analyse the data. Only the research team have access to the code to match your name if it is necessary to do so. Any information we collect will be treated as confidential and used only in this project unless

otherwise specified. The following people will have access to the information we collect in this research: the research team and, in the event of an audit or investigation, staff from the Curtin University Office of Research and Development.

- Electronic data will be password-protected and hard copy data (including audio tapes) will be in locked storage.
- The information we collect in this study will be kept under secure conditions at Curtin University for 7 years after the research is published and then it will be destroyed.
- The results of this research may be presented at conferences or published in professional journals. You will not be identified in any results that are published or presented.

Will you tell me the results of the research?

- If you are interested in obtaining a summary of the results please contact the researchers after December 2019. Results will not be individual but based on all the information we collect and review as part of the research.

Do I have to take part in the research project?

- Taking part in a research project is voluntary. It is your choice to take part or not. You do not have to agree if you do not want to. If you decide to take part and then change your mind, that is okay, you can withdraw from the project. If you choose not to take part or start and then stop the study, it will not affect your relationship with the University, staff or colleagues.
- You are free to withdraw from the study prior to approving your transcript.
- With your permission, if you chose to leave the study we will use any information collected unless you tell us not to.

What happens next and who can I contact about the research?

- You can contact Dr Andrea Begley on 9266 2773 or a.begley@curtin.edu.au to obtain further information or ask any questions.
    - If you decide to take part in this research we will ask you to sign the consent form. By signing it is telling us that you understand what you have read and what has been discussed. Signing the consent indicates that you agree to be in the research project and have your information used as described. Please take your time and ask any questions you have before you decide what to do. You will be given a copy of this information and the consent form to keep.

Curtin University Human Research Ethics Committee (HREC) has approved this study (HREC number HRE2019-0179). Should you wish to discuss the study with someone not directly involved, in particular, any matters concerning the conduct of the study or your rights as a participant, or you wish to make a confidential complaint, you may contact the Ethics Officer on (08) 9266 9223 or the Manager, Research Integrity on (08) 9266 7093 or email hrec@curtin.edu.au.

E.3 Demographic questionnaire

DEMOGRAPHIC INFORMATION

Number of years practising medicine: _____

Number of years since qualifying as a neurologist: _____

Country of medical training: _____

In the last 3 days, approximately what proportion (%) of your time was spent with patients with multiple sclerosis? _____

Would you like a copy of your interview transcript posted to your clinic rooms?

Yes ☐ No ☐

Or alternative address:

Name: _____

Postal address: _____

Suburb: _____ Postcode: _____

Appendix F: Documents from Chapter 5: How people with multiple sclerosis navigate dietary advice

F.1 Consent form

A dietary education program for MS

Curtin University

CONSENT FORM

HREC Project Number:	HRE2019-0179
Project Title:	Developing a dietary education program for people with multiple sclerosis
Chief Investigator:	Dr Andrea Begley, Senior Lecturer
Student researcher:	Rebecca Russell
Version Number:	V1
Version Date:	08/03/2019

- I have read the information statement version listed above and I understand its contents.
- I believe I understand the purpose, extent and possible risks of my involvement in this project.
- I voluntarily consent to take part in this research project.
- I have had an opportunity to ask questions and I am satisfied with the answers I have received.
- I understand that this project has been approved by Curtin University Human Research Ethics Committee and will be carried out in line with the National Statement on Ethical Conduct in Human Research (2007).
- I understand I will receive a copy of this Information Statement and Consent Form.

☐ I do	☐ I do not	consent to being audio-recorded
☐ I do	☐ I do not	consent to be contacted about future research projects that are related to this project
☐ I do	☐ I do not	consent to the storage and use of my information in future ethically-approved research projects related to this (project/disease)
☐ I do	☐ I do not	consent to you using any data I provided before withdrawing from the study, if relevant

Participant Name	
Participant Signature	
Date	

Declaration by researcher: I have supplied an Information Letter and Consent Form to the participant who has signed above, and believe that they understand the purpose, extent and possible risks of their involvement in this project.

Researcher Name	
Researcher Signature	
Date	

Note: All parties signing the Consent Form must date their own signature.

F.2 Participant information statement

Curtin University

A dietary education program for MS

PARTICIPANT INFORMATION STATEMENT

HREC Project Number:	HRE2019-0179
Project Title:	Developing a dietary education program for people with multiple sclerosis
Chief Investigator:	Dr Andrea Begley, Senior Lecturer
Student researcher:	Rebecca Russell
Version Number:	V2
Version Date:	29/03/2019

What is the Project About?

- We know that dietary education has the potential to improve the health, wellbeing, and sense of control in people with multiple sclerosis (MS), but there has been very little research done in this area. There is lots of information about diets available online, but it's not always based on current evidence and it can be confusing trying to work out what is reliable information.
- Currently, there are no dietary education programs being run by MS organisations in Australia.
- With this project, we aim to find out exactly what people with MS want from a dietary education program. For example, what topics do they want covered, how long should the program run for, and in what format (e.g. online or in-person) etc.
- This research is important, so that we can develop a dietary education program that reflects the wants, needs, values, and behaviours of people with MS.
- We will run at least 2 focus groups from each of the following categories, with 5-15 people in each group:
    - People with MS
    - MS nurses
    - Other MS allied health professionals

Who is doing the Research?

- The project is being conducted by PhD candidate Rebecca Russell, and her supervisors Dr Andrea Begley and Dr Lucinda Black.
- The results of this research project will be used by Rebecca Russell to obtain a Doctor of Philosophy at Curtin University and is funded by the University.
- There will be no costs to you and you will not be paid for participating in this project. You will receive a gift voucher as a thank you for your time.

Why am I being asked to take part and what will I have to do?

- You have been asked to take part because you have either been diagnosed with MS, or you are a health professional who works with people with MS
- Your participation will involve taking part in a one-off focus group, with other people with MS or other MS health professionals. Rebecca will run the focus group, which will last for around 1.5 hours. You will be asked to fill out a short questionnaire before the focus group starts, with some standard questions about your age, sex, employment status etc.
- The study will take place at the MSWA Wilson location: 29 Parkhill Way, Wilson, or another MSWA venue. There is free parking available onsite.
- The focus group questions will cover topics such as "What are some things that could make it easier for people with MS to make healthier food choices?" and "What topics would you like to have covered in a dietary education program for MS?"
- We will make a digital audio recording so we can concentrate on what you have to say and not distract ourselves with taking notes. After the focus group we will make a full written copy of the recording.
  - Optional Consent Future Research: We would like you to consider allowing us to send you information about future research projects. Once you receive the information it is your choice if you decide to take part or not.
  - Optional Consent: We would like you to consider letting us share the information we collect during this research with other researchers working in this area. The information will be de-identified then re-identifiable, meaning your name will be changed.

Are there any benefits' to being in the research project?

- There may be no direct benefit to you from participating in this research, but sometimes, people appreciate the opportunity to discuss their opinions.
- We hope the results of this research will allow us to:
  - Develop a dietary education program for people with MS that is tailor-made for them.

Are there any risks, side-effects, discomforts or inconveniences from being in the research project?

- We have been careful to make sure that the questions in the focus groups do not cause you any distress. But, if you feel anxious about any of the questions you do not need to answer them. If the questions cause any concerns or upset you, we can refer you to a counsellor.
- Apart from giving up your time, we do not expect that there will be any risks or inconveniences associated with taking part in this study.

Who will have access to my information?

- The information collected in this research will be re-identifiable (coded). This means that we will collect data that can identify you, but will then remove identifying information on any data or sample and replace it with a code when we analyse the data. Only the research team have access to the code to match your name if it is necessary to do so. Any information we collect will be treated as confidential and used only in this project unless otherwise specified. The following people will have access to the information we collect in

this research: the research team and, in the event of an audit or investigation, staff from the Curtin University Office of Research and Development.

- Electronic data will be password-protected and hard copy data (including audio tapes) will be in locked storage.
- The information we collect in this study will be kept under secure conditions at Curtin University for 7 years after the research is published and then it will be destroyed.
- The results of this research may be presented at conferences or published in professional journals. You will not be identified in any results that are published or presented.
- Whilst all care will be taken to maintain privacy and confidentiality of any information shared at a focus group or group discussion, you should be aware that you may feel embarrassed or upset if one of the group members repeats things said in a confidential group meeting.

Will you tell me the results of the research?

- If you are interested in obtaining a summary of the results please contact the researchers after December 2019. Results will not be individual but based on all the information we collect and review as part of the research.

Do I have to take part in the research project?

- Taking part in a research project is voluntary. It is your choice to take part or not. You do not have to agree if you do not want to. If you decide to take part and then change your mind, that is okay, you can withdraw from the project. If you choose not to take part or start and then stop the study, it will not affect your relationship with the University, staff or colleagues.
- You are free to withdraw from the study prior to approving your transcript.
- With your permission, if you chose to leave the study we will use any information collected unless you tell us not to.

What happens next and who can I contact about the research?

- You can contact Dr Andrea Begley on 9266 2773 or a.begley@curtin.edu.au to obtain further information or ask any questions.
    - If you decide to take part in this research we will ask you to sign the consent form. By signing it is telling us that you understand what you have read and what has been discussed. Signing the consent indicates that you agree to be in the research project and have your information used as described. Please take your time and ask any questions you have before you decide what to do. You will be given a copy of this information and the consent form to keep.

Curtin University Human Research Ethics Committee (HREC) has approved this study (HREC number HRE2019-0179). Should you wish to discuss the study with someone not directly involved, in particular, any matters concerning the conduct of the study or your rights as a participant, or you wish to make a confidential complaint, you may contact the Ethics Officer on (08) 9266 9223 or the Manager, Research Integrity on (08) 9266 7093 or email hrec@curtin.edu.au.

F.3 Demographic questionnaire

A dietary education program for MS

Curtin University

DEMOGRAPHIC INFORMATION

Age: _____

Gender: _____

Country of birth: _____

Occupation: _____

How long ago were you diagnosed with MS? _____

What type of MS do you have (please tick)?

Relapsing-Remitting ☐ Primary-Progressive ☐ Secondary-Progressive ☐ Other or unsure ☐

Current employment status (please tick):

Employed ☐ Disability pension ☐ Other (please specify): ☐

_____

Would you like a copy of the focus group transcript posted to you?

Yes ☐ No ☐

If yes:

Name: _____

Postal address: _____

Suburb: _____ Postcode: _____

F.4 Program preference questions

A dietary education program for MS

Curtin University

PROGRAM QUESTIONS

Tell us about your preferences for MS-specific education programs:

What program delivery method do you like? *Tick as many that apply*

In-person ☐ Online ☐ Web-conference ☐ Other *(write below):* ☐

_____

Which of the following do you prefer? *Tick as many that apply*

Twice a week ☐ Once a week ☐ Once a fortnight ☐ Self-paced ☐ No preference ☐

Do you have a preference for time of day (morning, afternoon, or evening)?

_____

How many sessions do you like to attend? *Tick as many that apply*

One off ☐ 2-4 ☐ 5-7 ☐ 8 or more ☐ No preference ☐

How much time do you like to commit for a session?

Less than 1 hour ☐ 1-1.5 hours ☐ 1.5-2 hours ☐ 2 hours or more ☐

What topics would you like to have covered in a nutrition program? *List/describe as many as you like*

_____

_____

_____

_____

What goals would you like to achieve from a nutrition program?

_____

_____

_____

Any other comments about attending a nutrition program?

_____

Program Questionnaire Version 3, 23/09/2019

Appendix G: Documents from Chapter 6: Using co-design principles to develop an online nutrition education program for people with multiple sclerosis

G.1 Consumer workshops: consent form

CONSENT FORM

HREC Project Number:	HRE2019-0179
Project Title:	Developing, piloting, and evaluating a dietary education program for people with multiple sclerosis
Chief Investigator:	Associate Professor Andrea Begley
Student researcher:	Rebecca Russell
Version Number:	5
Version Date:	3rd May 2021

- I have read the Information Statement version listed above and I understand its contents.
- I believe I understand the purpose, extent and possible risks of my involvement in this project.
- I voluntarily consent to take part in this research project.
- I have had an opportunity to ask questions and I am satisfied with the answers I have received.
- I understand that this project has been approved by Curtin University Human Research Ethics Committee and will be carried out in line with the National Statement on Ethical Conduct in Human Research (2007).
- I understand I will receive a copy of the Information Statement and Consent Form.

Participant Name	
Participant Signature	
Date	

Declaration by researcher: I have supplied an Information Statement and Consent Form to the participant who has signed above.

Researcher Name	Rebecca Russell
Researcher Signature	
Date	

Note: All parties signing the Consent Form must date their own signature.

G.2 Consumer workshops: participant information statement

A nutrition education program for people with MS

Curtin University

PARTICIPANT INFORMATION STATEMENT

HREC Project Number:	HRE2019-0179
Project Title:	Developing, piloting, and evaluating a dietary education program for people with multiple sclerosis
Chief Investigator:	Associate Professor Andrea Begley
Student researcher:	Rebecca Russell
Version Number:	7
Version Date:	19th May 2021

What is the project about?

- We know that nutrition education has the potential to improve the health and wellbeing of people with multiple sclerosis (MS). There is lots of information online about diets for MS, but it's not always based on current evidence and it can be confusing trying to work out what is reliable information.
- In this project, we aim to develop an online nutrition education program for people with MS that has been co-designed by people with MS.
- In our previous research involving people with MS, we have collected information on the preferred characteristics and content of a nutrition education program.
- To ensure the program best meets the needs of people with MS, we are conducting two 2-3 hour workshops with eight (8) people with MS as we continue to develop the program.
- We will use the feedback to make changes to our program, which we will run in late 2021.

Who is doing the research?

- The project is being conducted by PhD student Rebecca Russell, and her supervisors Associate Professor Andrea Begley and Associate Professor Lucinda Black.
- The results of this research project will be used by Rebecca to obtain a Doctor of Philosophy at Curtin University, and is funded by the University. Rebecca is supported by an Australian Government Research and Training Program Scholarship, and an MSWA PhD Top-Up Scholarship.

Why am I being asked to take part and what will I have to do?

- You have been asked to take part because you have been diagnosed with MS within the last five (5) years.
- Your participation will involve taking part in two 2-3 hour workshops: one in June, and one in August 2021.
- The workshops will be held online.

- There will be no cost to you for taking part in this research, and we will pay you a $35 honorarium per workshop.
- During the workshops we will discuss the development of the nutrition education program, and ask for your input and feedback on the content and structure.
- We will take notes to record input and feedback from the workshops.

Are there any benefits to being in the research project?

- There may be no direct benefit to you from participating in this research, but sometimes people appreciate the opportunity to discuss their ideas and opinions.
- The results of this research will allow us to develop a nutrition education program for people with MS that has been co-designed by people with MS, so that it is tailor-made for them.
- By taking part in the workshops, you will not be able to enrol in the trial to test the nutrition education program, but you will have the opportunity to enrol in the program once the trial has been completed (late 2021).

Are there any risks, side-effects, discomforts or inconveniences from being in the research project?

- We do not anticipate that the content of the nutrition education program will cause you any distress. But if you feel anxious about any of the topics, you do not need to provide input on them. If the topics cause any concerns or upset you, we can refer you to a counsellor.
- Apart from giving up your time, we do not expect that there will be any risks or inconveniences associated with taking part in this study.
- To minimise any risks in relation to the COVID-19 pandemic, the workshops will be held online.

Who will have access to my information?

- The information collected in this research will be re-identifiable (coded). This means that we will collect data that can identify you, but will then remove identifying information on any data or sample and replace it with a code when we analyse the data. Only the research team have access to the code to match your name if it is necessary to do so. Any information we collect will be treated as confidential and used only in this project unless otherwise specified. The following people will have access to the information we collect in this research: the research team and, in the event of an audit or investigation, staff from the Curtin University Office of Research and Development
- Electronic data will be password-protected and hard copy data (including consent forms) will be in locked storage.
- The information we collect in this study will be kept under secure conditions at Curtin University for 7 years after the research is published and then it will be destroyed.
- The results of this research may be presented at conferences or published in professional journals. You will not be identified in any results that are published or presented.
- Whilst all care will be taken to maintain privacy and confidentiality of any information shared at a focus group or group discussion, you should be aware that you may feel embarrassed or upset if one of the group members repeats things said in a confidential group meeting.

Will you tell me the results of the research?

- If you are interested in obtaining a summary of the results, please contact the researchers after October 2021. Results will not be individual, but based on all the information we collect and review as part of the research.

Do I have to take part in the research project?

- Taking part in the research project is voluntary. It is your choice to take part or not. You do not have to agree if you do not want to. If you decide to take part and then change your mind, that is okay, you can withdraw from the project. If you choose not to take part or start and then stop the study, it will not affect your relationship with the University, staff or colleagues, or any MS organisations.
- With your permission, if you choose to leave the study we will use any information collected unless you tell us not to.

What happens next and who can I contact about the research?

- You can contact Associate Professor Andrea Begley on 9266 2773 or MSDietProject@curtin.edu.au to obtain further information or ask any questions.
- If you decide to take part in this research we will ask you to sign the consent form (either a digital signature; or you can print, scan, and email it back; or we can post you a form with a reply-paid envelope). By signing it is telling us that you understand what you have read and what has been discussed. Signing the consent form indicates that you agree to be in the research project and have your information used as described. Please take your time and ask any questions you have before you decide what to do. You will be given a copy of this information and the consent form to keep.

Curtin University Human Research Ethics Committee (HREC) has approved this study (HREC number HRE2019-0179). Should you wish to discuss the study with someone not directly involved, in particular, any matters concerning the conduct of the study or your rights as a participant, or you wish to make a confidential complaint, you may contact the Ethics Officer on (08) 9266 9223 or the Manager, Research Integrity on (08) 9266 7093 or email hrec@curtin.edu.au.

G.3 Consumer workshops: discussion guide workshop #1

Project title: Developing, piloting, and evaluating a dietary education program for people with multiple sclerosis.
Formative workshops. Discussion and topic guide

Workshop #1.

Welcome:

- Purpose of the workshops - provide a valuable lived experience perspective on the program including topics – order, what is missing, and the discussion board rules. In the second workshop we will look at the program design, language, and format.
- This workshop is a 2-way conversation, and everyone will have the opportunity to express their views. Some of you may have different views to others, so we please ask that we all are respectful of each other.
- We are developing a nutrition education program that we plan to test how well it is received in a pilot study later this year (Oct-Nov 2021). At some stage after the pilot study has been completed, you will be offered the opportunity to enrol and complete the program.

Confidentiality agreements

House-keeping:

- Mic and camera control icons down the bottom
  - Mute/unmute microphones – unmute to talk, and then mute when finished speaking
  - Cameras on/off – can turn off if you like when you get up to go to the bathroom etc
- How to use 'raise hand' and chat features – down the bottom (may need to press 3 dots on mobile device to see options)

Format of the workshop:

- 2-3 hours.
- Scheduled breaks, but please feel welcome to break as you need for bathroom etc, and eat/drink as you need.
- I will present a small section of information, then ask each participant for comments – you may like to write down your comments or questions.
- Student dietitian is assisting with this research and will take notes so that we can incorporate your feedback into the program (no video or audio recording).

Introductions & icebreaker: researchers, each participant. Icebreaker topic: If you could go anywhere in Australia tomorrow, without COVID restrictions, where would you go?

Background: Research journey timeline – beginning with my Honours research in 2017… the research projects that have informed the current form of the nutrition education program… involvement of consumer and stakeholders

Format: present the current format of the program (number of weeks, number of sessions, duration to complete each session). Invite participants to comment.

Topics: present the topics and subtopics for each of the modules. Invite participants to comment and discuss as we present each module.
Module 0 - Present discussion board rules and invite participants to comment

Program name: brainstorm name for program
Summary of key points by one student dietitian

Wrap-up: Thank participants. Reminder of next workshop.

G.4 Consumer workshops: discussion guide workshop #2

Curtin University

Project title: Developing, piloting, and evaluating a dietary education program for people with multiple sclerosis.

Formative workshops. Discussion and topic guide

Workshop #2.

Welcome:

- Reminder of the purpose of the workshops - provide a valuable lived experience perspective, and input on the nutrition program activities.
- This workshop is a 2-way conversation, and everyone will have the opportunity to express their views. Some of you may have different views to others, so we please Q# that we all are respectful of each other.

Confidentiality

House-keeping:

- Mic and camera control icons down the bottom
  - Mute/unmute microphones – unmute to talk, and then mute when finished speaking
  - Cameras on/off – can turn off if you like when you get up to go to the bathroom etc

Format of the workshop:

- 2-3 hours.
- Scheduled breaks, but please feel welcome to break as you need for bathroom etc, and eat/drink as you need.
- I will present a small section of information, then ask each participant for comments – you may like to write down your comments or questions.
- Student dietitians assisting with this research and will take notes so that we can incorporate your feedback into the program (no video or audio recording).

Icebreaker: What is your favourite season, and why?

Reminder – nutrition program format:

- Online, asynchronous, 6 weeks
- 1-2 hours per week
- Written content, videos, workbook activities, discussion board topics, links for further reading
- Q#: Has anyone done any other online courses (not just nutrition?) What did they like, what didn't they like

Activities:

- Present proposed activities (by module. Show slide, then show Word Doc activity book). Invite comments.
  - **Activities to try:**
    - Goal setting
    - Recipe modification (if time)
- **Answers for activities**: Where would they like to have the answers (e.g. at the back of the workbook, right after the activity, at the start of the following module, or other suggestions?)

Summary of key points by one student dietitian

Wrap-up: Any other questions or comments? Thank participants. Advise participants that the timeline is evolving ("talk aloud" interviews to test module 1 online (Sept-Nov). But they will still be invited to complete the program once a pilot study has taken place.

G.5 Cognitive interviews: consent form

CONSENT FORM

HREC Project Number:	HRE2019-0179
Project Title:	Developing, piloting and evaluating a dietary education program for people with multiple sclerosis.
Chief Investigator:	Associate Professor Andrea Begley
Student researcher:	Rebecca Russell
Version Number:	1
Version Date:	5th August 2021

The following boxes will appear at the top of the demographic questionnaire.

☐ I have received information regarding this research project and had an opportunity to ask questions. I believe I understand the purpose, extent and possible risks of my involvement in this project and I voluntarily consent to take part.

☐ I do ☐ I do not consent to you using any data I provided before withdrawing from the study, if relevant

☐ I do ☐ I do not consent to be contacted about future research projects that are related to this project

If 'I do', please enter email address _____

G.6 Cognitive interviews: participant information statement

PARTICIPANT INFORMATION STATEMENT

HREC Project Number:	HRE2019-0179
Project Title:	Developing, piloting, and evaluating a dietary education program for people with multiple sclerosis
Chief Investigator:	Associate Professor Andrea Begley
Student researcher:	Rebecca Russell
Version Number:	3
Version Date:	2nd November 2021

What is the project about?

- We know that nutrition education has the potential to improve the health and wellbeing of people with multiple sclerosis (MS). We are wanting to work with people with MS to design an online nutrition education program.
- In our previous research involving people with MS, we have collected information on the preferred topics and format of a nutrition education program.
- This project will use interviews to explore the ease of understanding, and the use of content, format, and design of one module in the online nutrition program.
- To ensure the information in the program to easy to understand and best meets the needs of people with MS, we are conducting one-on-one interviews with six (6) to eight (8) people as they work through one module of the program.
- We will use the feedback to make changes to our program, which we will run in 2022.

Who is doing the research?

- The project is being conducted by PhD student Rebecca Russell, and her supervisors Associate Professor Andrea Begley and Associate Professor Lucinda Black.
- The results of this research project will be used by Rebecca to obtain a Doctor of Philosophy at Curtin University, and is funded by the University. Rebecca is supported by an Australian Government Research and Training Program Scholarship, and an MSWA PhD Top-Up Scholarship.

Why am I being asked to take part and what will I have to do?

- You have been asked to take part because you have been diagnosed with MS within the last five (5) years.
- Your participation will involve having an online one-on-one interview with Rebecca.
- You will be asked to complete a short online questionnaire before the interview starts with some standard questions about your age, sex etc.
- We will also ask you to complete a short online questionnaire after the interview has finished, at a time that is convenient for you.

- The interview will last for one (1) hour.

- Rebecca will ask you to work through sections of the online nutrition education program, and she will ask you to "think aloud" as you work through. After each section, Rebecca will ask you questions about how easy or difficult the information was to understand.
- We also want your opinions on the content, format, and design.
- There will be no cost to you for taking part in this research.
- We will make a digital video recording so we can concentrate on what you have to say and not distract ourselves with taking notes. After the interview we will make a full written copy of the recording.

    - Optional Consent Future Research: We would like you to consider allowing us to send you information about future research projects. Once you receive the information it is your choice if you decide to take part or not.

Are there any benefits to being in the research project?

- There may be no direct benefit to you from participating in this research, but sometimes people appreciate the opportunity to discuss their ideas and opinions.
- We hope that the results of this research will allow us to develop a nutrition education program for people with MS that has been co-designed by people with MS, so that it is tailor-made for them.
- By taking part in the interviews, you will not be able to enrol in the pilot study to test the nutrition education program, but you will have the opportunity to enrol in the nutrition program once the pilot study has been completed (2022).

Are there any risks, side-effects, discomforts or inconveniences from being in the research project?

- We do not anticipate that the content of the nutrition education program will cause you any distress. But if you feel anxious about any of the topics, you do not need to provide input on them. If the topics cause any concerns or upset you, we can refer you to a counsellor.
- Apart from giving up your time, we do not expect that there will be any risks or inconveniences associated with taking part in this study.
- To minimise any risks in relation to the COVID-19 pandemic, the interviews will be held online.

Who will have access to my information?

- The information collected in this research will be re-identifiable (coded). This means that we will collect data that can identify you, but will then remove identifying information on any data and replace it with a code when we analyse the data. Only the research team have access to the code to match your name if it is necessary to do so. Any information we collect will be treated as confidential and used only in this project unless otherwise specified. The following people will have access to the information we collect in this research: the research team and, in the event of an audit or investigation, staff from the Curtin University Office of Research and Development
- Electronic data will be password-protected and hard copy data will be in locked storage.

- The information we collect in this study will be kept under secure conditions at Curtin University for 7 (7) years after the research is published and then it will be destroyed.
- The results of this research may be presented at conferences or published in professional journals. You will not be identified in any results that are published or presented.

Will you tell me the results of the research?

- If you are interested in obtaining a summary of the results, please contact the researchers after January 2022. Results will not be individual, but based on all the information we collect and review as part of the research.

Do I have to take part in the research project?

- Taking part in the research project is voluntary. It is your choice to take part or not. You do not have to agree if you do not want to. If you decide to take part and then change your mind, that is okay, you can withdraw from the project. If you choose not to take part or start and then stop the study, it will not affect your relationship with the University, staff or colleagues, or any MS organisations.
- You are free to withdraw from the study prior to approving your transcript.
- With your permission, if you chose to leave the study we will use any information collected unless you tell us not to.

What happens next and who can I contact about the research?

- You can contact Associate Professor Andrea Begley on 9266 2773 or MSDietProject@curtin.edu.au to obtain further information or ask any questions.
- If you decide to take part in this research we will ask you tick a box on the online consent form. Ticking the box tells us that you understand what you have read and what has been discussed. Ticking the box on the online consent form indicates that you agree to be in the research project and have your information used as described. Please take your time and ask any questions you have before you decide what to do. You will be given a copy of this information and the consent form to keep.

Curtin University Human Research Ethics Committee (HREC) has approved this study (HREC number HRE2019-0179). Should you wish to discuss the study with someone not directly involved, in particular, any matters concerning the conduct of the study or your rights as a participant, or you wish to make a confidential complaint, you may contact the Ethics Officer on (08) 9266 9223 or the Manager, Research Integrity on (08) 9266 7093 or email hrec@curtin.edu.au.

G.7 Cognitive interviews: demographic questionnaire

Curtin University

Project title: Developing, piloting and evaluating a dietary education program for people with multiple sclerosis.

SOCIO-DEMOGRAPHIC INFORMATION

Qualtrics survey

Age (years)

_____

Sex: (select one)
Female
Male
Non-binary

Which type of MS have you been diagnosed with? (select one)
Relapsing-remitting
Secondary-progressive
Primary-progressive
Progressive-relapsing
Unsure
Other (please specify) _____

How long ago were you diagnosed with MS? (Years and months) *If less than 1 year, enter '0' for years*
____ (years)
____ (months)

What is the highest level of education that you have completed? (select one)
Below year 12
Year 12 or equivalent
Trade/Apprenticeship
TAFE technical certificate/Diploma
Bachelor degree
Postgraduate degree

What is your current employment status?
Employed (including full-time, part-time, casual, and self-employed)
Unemployed (including looking for work, volunteering, and homemaker)
Retired
Disability pension
Student
Other (please specify) _____

Would you like a copy of your interview transcript emailed to you?
No
Yes
If yes, email address: _____

G.8 Cognitive interviews: discussion guide

Project title: Developing, piloting, and evaluating a dietary education program for people with multiple sclerosis.

Cognitive interviews. Topic guide

Welcome:

- **Introductions**, thank participant.
- **Establish rapport**
- **Purpose** of the project, and interest in hearing what s/he has to say about the materials
- Explain how the interview will go **(format)**
- Confirm audio & video **recording**

General questions and probes (after each section):

- What thoughts came to mind while reading that? / What thoughts came to mind while looking at that (graphic/video)?
    - Probe - usefulness
- Was it easy or difficult to understand?
    - Why do you say that?

Theory-driven probes (motivation):

- **External motivations**
    - After sections with potential benefits – what are your thoughts on that section?
    - Workbook activity (1A) – what are your thoughts on that activity?
    - Discussion board activities – what do you think about these discussion board activities?

- **Autonomous motivation**
    - After section on symptoms – if you made changes to your diet and noticed some of your symptoms improve, how would you feel about those changes that you had made?

After the module is completed:

- What are your overall thoughts on this module?
- What thoughts did you have about the design and style (amount of text, videos, etc)
- Based on this module, how likely are you to do the next 5 modules of the program?
- Would you be more or less likely to make changes to your current eating habits after completing this module or program? **(theory-driven probe – external motivation)**

Wrap up: Thank you for taking the time to answer these questions. Please feel free to share any other comments that you haven't shared at this point.

Appendix H: Higher resolution images from Chapter 7 figure 2

[images and weblink redacted]