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GERMAN TRANSLATION, CULTURAL ADAPTION AND VALIDATION OF THE UNIDIMENSIONAL SELF-EFFICACY SCALE FOR MULTIPLE SCLEROSIS: STUDY PROTOCOL

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GERMAN TRANSLATION, CULTURAL ADAPTION AND VALIDATION OF THE UNIDIMENSIONAL SELF-EFFICACY SCALE FOR MULTIPLE SCLEROSIS: STUDY PROTOCOL

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ABSTRACT

Introduction: Self-efficacy refers to individuals' confidence in their ability to perform relevant tasks to accomplish desired goals. This is independent of their actual abilities. In people with multiple sclerosis (PwMS), self-efficacy has been shown to powerfully influence motivation and health-related behaviour, such as adherence to prescribed treatment or physical activity. So far, a rigorously tested German language self-efficacy questionnaire for PwMS has not been found.

Methods: Based on Bandura's concept of self-efficacy and international guidelines for questionnaire development, this study will translate the original Unidimensional Self-Efficacy Scale for Multiple Sclerosis (USE-MS) into German. The patient-led development of the pre-final German version will involve a forward-backward translation process, synthesis of translations, expert committee review and consensus with the original test developers. At two centres in the region of Tyrol, Austria, content and face validity and cultural adaption for Austria will be established using face-to-face semi-structured cognitive interviews of 30 PwMS. A further 292 PwMS with minimal to severe disability will be tested at two time-points to validate the German USE-MS (USE-MS-G).

Analysis: Mixed methods analyses will be applied. Interviews will be transcribed and analysed employing qualitative content analysis. External validity will be explored using Spearman's Rank correlation coefficients of the USE-MS-G with the 13-item Resilience Scale, General Self-Efficacy Scale, Multiple Sclerosis International Quality of Life questionnaire, Hospital Anxiety and Depression Scale and MS-specific Neurological Fatigue Index. Test-retest reliability, internal consistency and floor and ceiling effects will be evaluated. Internal validity will be examined using Rasch analysis.

Ethics and dissemination: Ethical approval was received from the Ethics Committee of the Medical University of Innsbruck, Austria (reference number EK1260/2018; 13.12.2018). Results from this study will be disseminated to the participants via mail and MS Societies (Austria, Germany, Switzerland and UK), and to clinicians and researchers through peer-reviewed publications and conferences.

Study registration: ISRCTN Registry; trial ID ISRCTN14843579; prospectively registered on 02. 01. 2019; http://www.isrctn.com/ISRCTN14843579

Study protocol, version 1, 31.1.2019

Keywords: multiple sclerosis; self-efficacy; Unidimensional Self-efficacy Scale for Multiple Sclerosis; German version; cross-cultural adaption for Austria; Rasch analysis

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ARTICLE SUMMARY

Strengths and limitations of this study

- This study protocol describes the German translation of the original English language Unidimensional Self-efficacy Scale for Multiple Sclerosis (USE-MS), upon permission of the scale developers and applying international recommendations.

- Consistent with the conceptual framework of the English USE-MS, Bandura's concept of self-efficacy will be adhered to.

- Employing a patient-led process in phase 1, 30 people with MS (PwMS) will be interviewed about the pre-final German USE-MS, to establish face and content validity and cultural adaption for PwMS in Austria.

- In phase 2, the German USE-MS will be validated in a larger sample of 292 PwMS.

- Applying classical test theory and Rasch analysis approaches, internal and external validity, internal consistency and test retest reliability will be explored.

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INTRODUCTION

Multiple sclerosis (MS) is one of the most common neurological diseases in young adults worldwide, with increasing prevalence[1]. MS is characterised by a wide variety of symptoms and different disease courses[2]. Despite the development of novel disease modifying drugs and neurorehabilitation strategies, the unpredictability of the disease with psychological distress, losses in social contact and quality of life (QoL) are concerning for PwMS. However, individuals' self-knowledge can modulate their approach to day-to-day activities. According to Bandura's social cognitive theory, psychosocial functioning is regulated by reciprocal interactions between behaviour, personal factors and environmental conditions[3]. Self-regulation and intrinsic motivation enable individuals to set and pursue their own goals, observe and evaluate themselves in relation to attained goals[4]. Bandura defined self-efficacy as individuals' beliefs regarding their capability to perform significant tasks, to achieve goals that are meaningful for their daily lives[3]. Self-efficacy beliefs considerably influence people's feelings, thoughts and motivation[5] while, notably, being independent of their physical performance[5]. Such a concept appears important for people with disabilities because it may shape their motivation to initiate and adhere to treatment, particularly when facing side effects.

Perceived self-efficacy influences health-related behaviour such as adhering to medication[6] or engaging in physical activity in PwMS[7]. Health status evaluations of responses to rehabilitation and steroid treatment after an MS relapse can be predicted by self-efficacy levels[8]. Also, higher self-efficacy levels are associated with better long-term perceived cognitive functioning[9] and QoL[10, 11]. PwMS who report higher perceived self-efficacy also state lower levels of fatigue, depression and anxiety[12]. Recent evidence has provided insight into the importance of self-management and intrinsic motivation for motor learning[13]. Recognising the relevance of self-efficacy especially for people with disabilities, valid and reliable measurement tools are still needed for its assessment. Three generic self-efficacy scales were found in the literature[7, 14-16]. However, generic questionnaires may not adequately cover the construct of self-efficacy in a chronic neurological disease like MS. The initial impact of a diagnosis of MS, in addition to the manifold symptoms and necessity of managing a progressive disease may affect individuals' self-efficacy perceptions. Studies

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demonstrated that the capability to effectively solve problems, consistent with higher self-efficacy levels, is strongly associated with PwMS' psychological adaptation to their disability[17], supporting the choice of a disease-specific over a generic self-efficacy questionnaire. MS-specific self-efficacy scales include the Liverpool Self-efficacy Scale (LSES)[18], Multiple Sclerosis Self-Efficacy Scale (MSSS)[19], MS Self-Efficacy Scale (MSSE)[20], Unidimensional Self-Efficacy Scale for Multiple Sclerosis (USE-MS)[21] and University of Washington Self-Efficacy Scale for people with disabilities[22].

Following current guidelines, patients should be involved in the translation and development process of disease specific questionnaires, to ensure the scale reflects their experiences[23]. LSES and MSSS development used in-depth patient interviews while the USE-MS consists of items from both the LSES and MSSS. Bandura's concept of self-efficacy is reflected in the wording of all three questionnaires. The USE-MS study sample was the largest thereof (N=303), and only the USE-MS was exposed to Rasch analysis assessing internal construct validity, in addition to conventional external construct validity and reliability testing. Fit to the Rasch model was demonstrated, and good external validity and reliability[21]. Consequently, the USE-MS appears to be appropriate for use in clinical practice and research. However, so far no validated German language version of the USE-MS is available. The purpose of this study will therefore be to translate the USE-MS into German and validate the German language version in a larger sample of PwMS.

METHODS

Study aims

The first aim of this patient led study is to translate the original English USE-MS, developed by Young et al. (2012) into German, based on international guidelines.

The second aim is to establish face and content validity and cultural adaption of the German version for PwMS in Austria, using individual semi-structured cognitive interviews.

The third aim is to evaluate internal and external validity, internal consistency and testretest reliability of the German USE-MS (USE-MS-G), using classical test theory and Rasch analysis.

Study design

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This will be a bi-centre prospective cross-sectional translation and validation study with repeated measures, consisting of Phase 1 and Phase 2. The SPIRIT 2013 and SPIRIT-PRO Extension checklist for study protocols[24] is presented as Supplementary File 1.

Study setting and timeline

Locations will be the outpatient MS-Clinic of the Clinical Department of Neurology, Medical University of Innsbruck, Austria and Department of Neurology, Clinic for Rehabilitation Münster, Austria.

The expected overall study duration is 33 months, from 01.02.2019 to 31.10. 2021.

Participants and recruitment

A random cross-sectional cohort of patients with clinically definite MS will be recruited from the two centres. The study will be advertised in the MS-Clinic, the Rehabilitation Centre and on the Austrian MS Society website. Further interested PwMS will be examined for eligibility by neurologists at the two study locations. Severely disabled PwMS (EDSS ≥8) will be offered home visits to enable their participation. Written informed consent will be obtained either by neurologists (FD, CB and RE) or the first author (BS). Participants may withdraw from the study at any time and for any reasons without prejudice. Outpatient participants will be reimbursed for travel expenses only. Inclusion and exclusion criteria are listed in Table 1.

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Table 1 Inclusion and exclusion criteria

Inclusion criteria	Exclusion criteria
People with any MS phenotype according to the version of the McDonald's criteria valid at the time of diagnosis	Concomitant diseases which may affect subjective self-efficacy ratings (e.g. malignant diseases, other neurological or psychiatric disorders)
Aged 18 years or over	A relapse of MS within the last two months
Any ethnicity	Any medication change within four weeks prior to the study
Disability status score on the Expanded Disability Status Scale (EDSS) of 0 to 9.0	A relapse between testing 2 and 3 would necessitate the exclusion of the participant.
Able to speak and understand German language, or German language as the first spoken language	

References: McDonald's criteria[25-27]; Expanded Disability Status Scale[28].

Ethics approval, permissions and dissemination plan

Ethical approval for both centres was received from the Ethics Committee of the Medical University of Innsbruck, Austria (reference number EK1260/2018; 13.12.2018). Due to the absence of an intervention, no insurance policy is required for this study and no harm to participants is expected.

Permission to translate into German and validate the original USE-MS[21] was provided by the test developers who hold the copyright for the USE-MS-G.

Results from this study will be disseminated to the participants via mail and MS Societies (Austria, UK). Findings will be disseminated to clinicians and researchers through peer-reviewed publications and conferences.

Patient and public involvement

In Phase 1, patients will be lay members of the expert committee to consolidate all the translations and back translations of the USE-MS. Their role regarding the item and response option wording and sentence structure will be crucial, as the final questionnaire should be understood by PwMS. Patients will also be involved using face-to-face cognitive interviewing, to gain insight into their views about the clarity of the wording, meaning and completeness of the questions of the pre-final USE-MS-G. The Austrian MS (recruitment) and MS Research Societies (funding) will be involved in this study, with whom the findings will be shared as soon as available (patient magazine, meetings). The findings will also be disseminated to the UK MS Society and MS Trust.

Sample size

Phase 1

Using a power of 80% (β =0.2) and a 5% type I error rate, a sample size of 27 participants is necessary to detect a problem within a questionnaire, which occurs at a prevalence of 0.06% (1 in 1666.67)[29]. Thus, including a 10-12% attrition rate, 30 participants will be required. As recommended for qualitative interviewing, it is expected that 30 participants will be sufficient to reach saturation[30].

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Phase 2

Rasch analysis sample size requirements are predicated upon the degree of precision required for estimating item and person difficulties. Regardless of targeting, one can be 99% confident that a sample size of 243 participants is adequately large to obtain a (high) precision of ± 0.5 log odd units (logits). Good targeting provided, a sample size of 108 people would be sufficient[21, 31]. Hence, including a 20% attrition rate, 292 participants will be aimed at in this study.

Outcomes and data collection

Assessments used in this study were developed using patient involvement and/or recommended by governmental or patient organisations (Supplementary File 2). Study outcomes and methods for their assessment are presented in Figure 1. Participant characteristics and assessments used at all time-points are shown in Table 2.

Figure 1 around here

Figure 1 Study outcomes and their assessment

Figure legend: GSE: General Self-Efficacy Scale; RS-13: Resilience Scale, short version; MusiQol: Multiple Sclerosis International Quality of Life questionnaire; HADS: Hospital Anxiety and Depression Scale; NFI-MS: Neurological Fatigue Index.

Table 2 Participant characteristics and assessments used in this study

Participant characteristics and assessments	Phase 1	Phase 2	
(assessments will be collected in a random order to	T1	T2	T3
avoid order effect)			
Participant identifier (ID)	X	X	X
Age	X	X	
Gender	X	X	
MS phenotype ¹	X	X	
Disease duration	X	X	
Expanded Disability Status Scale (EDSS) ²	X	X	
Disease modifying treatment (DMT) ³	X	X	X
(Pre-final) German version of Unidimensional	X	X	X
Self-Efficacy Scale for Multiple Sclerosis			
Qualitative cognitive interview	X		
Resilience Scale, short version		X	X
General Self-Efficacy Scale	0	X	X
Multiple Sclerosis International Quality of Life	5	X	X
questionnaire			
Hospital Anxiety and Depression Scale	5	X	X
Neurological Fatigue Index		X	X

¹Relapsing-remitting; primary progressive; secondary progressive multiple sclerosis[32]

²EDSS groups: 0-4.0; 4.5-6.5; 7.0-7.5; 8.0-9.0[28]

³(a) No DMTs; (b) low effective DMTs: interferon-b 1a, interferon-b 1a, interferon-b 1b, pegylated interferon-b 1a, glatiramer acetate, dimethyl fumarate, teriflunomide, azathioprin, intravenous immunoglobulins; (c) high effective DMTs: alemtuzumab, cladribine, fingolimod, natalizumab, ocrelizumab, cyclophosphamide, mitoxantrone,

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rituximab[33, 34]

At recruitment, disability will be assessed by neurologists (FD, CB or RE) using the EDSS, ranging from 0 to 10, with higher scores representing higher levels of disability[28]. Although psychometric validation studies criticised its low responsiveness to changes, the EDSS has no floor or ceiling effects[35], has been shown to be valid and reliable[36] and is therefore recommended for use in clinical studies[37].

Excellent internal and external validity and reliability of the original USE-MS has been shown[21]. Scoring of the USE-MS draws results from all 12 items while items 7, 8, 10, 11 and 12 are inversely scored. Higher numbers represent stronger self-efficacy beliefs in participants[21]. The USE-MS includes a 4-point Likert scale (1= strongly disagree to 4=strongly agree).

To assess external construct validity, the following questionnaires will be administered:

The validated German version[38] of the 10-item General Self-Efficacy Scale (GSE)[15] is a self-administered 4-point Likert scale with a summary score ranging from "not at all true" to "exactly true". The total GSE score ranges between 10 and 40, higher scores signifying greater self-efficacy. Psychometric testing demonstrated high internal consistency, moderate concurrent validity and unidimensionality[15].

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The validated German version[39] of the 13-item Resilience Scale (RS-13)[40], based on the 25-item Resilience Scale[41] will be used. RS-13 item scores from a 7-point Likert scale are added up, indicating low (13-66 points), moderate (67-72 points) or high (73-91 points) resilience[40]. The German RS-13 showed high internal consistency and moderate test-retest reliability. Confirmatory factor analysis indicated an acceptable model fit[40].

The validated German version[42] of the 31-item Multiple Sclerosis International Quality of Life (MusiQol) questionnaire[43] will be employed. Response options use a 6-point Likert scale, from 1= "never/not at all" to 5= "always/very much" and 6= "not applicable". Negatively worded item scores are reversed, and for each participant mean scores for each dimension of the item scores are calculated. All nine dimension scores are linearly transformed to a 0-100 scale, their mean representing the global index score, 0 indicating the worst level of health-related QoL and 100 the best. Psychometric testing

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showed satisfactory internal and external validity and acceptable reliability for all MusiQol dimensions[43].

The validated German version[44] of the 14-item Hospital Anxiety Depression Scale (HADS)[45] will be used. The HADS is a self-report questionnaire with a 4-point Likert scale and a 42 point maximum, higher scores representing higher levels of anxiety or depression. Odd items are added to score the anxiety subscale (0-21 points), and even items are added to generate the depression subscale (0-21 points). Testing of the German version demonstrated good internal consistency and acceptable test-retest reliability [44]. The two-factor structure of the scale was confirmed[44].

The validated German version[46] of the 23-item Neurological Fatigue Index (NFI-MS) will be used[47]. Four factors of the NFI-MS were confirmed by principal component analysis and explained 62% of the variance. The four subscales and total scale showed acceptable responsiveness[48], good test-retest reliability, moderate convergent validity and fit to Rasch model expectations[47]. Items are scored on a 4-point Likert scale from 0= "strongly disagree" to 3= "strongly agree". For scoring, the following item values are added: 1-8= "physical subscale"; 9-12= "cognitive subscale"; 13-18= "relief by diurnal sleep or rest subscale"; 19-23= "abnormal nocturnal sleep and sleepiness subscale"; and 1-7, 9, 11-12 = "physical and cognitive summary score"[47].

Assessments will be performed by trained physiotherapists holding a Master's (SK) and PhD degree (BS) and a clinical neuropsychologist (LZ). The number of participants who decline to participate or drop out will be recorded, together with reasons (CONSORT flow chart). Any health problems will be recorded.

Phase 1: data will be collected at one time-point (Testing 1, T1), with an expected duration of 45-60 minutes.

Phase 2: for the test-retest reliability assessment, data will be collected at two timepoints and will last 60-90 minutes: Testing 2 (T2) and Testing 3 (T3), 14-21 days after T2[47, 49].

Data management

With regard to confidentiality, the Austrian and Tyrolean Data Protection Acts will be adhered to. Double data entry and range checks for data values will be used. For qualitative content analysis, double coding of the data set will be performed. Only the 12

research team will have access to the data. All personal data will be anonymised by a participant ID. Data and files will be saved on a password protected computer, will not be transferred via emails and will be only used for the purposes for which they were collected. Participants will be informed about their right to disclosure for their own data even if these data lack clinical utility. Anonymised data will be kept for 15 years following completion of the study. Blank data collection forms can be requested from the corresponding author.

Study procedures

This study will follow the Beaton et al. guidelines for the cross-cultural adaptation of patient reported outcomes[50] and its enhanced version from the University of Leeds, UK.

Phase 1

Stage 1: Forward translation of the items, response options, instructions and scoring information into German will be performed by three independent translators; translator 1 is a medical professional and informed about self-efficacy, while translators 2 and 3 have no medical knowledge and are "naïve" to self-efficacy. Translators are bilingual German native speakers and will create a written report for all translations (T1, T2 and T3), which will then be compared, to distinguish any wording differences or ambiguities[51].

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Stage 2 will be a synthesis of T1-3 into T-123. Involving a fourth, unbiased person, the three versions will be discussed with the translators and any discrepancies solved by consensus. A revised questionnaire and comprehensive report will be produced[50].

Stage 3: Backward translation of T-123 into English will be done by three bilingual English native speakers who are blind to the original version. Translators are "naïve" to self-efficacy and medicine, to minimise bias[51]. Vague wording, obvious inconsistencies or theoretical errors in the translations shall be detected. A report for each version, TB1, TB2 and TB3, will be written by the translators. To maximise comprehension, language will be used which can be understood by a 12 year old[52, 53], indicated by a Flesch reading-ease score of 80-90[54]. The German Flesch value=180-ASL-(58,5*ASW), where ASL=average sentence length and ASW=average number of syllables per word[54].

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Stage 4: Considering written documentations, an Expert Committee will review and integrate all versions of the questionnaire, involving instructions and scoring documentation, and develop the pre-final version of the USE-MS-G. The Expert Committee will consist of three neurologists, two physiotherapists, a neuropsychologist, a methodologist, two language professionals, the translators, three lay PwMS and the translation synthesis recorder. The Expert Committee will be in close contact with the original USE-MS developers. A written report of the consensus process will be created. Decision-making will be based on guidelines to accomplish cross-cultural equivalence between the original and German versions in four areas[51], shown in Figure 2.

Figure 2 around here

Figure 2 Cross-cultural equivalence areas to be achieved between original and German USE-MS (USE-MS-G); adapted from[51]

Stage 5: Pretesting of the pre-final USE-MS-G will be performed in 30 PwMS, involving completion of the scale and face-to-face cognitive interviews. Cognitive interviewing will be used to evaluate whether survey questions are easily comprehended, response categories match natural responses, and if people are motivated to respond truthfully and accurately [55-57]. Leading questions will be avoided to minimise bias. Enquiries for comprehension and meaning will be used, and repetition of content by patients [55, 57]. Probing will be applied to explore cognitive processes such as memory, underlying reasons for certain responses and overall level of difficulty or confidence[56]. Verbal probes, following Willis' model, will be used immediately after the guestions[58]: (a) standardised, anticipated probes: scripted; (b) standardised, conditional probes: scripted, but will be used only if activated by certain participant behaviors such as hesitation[59]; (c) non-standardised, spontaneous probes: flexible, at researcher's digression: and (d) non-standardised, emergent probes: applied in reaction to participant behaviour[60]. The interview guide is presented in Table 3. Recording and field notes will be used, reviewed for inconsistencies or gaps shortly before the end of the interview.

Table 3 Questions used for semi-structured interview

Parti MS.	Participants will be given sufficient time to complete the pre-final German USE- MS.					
1.	Having read the questions in the questionnaire, what are your thoughts about them?					
2.	Would you please repeat this question in your own words?					
3.	What do you think this question is asking?					
4.	What do you think about that particular question?					
5.	What do you think about the wording of this question, in terms of its clarity?					
6.	How easy or hard was this to answer?					
7.	How sure are you of your answer?					
8.	Could you talk me through your answers in more detail?					
9.	What were you thinking of when you answered this question?					
10.	Do you have any other comments?					
11.	If responses from participants are somewhat unclear, the interviewer asks: "Why so?"					
12.	Should a participant hesitate, the interviewer conveys: "You spent some time answering that question - what were you thinking about?"					
Adap	Adapted from [56] and [61]					

An overview of study procedures is presented in Figure 3.

Figure 3 around here

Figure 3 Flowchart of the study procedures

Figure legend: MS: multiple sclerosis; T1 (2; 3): testing 1 (2; 3).

Phase 2

 The USE-MS-G will be validated in a larger sample of 292 PwMS who will complete the above described questionnaires at T1 and T2.

Data analyses

Mixed methods data analyses will be used.

Phase 1- Qualitative analyses

Interviews will be transcribed and analysed using qualitative content analysis (QCA)[62, 63] using QDA MINER LITE software (Provalis Research, Montreal, Canada) and adhering to the Consolidated Criteria for Reporting Qualitative Research (COREQ)[64]. Analysis steps will be performed as follows[65-68]:

- Data organisation based on the research question
- Identification of recurring ideas, concepts, themes and words
- Development of a coding frame (requirements: unidimensionality, mutual exclusivity of subcategories within dimensions, exhaustiveness of subcategories and saturation, where each subcategory is used at least once)
- Selection of relevant material, structuring, marking and segmentation of text sections, based on Bandura's concept of self-efficacy and the original USE-MS, to identify main and subcategories
- Definition, naming and characterisation of categories and decision rules, to enable consistent assignment of data segments
- Illustration of categories and subcategories using citations
- Creation of a data matrix, followed by quantitative data analysis (descriptive statistics, e.g. frequencies)
- Report
- Rigor and credibility will be maximised by[69-71]

- systematic and consistent approach throughout the analysis
- revision and expansion of the coding frame
 double coding of the whole dataset by two independent researchers (10-14 days after initial coding)
- checking for researcher effects (reflexivity)[72]
Phase 2 - Quantitative analyses
Descriptive statistics and reliability estimates will be performed using IBM SPSS software, release 25.0 (IBM Corporation, Armonk, NY, USA). Rasch Analysis will be conducted with RUMM2030 software[73]. Statistical significance is defined as two-tailed p value <0.05.
Missing data will be treated as follows:
 Missing data should be avoided by checking questionnaires for missing item responses and asking participants for completion.
 Rasch analysis calculates an estimate from all available data and does not require a complete data set[74].
Test-retest reliability
Test-retest reliability will be evaluated using Lin's concordance correlation coefficient (r_c) between T2 and T3 (0-1)[75, 76]. R_c s will be calculated with their 95% confidence intervals (CI). Values of <0 will be considered to indicate poor, 0–0.20 slight, 0.21–0.40 fair, 0.41–0.60 moderate, 0.61–0.80 substantial and 0.81–1 almost perfect agreement[77].
External validity
It is hypothesised that scores on the German USE-MS will demonstrate moderate to high positive correlations with scales assessing conceptually similar constructs (convergent validity; with the GSE, RS-13 and MusiQol) and moderate to high negative correlations with scales measuring divergent constructs (divergent validity; with the HADS and NFI-MS); Spearman's Rank correlation coefficients of 0.3-0.49 being
Internal validity - Rasch analysis

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Rasch analysis[79] assumes the probability of a person endorsing an item is a logistic function of the difference between the "person ability" (perceived self-efficacy) and the "item difficulty" (level of self-efficacy) expressed[80]. Item characteristic curves, arranged on the log-odds units (logit) scale, will be used to visualise the probability of a person's correct response in relation to the item difficulty[81].

The polytomous Rasch model will be chosen for this study, suitable for scales with multiple response categories for their items[82]. A significant likelihood ratio test signifying inconsistent distance between response category thresholds would require the use of Masters' unrestricted (partial credit) model[83], otherwise Andrich's rating scale model[84]. Category thresholds are located centrally between two adjacent categories where either response is equally likely[82, 85]. The 4-point USE-MS includes three thresholds.

Ordered item category thresholds

Category probability curves will be inspected, checking regular distribution and monotonic advance of measures across categories[85].

Targeting

Targeting refers to the degree to which the scale captures the full range of self-efficacy. Inspecting person-item threshold distribution maps, the mean location score for the respondents will be compared with the default items zero value. A well-targeted scale is centred around zero logits (±0.5 logits), corresponding to the scale's item of mean difficulty[86].

The proportion of floor and ceiling effects will be monitored, considered noteworthy if >5%[87].

Local independence

Local independence means there should be no associations between the items. Inspection of the correlation matrix of item standardised residuals should show Pearson's correlations of <0.2 above the mean value of the matrix as a whole.

Unidimensionality

Unidimensionality as a Rasch model requirement allows a summary score measurement of a single construct. Using a PCA of the residuals, positively and 18

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negatively loadings of the first component will be identified, generating two subsets and separate person estimates. Independent t-tests will explore significant differences[88]. If less than 5% of t-tests are significant or the lower bound of the binominal CI overlaps 5%, unidimensionality is supported[89, 90].

Fit to the Rasch model

Different fit statistics will seek to determine if the assumption of a probabilistic ordering of items is satisfied:

(a) Summary Chi-square interaction statistics and individual item Chi-square statistics are expected to be non-significant (Bonferroni-adjusted p-values for the number of items)[47].

(b) Individual person and item fit residuals are expected to be between ±2.5 (99% CI)[91].

(c) Person and summary item fit residuals reflect perfect model fit if their mean and standard deviation are close to 0 and 1, respectively[92, 93].

Reliability

Reliability is indicated by the person separation index (PSI; range 0-1)[94] and Cronbach's alpha (missing data excluded), which should be ≥ 0.85 for individual use or 0.70 for group use[47, 95]. Erasmushogeschool . Protected by copyright, including for uses related to text and data mining, Al training, and similar technologies.

Invariance and differential item functioning (DIF)

Invariance means that all persons completing a questionnaire, regardless of their ability (or self-efficacy), recognise the difficulty in identical items[94]. Any likelihood of differently scored items between the groups violates the assumption of invariance, called DIF[96, 97]. Absence of DIF will be tested in gender (female; male), age (quartile groups) and disease duration (quartile groups) and indicated by a non-significant ANOVA of the residuals (5% alpha with Bonferroni correction) where the group is the main factor[97, 98]. Any observed DIF will be examined to know whether it cancels out at the test level[96].

If model fit is not achieved, an iterative stepwise procedure will be initiated, involving

strategies for combining response categories, stepwise deletion of the worst fitting item, testlet (superitem) construction and adjusting for DIF as appropriate[99].

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Author contributions

All authors critically and substantially revised the manuscript and approved the current version to be submitted for publication. BS devised and designed the study and drafted the manuscript. RK developed the qualitative data analysis plan. LZ substantially contributed to the conception and design of the study. MR provided input on the study methodology and quantitative analysis. RM provided relevant advice on the Rasch analysis. SK substantially contributed to the development of the study protocol. FD is a study manager at his centre and substantially contributed to the development of the study protocol. CB is a study manager at his centre and substantially contributed to the design of the study protocol. RE is a study manager at his centre and substantially contributed to the design of the study protocol. RE is a study manager at his centre and substantially contributed to the design of the study protocol. RE is a study manager at his centre and substantially contributed to the design of the study protocol.

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Competing Interests

None declared.

Data sharing statement

Data generated by this research that support any publications will be made available upon reasonable request as soon as possible. In addition, meaningful data from this research will be available online as a "scientific use file", wherever legally and ethically possible.

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Figure 1

254x190mm (96 x 96 DPI)



Semantic

Meaning of words including subtle meaning differences of words within context

Grammar and sentence structure should be considered

Idiomatic

Idioms and colloquialisms are a type of informal language

They cannot be translated literally

Equivalent expressions or item replacement with comparable meaning need to be found

EQUIVALENCE

Experiential

Situations and daily life activities described by the translated questionnaire should match Austrian cultural context

Conceptual

The concept of self-efficacy and events described by the questionnaire items may be differently perceived and experienced by people with MS in different countries or cultures.

For example, the concept of family, friends, fatigue or the condition of MS itself

Figure 2

254x190mm (96 x 96 DPI)

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Contact and

consensus with the original test

developers from

The Walton Centre for Neurology and

Forward translation into German

Synthesis of forward translations





Figure 3

190x254mm (96 x 96 DPI)



Calvert M, Kyte D, Mercieca-Bebber R, et al. Guidelines for Inclusion of Patient-Reported Outcomes in (a) Billing Hard Protocols: The SPIRIT-PRO Extension. JAMA : the journal of the American Medical Association 2018;319(5):483-94 doi: 10.1001/jana 2018;21903[published Online First: Epub Date])

Section/item	ItemNo	Description	SPIRIT-PRO Item No.	SPIRIT-PRO Extension or a solution of the second se		Addressed on Page No.
Administrative in	formation			hining	om h	
Title	1	Descriptive title identifying the study design, population, interventions, and, if applicable, trial acronym	er	, Al training,	:tp://bmjopen	1
Trial registration	2a	Trial identifier and registry name. If not yet registered, name of intended registry		and similar t	bmj.com/ o	2
	2b	All items from the World Health Organization Trial Registration Data Set		echnolog	n June 10	2; 22
Protocol version	3	Date and version identifier		le. s.	, 2025	2
Funding	4	Sources and types of financial, material, and other support			at Depar	22
					tment GEZ-L1	<u>.</u>

			BMJ Open	/bmjopen-201; by copyright,	
Roles and responsibilities	5a	Names, affiliations, and roles of protocol contributors		including f	1; 22
	5b	Name and contact information for the trial sponsor	SPIRIT-5a- PRO Elaboration	Specify the individual(s) responsible for the PRO content of the trial potocol.	BS
	5c	Role of study sponsor and funders, if any, in study design; collection, management, analysis, and interpretation of data; writing of the report; and the decision to submit the report for publication, including whether they will have ultimate authority over any of these activities	to.	2019. Downloaded from http:// smushogeschool . ed to text and data mining, Al t	8
	5d	Composition, roles, and responsibilities of the coordinating centre, steering committee, endpoint adjudication committee, data management team, and other individuals or groups overseeing the trial, if applicable (see Item 21a for data monitoring committee)		bmjopen.bmj.com/ on June '	7; 13; 14; 22
ntroduction				ogies,	
Background and rationale	6a	Description of research question and justification for undertaking the trial, including summary of relevant studies (published and unpublished) examining benefits and harms for each intervention	SPIRIT-6a- PRO Extension	Describe the PRO-specific research question and rationale for PRO assessment and summarize RRO findings in relevant studies.	5-6

	6b	Explanation for choice of comparators		ncluding for	11-13; Supplem File 2
Objectives	7	Specific objectives or hypotheses	SPIRIT-7- PRO Extension	State specific PRO objectives or hypotheses (including relevant PRO concepts/domains).	6
Trial design	8	Description of trial design including type of trial (eg, parallel group, crossover, factorial, single group), allocation ratio, and framework (eg, superiority, equivalence, noninferiority, exploratory)		19. Downloaded from h nushogeschool . to text and data mining	6-7
Methods: Partici	pants, in	nterventions, and outcomes		,, Al	
Study setting	9	Description of study settings (eg, community clinic, academic hospital) and list of countries where data will be collected. Reference to where list of study sites can be obtained		bmjopen.bmj.com/ training, and simila	7
Eligibility criteria	10	Inclusion and exclusion criteria for participants. If applicable, eligibility criteria for study centres and individuals who will perform the interventions (eg, surgeons, psychotherapists)	SPIRIT-10- PRO Extension	Specify any PRO-specific eligibility criteria (eg, language/reading requirements or prerandonization completion of PRO). If PROS will not be collected from the entire study sample, provide a rationale and describe the method for obtaining the PROS subsample.	7-8

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Interventions	11a	Interventions for each group with sufficient detail to allow replication, including how and when they will be administered		9-029565 on 20 Au	NA
	11b	Criteria for discontinuing or modifying allocated interventions for a given trial participant (eg, drug dose change in response to harms, participant request, or improving/worsening disease)		igust 2019. Downloa Erasmushogesch related to text and	7-8 (concerning assessments) Interventions: NA
	11c	Strategies to improve adherence to intervention protocols, and any procedures for monitoring adherence (eg, drug tablet return, laboratory tests)		aded from http:// lool . data mining, Al t	7; 13
	11d	Relevant concomitant care and interventions that are permitted or prohibited during the trial		bmjopen.bm raining, and	7-8
Outcomes	12	Primary, secondary, and other outcomes, including the specific measurement variable (eg, systolic blood pressure), analysis metric (eg, change from baseline, final value, time to event), method of aggregation (eg, median, proportion), and time point for each outcome. Explanation of the clinical relevance of chosen efficacy and harm outcomes is strongly recommended	SPIRIT-12- PRO Extension	Specify the PRO concepted domains used to evaluate the intervention (a), overall health-related quality of life, specific domain, specific sympton and, for each one, the analysis metric (a), anage from baseline, final value, time to a the principal time point or period of interest.	9-13

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Page 33 of 43 1 2 3 4 5 6 7 8 9 10 11 12 13 14 15 16 17 18 19 20 21 22 23 24 25 26 27 28 29 30 31 32 24 25 26 27 28 29 30 31 32 33 34 35 36 37 38 39
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Participant timeline	13	Time schedule of enrolment, interventions (including any run-ins and washouts), assessments, and visits for participants. A schematic diagram is highly recommended (see Figure)	SPIRIT-13- PRO Extension	Include a schedule of PR assessments, providing a rationale for the time points, and justifying if the initial assessment is not prerandomization. Specify time windows, whether PRO collection is prior to clinical assessments, and the using multiple questionnaires, whether order of administration will be stard a beta beta beta
Sample size	14	Estimated number of participants needed to achieve study objectives and how it was determined, including clinical and statistical assumptions supporting any sample size calculations	SPIRIT-14- PRO Extension	When a PRO is the primary and point, state the required samples be (and how it was determined) and recruiting it for expected biss to follow- up). If sample size is not established based on the PRO end point, then discuss the power of the bring of PRO analyses.
Recruitment	15	Strategies for achieving adequate participant enrolment to reach target sample size		and similar te
Methods: Assig	jnment of i	nterventions (for controlled trials)		- June
Allocation:				logie NA

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Sequence generation	16a	Method of generating the allocation sequence (eg, computer-generated random numbers), and list of any factors for stratification. To reduce predictability of a random sequence, details of any planned restriction (eg, blocking) should be provided in a separate document that is unavailable to those who enrol participants or assign interventions		19-029565 on 20 August 2019. Downloac Erasmushogescho , including for uses related to text and d	NA
Allocation concealment mechanism	16b	Mechanism of implementing the allocation sequence (eg, central telephone; sequentially numbered, opaque, sealed envelopes), describing any steps to conceal the sequence until interventions are assigned		ded from http://bmjoper ool . ata mining, Al training,	NA
Implementation	16c	Who will generate the allocation sequence, who will enrol participants, and who will assign participants to interventions		ı.bmj.com/ on Ju and similar tech	NA
3linding (masking)	17a	Who will be blinded after assignment to interventions (eg, trial participants, care providers, outcome assessors, data analysts), and how		ine 10, 2025 at De nologies.	NA
			BMJ Open	omjopen-2019 y copyright, i	
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	17b	If blinded, circumstances under which unblinding is permissible, and procedure for revealing a participant's allocated intervention during the trial		-029565 on 20 Aug ncluding for uses r	NA
Methods: Data	collection,	management, and analysis		elate	
Data collection methods	18a	Plans for assessment and collection of outcome, baseline, and other trial data, including any related processes to promote data quality (eg, duplicate measurements, training of assessors) and a description of study instruments (eg, questionnaires, laboratory tests) along with their reliability and validity, if known. Reference to where data collection forms can be found, if not in the protocol	SPIRIT-18a (i)-PRO Extension	Justify the PRO instrume to be used and describe domains, not be used and describe domains, not be used indicating a good or poor but the measurement properties and patient action guidelines, and patient action guidelines, and patient action guidelines, and patient action guidelines, ideally in the period of interest. State whether the measure will be used in accordance with any user manual and specify and justify deviations if planned.	9-13
			SPIRIT-18a (ii)-PRO Extension	Include a data collection and setting (eg, clinic, home, bither).	7-8; 15
			SPIRIT-18a (iii)-PRO Extension	Specify whether more than 1 panguage version will be used and state whether translated versions have beer developed using currently recommended methods.	Total manuscript

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			SPIRIT-18a (iv)-PRO Extension	When the trial context requires someone other than a trial participant to answer on his or her behalf (a proxy reported outcome), state and justify the use of a proxy respondent. Provide or the evidence of the validity of the use assessment if available.	NA
	18b	Plans to promote participant retention and complete follow-up, including list of any outcome data to be collected for participants who discontinue or deviate from intervention protocols	SPIRIT-18b (i)-PRO Extension	Specify PRO data collect and management strategies for management avoidable missing data. at a mining mining mining avoidable missing data.	19
			SPIRIT-18b (ii)-PRO Elaboration	Describe the process of PPRO assessment for participants who discontinue or deviate from the assigned intervention protocol.	19
Data management	19	Plans for data entry, coding, security, and storage, including any related processes to promote data quality (eg, double data entry; range checks for data values). Reference to where details of data management procedures can be found, if not in the protocol		mj.com/ on June 10, 2025 a d similar technologies.	13-14; 18-19; 23
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Statistical methods	20a	Statistical methods for analysing primary and secondary outcomes. Reference to where other details of the statistical analysis plan can be found, if not in the protocol	SPIRIT- 20a- PRO Elaboration	si φ State PRO analysis metheds, Signcluding any plans for addressing multiplicity/type I (α) error. signal g Au g Au	18-22
	20b	Methods for any additional analyses (eg, subgroup and adjusted analyses)		Erasmush lated to te	20-22
	20c	Definition of analysis population relating to protocol non-adherence (eg, as randomised analysis), and any statistical methods to handle missing data (eg, multiple imputation)	SPIRIT- 20c- PRO Elaboration	State how missing data will be described and outline the methods to be described missing items or entire as the methods to be described approach to imputation and sonsitivity analyses).	19
Methods: Monit	oring			Al tra	
Data monitoring	21a	Composition of data monitoring committee (DMC); summary of its role and reporting structure; statement of whether it is independent from the sponsor and competing interests; and reference to where further details about its charter can be found, if not in the protocol. Alternatively, an explanation of why a DMC is not needed		njopen.bmj.com/ on June 10, 2025 aining, and similar technologies.	13; 17-18; 2

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	21b	Description of any interim analyses and stopping guidelines, including who will have access to these interim results and make the final decision to terminate the trial			-029565 on 20 Augu ncluding for uses rel	13
Harms	22	Plans for collecting, assessing, reporting, and managing solicited and spontaneously reported adverse events and other unintended effects of trial interventions or trial conduct	SPIRIT- 22- PRO Extension	State whether or not PRC monitored during the stud clinical care of individual and, if so, how this will be standardized way. Descr process will be explained eg, in the participant info and consent form.	will be will be will be will be will be again to participants will be again boo again boo	13-17
Auditing	23	Frequency and procedures for auditing trial conduct, if any, and whether the process will be independent from investigators and the sponsor			://bmjopen.bmj.c	17
Ethics and disse	mination			0	:om/ c milar	
Research ethics approval	24	Plans for seeking research ethics committee/institutional review board (REC/IRB) approval			on June 10, 2 technologies	8
		For peer review only - http://b	mjopen.bmi.cor	n/site/about/guidelines.xhtm)25 at Department GEZ-LTA	

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Protocol amendments	25	Plans for communicating important protocol modifications (eg, changes to eligibility criteria, outcomes, analyses) to relevant parties (eg, investigators, REC/IRBs, trial participants, trial registries, journals, regulators)		g	9-029565 on 20 August 20 Eras	NA
Consent or assent	26a	Who will obtain informed consent or assent from potential trial participants or authorised surrogates, and how (see Item 32)			019. Downloaded mushogeschool	7
	26b	Additional consent provisions for collection and use of participant data and biological specimens in ancillary studies, if applicable	revi	g	I from http://bmj	NA
Confidentiality	27	How personal information about potential and enrolled participants will be collected, shared, and maintained in order to protect confidentiality before, during, and after the trial		24001/	open.bmj.com/ on J	8; 12-13
Declaration of interests	28	Financial and other competing interests for principal investigators for the overall trial and each study site			une 10, 2025	22
Access to data	29	Statement of who will have access to the final trial dataset, and disclosure of contractual agreements that limit such access for investigators			at Department	12-13

			BMJ Open		5/bmjopen-20 by copyright,	
Ancillary and post- trial care	30	Provisions, if any, for ancillary and post- trial care, and for compensation to those who suffer harm from trial participation			19-029565 on 20 , including for u	NA
Dissemination policy	31a	Plans for investigators and sponsor to communicate trial results to participants, healthcare professionals, the public, and other relevant groups (eg, via publication, reporting in results databases, or other data sharing arrangements), including any publication restrictions			 August 2019. Downloaded fro Erasmushogeschool . ses related to text and data mi 	8; 23
	31b	Authorship eligibility guidelines and any intended use of professional writers			om http:// ning, Al t	22
	31c	Plans, if any, for granting public access to the full protocol, participant-level dataset, and statistical code		24	bmjopen.bm) raining, and	23
Appendices				06	i.com/ simila	
Informed consent materials	32	Model consent form and other related documentation given to participants and authorised surrogates			on June 10, ; r technologie	23
Biological specimens	33	Plans for collection, laboratory evaluation, and storage of biological specimens for genetic or molecular analysis in the current trial and for future use in ancillary studies, if applicable			2025 at Department s.	NA

BMJ Open Abbreviations: SPIRIT, Standard Protocol Items: Recommendations for Interventional Trials; PRO, patient-reported outcome. *It is strongly recommended that this checklist be read in conjunction with the SPIRIT 2013 Explanation 💈 E boration for important clarification on The strongly recommended that this checklist be read in conjunction with the SPIRIT 2013 Explanation & Egipts the items. Amendments to the protocol should be tracked and dated. The SPIRIT checklist is copyrighted by the Commons "Attribution-NonCommercial-NoDerivs 3.0 Unported" license and is reproduced with permission. The Transmission of the tracked and dated the spirit of the the items. Amendments to the protocol should be tracked and dated. The SPIRIT checklist is copyrighter by the SPIRIT Group under the Creative

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Supplementary File 2

Except good to excellent psychometric properties, the selection of outcome measures depended also on patient involvement during their development, patient acceptability of the tools or recommendations from relevant governmental organisations or MS Societies. The MusiQol was developed using individual semistructured interviews of 1992 PwMS in five countries, followed by qualitative content analysis. The MS-EDGE Outcome Measure Task Force reviewed and recommended the MusiQol for use in PwMS[1]. Similarly, both the German long and short versions of the Resilience Scale are based on face-to-face interviews of 2031 people. The GSE was exposed to repeated validation studies in thousands of participants where excellent validity and reliability were confirmed. A study explored the validity of the HADS in PwMS using interviews by an interviewer who was blind to the HADS scores [2]. The acceptability of the German HADS to various patient populations was tested and confirmed [3]. The UK National Institute for Health and Care Excellence (NICE) 2018 Guidelines for MS[4] recommended the HADS anxiety subscale as a sensitive and specific anxiety screening tool, based on a review[5]. The NFI-MS was developed through a two-stage process in 635 PwMS; guantitative validation and qualitative interviews with patients. The NFI-MS was reviewed and recommended by the MS International Federation (MSIF)[6].

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GERMAN TRANSLATION, CULTURAL ADAPTION AND VALIDATION OF THE UNIDIMENSIONAL SELF-EFFICACY SCALE FOR MULTIPLE SCLEROSIS: STUDY PROTOCOL

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GERMAN TRANSLATION, CULTURAL ADAPTION AND VALIDATION OF THE UNIDIMENSIONAL SELF-EFFICACY SCALE FOR MULTIPLE SCLEROSIS: STUDY PROTOCOL

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Word count: 3960 words excluding declarations; 4206 words including declarations

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ABSTRACT

Introduction: Self-efficacy refers to individuals' confidence in their ability to perform relevant tasks to accomplish desired goals. This is independent of their actual abilities. In people with multiple sclerosis (MS), self-efficacy has been shown to powerfully influence motivation and health-related behaviour, such as adherence to prescribed treatment or physical activity. So far, a rigorously tested German language self-efficacy questionnaire for people with MS is missing.

Methods: The purpose of this study is to translate the original Unidimensional Self-Efficacy Scale for Multiple Sclerosis (USE-MS) into German and to validate the German USE-MS (USE-MS-G). Based on Bandura's concept of self-efficacy and international guidelines for questionnaire development, the patient-led development of the pre-final German version will involve a forward-backward translation process, synthesis of translations, expert committee review and consensus with the original test developers. At two centres in Tyrol, Austria, content and face validity and cultural adaption for Austria will be established using face-to-face semi-structured cognitive interviews of 30 people with MS. A further 292 people with MS with minimal to severe disability will be tested at two time-points to validate the USE-MS-G.

Results: Mixed methods analyses will be applied. Interviews will be transcribed and analysed employing qualitative content analysis. External validity will be explored using Spearman's Rank correlation coefficients of the USE-MS-G with the 13-item Resilience Scale, General Self-Efficacy Scale, Multiple Sclerosis International Quality of Life questionnaire, Hospital Anxiety and Depression Scale and MS-specific Neurological Fatigue Index. Test-retest reliability, internal consistency and floor and ceiling effects will be evaluated. Internal validity will be examined using Rasch analysis.

Ethics and dissemination: Ethical approval was received from the Ethics Committee of the Medical University of Innsbruck, Austria (reference number EK1260/2018; 13.12.2018). Results from this study will be disseminated to the participants and MS Societies, and to clinicians and researchers through peer-reviewed publications and conferences.

Study registration: ISRCTN Registry; trial ID ISRCTN14843579; prospectively registered on 02. 01. 2019; http://www.isrctn.com/ISRCTN14843579

Keywords: Multiple Sclerosis; Self Efficacy; Patient Reported Outcome Austria; Cross-Cultural Comparison; Validation Studies as Topic.	Study pro	tocol, revision 1, 26.6.2019
	Keywords	s: Multiple Sclerosis; Self Efficacy; Patient Reported Outcome ross-Cultural Comparison: Validation Studies as Tonic

ARTICLE SUMMARY

Strengths and limitations of this study

- This study protocol describes the German translation of the original English language Unidimensional Self-efficacy Scale for Multiple Sclerosis (USE-MS), upon permission of the scale developers and applying international recommendations.

- Consistent with the conceptual framework of the English USE-MS, Bandura's concept of self-efficacy will be adhered to.

- Employing a patient-led process in phase 1, 30 people with MS (PwMS) will be interviewed about the pre-final German USE-MS, to establish face and content validity and cultural adaption for PwMS in Austria.

- In phase 2, the German USE-MS will be validated in a larger sample of 292 PwMS.

- Applying classical test theory and Rasch analysis approaches, internal and external validity, internal consistency and test retest reliability will be explored.

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INTRODUCTION

Multiple sclerosis (MS) is one of the most common neurological diseases in young adults worldwide, with increasing prevalence¹. MS is characterised by a wide variety of symptoms and different disease courses². Despite the development of novel disease modifying drugs and neurorehabilitation strategies, the unpredictability of the disease with psychological distress, losses in social contact and quality of life (QoL) are concerning for PwMS. However, individuals' self-knowledge can modulate their approach to day-to-day activities. According to Bandura's social cognitive theory, psychosocial functioning is regulated by reciprocal interactions between behaviour, personal factors and environmental conditions³. Self-regulation and intrinsic motivation enable individuals to set and pursue their own goals, observe and evaluate themselves in relation to attained goals⁴. Bandura defined self-efficacy as individuals' beliefs regarding their capability to perform significant tasks, to achieve goals that are meaningful for their daily lives³. Self-efficacy beliefs considerably influence people's feelings, thoughts and motivation⁵ while, notably, being independent of their physical performance⁵. Such a concept appears important for people with disabilities because it may shape their motivation to initiate and adhere to treatment, particularly when facing side effects.

Perceived self-efficacy influences health-related behaviour such as adhering to medication⁶ or engaging in physical activity in PwMS⁷. Health status evaluations of responses to rehabilitation and steroid treatment after an MS relapse can be predicted by self-efficacy levels⁸. Also, higher self-efficacy levels are associated with better long-term perceived cognitive functioning⁹ and QoL^{10 11}. PwMS who report higher perceived self-efficacy also state lower levels of fatigue, depression and anxiety¹². Recent evidence has provided insight into the importance of self-management and intrinsic motivation for motor learning¹³. Recognising the relevance of self-efficacy especially for people with disabilities, valid and reliable measurement tools are still needed for its assessment. Three generic self-efficacy scales were found in the literature^{7 14-16}. However, generic questionnaires may not adequately cover the construct of self-efficacy in a chronic neurological disease like MS. The initial impact of a diagnosis of MS, in addition to the manifold symptoms and necessity of managing a progressive disease may affect individuals' self-efficacy perceptions. Studies demonstrated that the

capability to effectively solve problems, consistent with higher self-efficacy levels, is strongly associated with PwMS' psychological adaptation to their disability¹⁷, supporting the choice of a disease-specific over a generic self-efficacy questionnaire. MS-specific self-efficacy scales include the Liverpool Self-efficacy Scale (LSES)¹⁸, Multiple Sclerosis Self-Efficacy Scale (MSSS)¹⁹, MS Self-Efficacy Scale (MSSE)²⁰, Unidimensional Self-Efficacy Scale for Multiple Sclerosis (USE-MS)²¹ and University of Washington Self-Efficacy Scale for people with disabilities²².

Following current guidelines, patients should be involved in the translation and development process of disease specific questionnaires, to ensure the scale reflects their experiences²³. LSES and MSSS development used in-depth patient interviews while the USE-MS consists of items from both the LSES and MSSS. Bandura's concept of self-efficacy is reflected in the wording of all three questionnaires. The USE-MS study sample was the largest thereof (N=303), and only the USE-MS was exposed to Rasch²⁴ ²⁵ analysis assessing internal construct validity, in addition to conventional external construct validity and reliability testing. Fit to the Rasch model was demonstrated, and good external validity and reliability²¹. Consequently, the USE-MS appears to be appropriate for use in clinical practice and research. However, so far no validated German language version of the USE-MS is available. The purpose of this study will therefore be to translate the USE-MS into German and validate the German language version in a larger sample of PwMS.

METHODS

Study aims

The first aim of this patient led study is to translate the original English USE-MS, developed by Young et al. (2012) into German, based on international guidelines.

The second aim is to establish face and content validity and cultural adaption of the German version for PwMS in Austria, using individual semi-structured cognitive interviews.

The third aim is to evaluate internal and external validity, internal consistency and testretest reliability of the German USE-MS (USE-MS-G), using classical test theory and Rasch analysis .

Study design

 BMJ Open

This will be a bi-centre prospective cross-sectional translation and validation study with repeated measures, consisting of Phase 1 and Phase 2. The SPIRIT 2013 and SPIRIT-PRO Extension checklist for study protocols²⁶ is presented as Supplementary File 1.

Study setting and timeline

Locations will be the outpatient MS-Clinic of the Clinical Department of Neurology, Medical University of Innsbruck, Austria and Department of Neurology, Clinic for Rehabilitation Münster, Austria.

The expected overall study duration is 33 months, from 01.02.2019 to 31.10. 2021.

Participants and recruitment

A random cross-sectional cohort of patients with clinically definite MS will be recruited from the two centres. Adult (\geq 18 years) people of any ethnicity and with any MS phenotype according to the McDonald's criteria²⁷⁻²⁹ version valid at the time of diagnosis will be included in the study. Their disability status score on the Expanded Disability Status Scale (EDSS)³⁰ may range from 0 (no disability) to 9.0 (severe disability). Patients will be included if they are able to speak and understand German language. Exclusion criteria are concomitant diseases which may affect subjective self-efficacy ratings (e.g. malignant diseases, other neurological or psychiatric disorders), a relapse of MS within the last two months or any medication change within four weeks prior to the study. A relapse between testing 2 and 3 would necessitate the exclusion of the participant.

The study will be advertised in the MS-Clinic, the Rehabilitation Centre and on the Austrian MS Society website. Further interested PwMS will be examined for eligibility by neurologists at the two study locations. Severely disabled PwMS (EDSS \geq 8) will be offered home visits to enable their participation. Written informed consent will be obtained by the first author (BS) who is not involved in the treatment of the patients. Participants may withdraw from the study at any time and for any reasons without prejudice. Outpatient participants will be reimbursed for travel expenses only.

Patient and public involvement

In Phase 1, patients will be lay members of the expert committee to consolidate all the translations and back translations of the USE-MS. Their role regarding the item and response option wording and sentence structure will be crucial, as the final

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questionnaire should be understood by PwMS. Patients will also be involved using faceto-face cognitive interviewing, to gain insight into their views about the clarity of the wording, meaning and completeness of the questions of the pre-final USE-MS-G. The Austrian MS (recruitment) and MS Research Societies (funding) will be involved in this study, with whom the findings will be shared as soon as available (patient magazine, meetings). The findings will also be disseminated to the UK MS Society and MS Trust.

Sample size

Phase 1

Patients will be recruited until saturation is achieved. Saturation is a standard term in qualitative methodology to signify the point when the analysis of data from new participants reveals no further emergent qualitative themes. Saturation is typically achieved after 10-30 people have been interviewed but is determined by the nature of the analysis and the participants themselves³¹.

Phase 2

Rasch analysis sample size requirements are predicated upon the degree of precision required for estimating item and person difficulties. Regardless of targeting, one can be 99% confident that a sample size of 243 participants is adequately large to obtain a (high) precision of ± 0.5 log odd units (logits). Good targeting provided, a sample size of 108 people would be sufficient^{21 32}. Using the formula N=n/(1-(z/100)) where n is the calculated number of participants and z the expected attrition rate of 15-20%, a total sample size of 286-304 participants will be aimed at in this study.

Outcomes and data collection

Assessments used in this study were developed using patient involvement and/or recommended by governmental or patient organisations (Supplementary File 2). Study outcomes and methods for their assessment are presented in Figure 1. Participant characteristics and assessments used at all time-points are shown in Table 1.

Figure 1 around here

Figure 1 Study outcomes and their assessment

Figure legend: GSE: General Self-Efficacy Scale; RS-13: Resilience Scale, short version; MusiQol: Multiple Sclerosis International Quality of Life questionnaire; HADS: 8

 Hospital Anxiety and Depression Scale; NFI-MS: Neurological Fatigue Index.

 Table 1 Participant characteristics and assessments used in this study

Participant characteristics and assessments	Phase 1	Pha	se 2
(assessments will be collected in a random order to	T1	T2	T3
avoid order effect)			
Participant identifier (ID)	X	X	X
Age	X	X	
Gender	X	X	
MS phenotype ¹	X	Х	
Disease duration	X	X	
Expanded Disability Status Scale (EDSS) ²	X	X	
Disease modifying treatment (DMT) ³	X	X	X
(Pre-final) German version of Unidimensional	X	X	X
Self-Efficacy Scale for Multiple Sclerosis			
Qualitative cognitive interview	X		
Resilience Scale, short version		X	X
General Self-Efficacy Scale		X	X
Multiple Sclerosis International Quality of Life		X	X
questionnaire			
Hospital Anxiety and Depression Scale	6	X	X
Neurological Fatigue Index	4	X	X

¹Relapsing-remitting; primary progressive; secondary progressive multiple sclerosis³³

²EDSS groups: 0-4.0; 4.5-6.5; 7.0-7.5; 8.0-9.0³⁰

³(a) No DMTs; (b) low effective DMTs: interferon-b 1a, interferon-b 1a, interferon-b 1b, pegylated interferon-b 1a, glatiramer acetate, dimethyl fumarate, teriflunomide, azathioprin, intravenous immunoglobulins; (c) high effective DMTs: alemtuzumab, cladribine, fingolimod, natalizumab, ocrelizumab, cyclophosphamide, mitoxantrone, rituximab^{34 35}

At recruitment, disability will be assessed by neurologists (FD, CB or RE) using the EDSS, ranging from 0 to 10, with higher scores representing higher levels of disability³⁰.

Although psychometric validation studies criticised its low responsiveness to changes, the EDSS has no floor or ceiling effects³⁶, has been shown to be valid and reliable³⁷ and is therefore recommended for use in clinical studies³⁸.

Excellent internal and external validity and reliability of the original USE-MS has been shown²¹. Scoring of the USE-MS draws results from all 12 items while items 5, 7, 8, 9 and 11 are reversed scored. Higher numbers represent stronger self-efficacy beliefs in participants²¹. The USE-MS includes a 4-point Likert scale (0= strongly disagree to 3=strongly agree).

To assess external construct validity, the following questionnaires will be administered:

The validated German version³⁹ of the 10-item General Self-Efficacy Scale (GSE)¹⁵ is a self-administered 4-point Likert scale with a summary score ranging from "not at all true" to "exactly true". The total GSE score ranges between 10 and 40, higher scores signifying greater self-efficacy. Psychometric testing demonstrated high internal consistency, moderate concurrent validity and unidimensionality¹⁵.

The validated German version⁴⁰ of the 13-item Resilience Scale (RS-13)⁴¹, based on the 25-item Resilience Scale⁴² will be used. RS-13 item scores from a 7-point Likert scale are added up, indicating low (13-66 points), moderate (67-72 points) or high (73-91 points) resilience⁴¹. The German RS-13 showed high internal consistency and moderate test-retest reliability. Confirmatory factor analysis indicated an acceptable model fit⁴¹.

The validated German version⁴³ of the 31-item Multiple Sclerosis International Quality of Life (MusiQol) questionnaire⁴⁴ will be employed. Response options use a 6-point Likert scale, from 1= "never/not at all" to 5= "always/very much" and 6= "not applicable". Negatively worded item scores are reversed, and for each participant mean scores for each dimension of the item scores are calculated. All nine dimension scores are linearly transformed to a 0-100 scale, their mean representing the global index score, 0 indicating the worst level of health-related QoL and 100 the best. Psychometric testing showed satisfactory internal and external validity and acceptable reliability for all MusiQol dimensions⁴⁴.

The validated German version⁴⁵ of the 14-item Hospital Anxiety Depression Scale

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(HADS)⁴⁶ will be used. The HADS is a self-report questionnaire with a 4-point Likert scale and a 42 point maximum, higher scores representing higher levels of anxiety or depression. Items 2, 4, 7, 9, 12 and 14 are reversed scored, odd items are added to score the anxiety subscale (0-21 points) and even items are added to generate the depression subscale (0-21 points). Testing of the German version demonstrated good internal consistency and acceptable test-retest reliability ⁴⁵. The two-factor structure of the scale was confirmed⁴⁵.

The validated German version⁴⁷ of the 23-item Neurological Fatigue Index (NFI-MS) will be used⁴⁸. Four factors of the NFI-MS were confirmed by principal component analysis and explained 62% of the variance. The four subscales and total scale showed acceptable responsiveness⁴⁹, good test-retest reliability, moderate convergent validity and fit to Rasch model expectations⁴⁸. Items are scored on a 4-point Likert scale from 0= "strongly disagree" to 3= "strongly agree". For scoring, the following item values are added: 1-8= "physical subscale"; 9-12= "cognitive subscale"; 13-18= "relief by diurnal sleep or rest subscale"; 19-23= "abnormal nocturnal sleep and sleepiness subscale"; and 1-7, 9, 11-12 = "physical and cognitive summary score"⁴⁸.

Assessments will be performed by trained physiotherapists holding a Master's (SK) and PhD degree (BS) and a clinical neuropsychologist (LZ). The number of participants who decline to participate or drop out will be recorded, together with reasons (CONSORT flow chart). Any health problems will be recorded.

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Phase 1: data will be collected at one time-point (Testing 1, T1), with an expected duration of 45-60 minutes.

Phase 2: for the test-retest reliability assessment, data will be collected at two timepoints and will last 60-90 minutes: Testing 2 (T2) and Testing 3 (T3), 14-21 days after T2^{48 50}.

Data management

With regard to confidentiality, the Austrian and Tyrolean Data Protection Acts will be adhered to. Double data entry and range checks for data values will be used. For qualitative content analysis, double coding of the data set will be performed. Only the research team will have access to the data. All personal data will be codified by a participant ID. Data and files will be saved on a password protected computer, will not be

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transferred via emails and will be only used for the purposes for which they were collected. Participants will be informed about their right to disclosure for their own data even if these data lack clinical utility. Codified data will be kept for 15 years following completion of the study. Blank data collection forms can be requested from the corresponding author.

Study procedures

This study will follow the Beaton et al. guidelines for the cross-cultural adaptation of patient reported outcomes⁵¹ and its enhanced version from the University of Leeds, UK.

Phase 1

Stage 1: Forward translation of the items, response options, instructions and scoring information into German will be performed by three independent translators; translator 1 is a medical professional and informed about self-efficacy, while translators 2 and 3 have no medical knowledge and are "naïve" to self-efficacy. Translators are bilingual German native speakers and will create a written report for all translations (T1, T2 and T3), which will then be compared, to distinguish any wording differences or ambiguities⁵².

Stage 2 will be a synthesis of T1-3 into T-123. Involving a fourth, unbiased person, the three versions will be discussed with the translators and any discrepancies solved by consensus. A revised questionnaire and comprehensive report will be produced⁵¹.

Stage 3: Backward translation of T-123 into English will be done by three bilingual English native speakers who are blind to the original version. Translators are "naïve" to self-efficacy and medicine, to minimise bias⁵². Vague wording, obvious inconsistencies or theoretical errors in the translations shall be detected. A report for each version, TB1, TB2 and TB3, will be written by the translators. To maximise comprehension, language will be used which can be understood by a 12 year old^{53 54}, indicated by a Flesch reading-ease score of 80-90⁵⁵. The German Flesch value=180-ASL-(58,5*ASW), where ASL=average sentence length and ASW=average number of syllables per word⁵⁵.

Stage 4: Considering written documentations, an Expert Committee will review and integrate all versions of the questionnaire, involving instructions and scoring documentation, and develop the pre-final version of the USE-MS-G. The Expert Committee will consist of three neurologists, two physiotherapists, a neuropsychologist,

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a methodologist, two language professionals, the translators, three lay PwMS and the translation synthesis recorder. The Expert Committee will be in close contact with the original USE-MS developers. A written report of the consensus process will be created. Decision-making will be based on guidelines to accomplish cross-cultural equivalence between the original and German versions in four areas⁵², shown in Figure 2.

Figure 2 around here

Figure 2 Cross-cultural equivalence areas to be achieved between original and German USE-MS (USE-MS-G); adapted from⁵²

Stage 5: Pretesting of the pre-final USE-MS-G will be performed in 30 PwMS, involving completion of the scale and face-to-face cognitive interviews. Cognitive interviewing will be used to evaluate whether survey questions are easily comprehended, response categories match natural responses, and if people are motivated to respond truthfully and accurately⁵⁶⁻⁵⁸. Leading questions will be avoided to minimise bias. Enquiries for comprehension and meaning will be used, and repetition of content by patients^{56 58}. Probing will be applied to explore cognitive processes such as memory, underlying reasons for certain responses and overall level of difficulty or confidence⁵⁷. Verbal probes, following Willis' model, will be used immediately after the questions⁵⁹: (a) standardised, anticipated probes: scripted; (b) standardised, conditional probes: scripted, but will be used only if activated by certain participant behaviors such as hesitation⁶⁰; (c) non-standardised, emergent probes: applied in reaction to participant behaviour⁶¹. The interview guide is presented in Table 2. Recording and field notes will be used, reviewed for inconsistencies or gaps shortly before the end of the interview.

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Table 2 Questions used for semi-structured interview

Parti MS.	cipants will be given sufficient time to complete the pre-final German USE-
1.	Having read the questions in the questionnaire, what are your thoughts about them?
2.	Would you please repeat this question in your own words?
3.	What do you think this question is asking?
4.	What do you think about that particular question?
5.	What do you think about the wording of this question, in terms of its clarity?
6.	How easy or hard was this to answer?
7.	How sure are you of your answer?
8.	Could you talk me through your answers in more detail?
9.	What were you thinking of when you answered this question?
10.	Do you have any other comments?
11.	If responses from participants are somewhat unclear, the interviewer asks: "Why so?"
12.	Should a participant hesitate, the interviewer conveys: "You spent some time answering that question - what were you thinking about?"
Adapt	ted from ⁵⁷ and ⁶²

An overview of study procedures is presented in Figure 3.

Figure 3 around here

Figure 3 Flowchart of the study procedures

Figure legend: MS: multiple sclerosis; T1 (2; 3): testing 1 (2; 3).

Phase 2

The USE-MS-G will be validated in a larger sample of 292 PwMS who will complete the above described questionnaires at T1 and T2.

Data analyses

Mixed methods data analyses will be used.

Phase 1- Qualitative analyses

Interviews will be transcribed and analysed using qualitative content analysis (QCA)^{63 64} using QDA MINER LITE software (Provalis Research, Montreal, Canada) and adhering to the Consolidated Criteria for Reporting Qualitative Research (COREQ)⁶⁵. Analysis steps will be performed as follows⁶⁶⁻⁶⁹:

- Data organisation based on the research question
- Identification of recurring ideas, concepts, themes and words
- Development of a coding frame (requirements: unidimensionality, mutual exclusivity of subcategories within dimensions, exhaustiveness of subcategories and saturation, where each subcategory is used at least once)

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- Selection of relevant material, structuring, marking and segmentation of text sections, based on Bandura's concept of self-efficacy and the original USE-MS, to identify main and subcategories
- Definition, naming and characterisation of categories and decision rules, to enable consistent assignment of data segments
- Illustration of categories and subcategories using citations
- Creation of a data matrix, followed by quantitative data analysis (descriptive statistics, e.g. frequencies)
- Report
- Rigor and credibility will be maximised by⁷⁰⁻⁷²

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- revision and expansion of the coding frame
- double coding of the whole dataset by two independent researchers (10-14 days after initial coding)
- checking for researcher effects (reflexivity)73

Phase 2 - Quantitative analyses

Descriptive statistics and reliability estimates will be performed using IBM SPSS software, release 25.0 (IBM Corporation, Armonk, NY, USA). Rasch Analysis will be conducted with RUMM2030 software⁷⁴. Statistical significance is defined as two-tailed p value <0.05.

Missing data will be treated as follows:

- 1) Missing data should be avoided by checking questionnaires for missing item responses and asking participants for completion.
- Rasch analysis calculates an estimate from all available data and does not require a complete data set⁷⁵.

Test-retest reliability

Test-retest reliability will be evaluated using Lin's concordance correlation coefficient (r_c) between T2 and T3 $(0-1)^{76}$ ⁷⁷. R_cs will be calculated with their 95% confidence intervals (CI). Values of <0 will be considered to indicate poor, 0–0.20 slight, 0.21–0.40 fair, 0.41–0.60 moderate, 0.61–0.80 substantial and 0.81–1 almost perfect agreement⁷⁸. The data will be racked for the analysis of the concordance correlation coefficient, and stacked for differential item functioning (DIF) by time point.

External validity

It is hypothesised that scores on the German USE-MS will demonstrate moderate to high positive correlations with scales assessing conceptually similar constructs (convergent validity; with the GSE, RS-13 and MusiQoI) and moderate to high negative correlations with scales measuring divergent constructs (divergent validity; with the HADS and NFI-MS); Spearman's Rank correlation coefficients of 0.3-0.49 being considered low, 0.5-0.69 moderate and \geq 0.7 strong⁷⁹.

Internal validity - Rasch analysis

Rasch analysis²⁵ assumes the probability of a person endorsing an item is a logistic function of the difference between the "person ability" (perceived self-efficacy) and the "item difficulty" (level of self-efficacy) expressed²⁴. Item characteristic curves, arranged on the log-odds units (logit) scale, will be used to visualise the probability of a person's correct response in relation to the item difficulty⁸⁰.

The polytomous Rasch model will be chosen for this study, suitable for scales with multiple response categories for their items⁸¹. A significant likelihood ratio test signifying inconsistent distance between response category thresholds would require the use of Masters' unrestricted (partial credit) model⁸², otherwise Andrich's rating scale model⁸³. Category thresholds are located centrally between two adjacent categories where either response is equally likely^{81 84}. The 4-point USE-MS includes three thresholds.

Ordered item category thresholds

Category probability curves will be inspected, checking regular distribution and monotonic advance of measures across categories⁸⁴.

Targeting

Targeting refers to the degree to which the scale captures the full range of self-efficacy. Inspecting person-item threshold distribution maps, the mean location score for the respondents will be compared with the default items zero value. A well-targeted scale is centred around zero logits (±0.5 logits), corresponding to the scale's item of mean difficulty⁸⁵.

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The proportion of floor and ceiling effects will be monitored, considered noteworthy if >5%⁸⁶.

Local independence

Local independence means there should be no associations between the items. Inspection of the correlation matrix of item standardised residuals should show Pearson's correlations of <0.2 above the mean value of the matrix as a whole.

Unidimensionality

Unidimensionality as a Rasch model requirement allows a summary score

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measurement of a single construct. Using a PCA of the residuals, positively and negatively loadings of the first component will be identified, generating two subsets and separate person estimates. Independent t-tests will explore significant differences⁸⁷. If less than 5% of t-tests are significant or the lower bound of the binominal CI overlaps 5%, unidimensionality is supported^{88 89}.

Fit to the Rasch model

Different fit statistics will seek to determine if the assumption of a probabilistic ordering of items is satisfied:

(a) Summary Chi-square interaction statistics and individual item Chi-square statistics are expected to be non-significant (Bonferroni-adjusted p-values for the number of items)⁴⁸.

(b) Individual person and item fit residuals are expected to be between ±2.5 (99% CI)⁹⁰.

(c) Person and summary item fit residuals reflect perfect model fit if their mean and standard deviation are close to 0 and 1, respectively^{91 92}.

Reliability

Reliability is indicated by the person separation index (PSI; range 0-1)⁹³ and Cronbach's alpha (missing data excluded), which should be ≥ 0.85 for individual use or 0.70 for group use^{48 94}.

Invariance, differential item functioning and differential test functioning

Invariance means that all persons completing a questionnaire, regardless of their ability (or self-efficacy), recognise the difficulty in identical items⁹³. Any likelihood of differently scored items between the groups violates the assumption of invariance, called DIF^{95 96}. The data will be pooled with a data set from the UK development sample and tested for invariance by language to equate the language versions. Absence of DIF will be tested in gender (female; male), age (quartile groups), disease duration (quartile groups), language (English, German), time point (retest) and centre and indicated by a non-significant ANOVA of the residuals (5% alpha with Bonferroni correction) where the group is the main factor^{96 97}. Any observed DIF will be examined to know whether it cancels out at the test level⁹⁵. If there are many items displaying DIF by language, Differential Test Functioning (DTF) will be performed.

If model fit is not achieved, an iterative stepwise procedure will be initiated, involving strategies for combining response categories, stepwise deletion of the worst fitting item, testlet (superitem) construction and adjusting for DIF as appropriate⁹⁸.

Ethics approval, permissions and dissemination plan

Ethical approval for both centres was received from the Ethics Committee of the Medical University of Innsbruck, Austria (reference number EK1260/2018; 13.12.2018). Due to the absence of an intervention, no insurance policy is required for this study and no harm to participants is expected.

Permission to translate into German and validate the original USE-MS²¹ was provided by the test developers who hold the copyright for the USE-MS-G.

Results from this study will be disseminated to the participants via mail and MS Societies (Austria, UK). Findings will be disseminated to clinicians and researchers through peer-reviewed publications and conferences.

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Author contributions

All authors critically and substantially revised the manuscript and approved the current version to be submitted for publication. BS devised and designed the study and drafted the manuscript. RM provided relevant advice on the Rasch analysis. RK developed the qualitative data analysis plan. LZ substantially contributed to the conception and design of the study. MR provided input on the study methodology and quantitative analysis. SK substantially contributed to the development of the study protocol. FD is a study manager at his centre and substantially contributed to the development of the study protocol. CB is a study manager at his centre and contributed to the design of the study protocol. RE is a study manager at his centre and substantially contributed to the design and development of the study protocol.

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Competing Interests

None declared.

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Data sharing statement

Data generated by this research that support any publications will be made available upon reasonable request as soon as possible. It will be considered submitting these data to the Open Science initiative once future analyses related to this data set are completed.

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170x173mm (300 x 300 DPI)
Semantic

Meaning of words including subtle meaning differences of words within context

Grammar and sentence structure should be considered

Idiomatic

Idioms and colloquialisms are a type of informal language

They cannot be translated literally

Equivalent expressions or item replacement with comparable meaning need to be found

EQUIVALENCE

Experiential

Situations and daily life activities described by the translated questionnaire should match Austrian cultural context

Conceptual

The concept of self-efficacy and events described by the questionnaire items may be differently perceived and experienced by people with MS in different countries or cultures.

For example, the concept of family, friends, fatigue or the condition of MS itself

Figure 2

254x190mm (250 x 250 DPI)

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Forward translation into German Contact and consensus with the original test developers from Synthesis of forward translations The Walton Centre for Neurology and Neurosurgery Backward translation into English Liverpool, UK and University of Leeds, Leeds, UK Expert committee consensus at all stages of the on pre-final German version translation process STUDY Participant recruitment of 30 people with MS PHASE 1 Testing of pre-final version by participants l Questionnaire Cognitive Qualitative completion (T1) interview content analysis V Contact with original test developers (summary checklist) Face and content validity, cultural adaption for Austria Creation of final German USE-MS-G, based on analysis Validation of the USE-MS-G in 292 people with MS \downarrow Recruitment Data Data collection (T2) collection (T3) V Quantitative data analysis, including Rasch analysis STUDY PHASE 2 Internal & external validity and reliability of USE-MS-G Consensus with the original test developers **Dissemination of findings**



190x254mm (250 x 250 DPI)

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Calvert M, Kyte D, Mercieca-Bebber R, et al. Guidelines for Inclusion of Patient-Reported Outcomes in Gigital Trial Protocols: The SPIRIT-PRO Extension. JAMA : the journal of the American Medical Association 2018;319(5):483-94 doi: 10.1001/jan22017.21903[published Online First: Epub Date])

Section/item	ItemNo	Description	SPIRIT-PRO Item No.	SPIRIT-PRO Extension of Item Description	ander Evaboration Graduata n fr	Addressed on Page No.
Administrative in	formation				rom h	
Title	1	Descriptive title identifying the study design, population, interventions, and, if applicable, trial acronym	er		tp://bmjoper	1
Trial registration	2a	Trial identifier and registry name. If not yet registered, name of intended registry		~~~~~~~~~~~~~~~~~~~~~~~~~~~~~~~~~~~~~~	bmj.com/ ol and similar t	2
	2b	All items from the World Health Organization Trial Registration Data Set			n June 10 echnolog	2: described in registry
Protocol version	3	Date and version identifier			, 2025 ies.	3
Funding	4	Sources and types of financial, material, and other support			at Depar	20
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Roles and responsibilities	5a	Names, affiliations, and roles of protocol contributors		including f	1; 20
	5b	Name and contact information for the trial sponsor	SPIRIT-5a- PRO Elaboration	Specify the individual(s) respectively the individual(s) respectively the for the PRO content of the trivel protocol.	BS
	5c	Role of study sponsor and funders, if any, in study design; collection, management, analysis, and interpretation of data; writing of the report; and the decision to submit the report for publication, including whether they will have ultimate authority over any of these activities	to.	2019. Downloaded from http:// smushogeschool . ed to text and data mining, Al t	20
	5d	Composition, roles, and responsibilities of the coordinating centre, steering committee, endpoint adjudication committee, data management team, and other individuals or groups overseeing the trial, if applicable (see Item 21a for data monitoring committee)		omjopen.bmj.com/ on June raining, and similar technol	7; 11-13
Introduction				10, 20 ogies	
Background and rationale	6a	Description of research question and justification for undertaking the trial, including summary of relevant studies (published and unpublished) examining benefits and harms for each intervention	SPIRIT-6a- PRO Extension	Describe the PRO-specific research question and rationale for PRO assessment and summarize RRO findings in relevant studies.	5-6

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	6b	Explanation for choice of comparators		including for u	10-11; Suppleme File 2
Objectives	7	Specific objectives or hypotheses	SPIRIT-7- PRO Extension	State specific PRO objectives or hypotheses (including relevants PRO concepts/domains).	6
Trial design	8	Description of trial design including type of trial (eg, parallel group, crossover, factorial, single group), allocation ratio, and framework (eg, superiority, equivalence, noninferiority, exploratory)		9. Downloaded from ushogeschool . o text and data mini	6-7
Methods: Partici	pants, in	terventions, and outcomes		http: ng, Al	
Study setting	9	Description of study settings (eg, community clinic, academic hospital) and list of countries where data will be collected. Reference to where list of study sites can be obtained	C/	//bmjopen.bmj.com/ training, and simila	7
Eligibility criteria	10	Inclusion and exclusion criteria for participants. If applicable, eligibility criteria for study centres and individuals who will perform the interventions (eg, surgeons, psychotherapists)	SPIRIT-10- PRO Extension	Specify any PRO-specific blightility criteria (eg, language/reading requirements or prerandonization completion of PRO). If PROS will not be collected from the entire study provide a rationale and describe the method for obtaining the PRO	7

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Interventions	11a	Interventions for each group with sufficient detail to allow replication, including how and when they will be administered		9-029565 on 20 Au including for uses	NA
	11b	Criteria for discontinuing or modifying allocated interventions for a given trial participant (eg, drug dose change in response to harms, participant request, or improving/worsening disease)		igust 2019. Downloa Erasmushogesch related to text and	7-8 (concerning assessments) Interventions: NA
	11c	Strategies to improve adherence to intervention protocols, and any procedures for monitoring adherence (eg, drug tablet return, laboratory tests)		aded from http:// tool . data mining, Al t	7; 13
	11d	Relevant concomitant care and interventions that are permitted or prohibited during the trial		bmjopen.bm	7
Outcomes	12	Primary, secondary, and other outcomes, including the specific measurement variable (eg, systolic blood pressure), analysis metric (eg, change from baseline, final value, time to event), method of aggregation (eg, median, proportion), and time point for each outcome. Explanation of the clinical relevance of chosen efficacy and harm outcomes is strongly recommended	SPIRIT-12- PRO Extension	Specify the PRO concepted dog nains used to evaluate the intervention (eg, overall health-related quality of light, specific domain, specific symptom areal, for each one, the analysis metric (eg, fnange from baseline, final value, time to egent) and the principal time point or period of interest.	8-11; Figure 1

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13	Time schedule of enrolment, interventions (including any run-ins and washouts), assessments, and visits for participants. A schematic diagram is highly recommended (see Figure)	SPIRIT-13- PRO Extension	Include a schedule of PR assessments, providing a rationale for the time points, and justifying if the initial assessment is not prerandomization. Specify time windows, whether PRO collection is prior to clinical assessments, and using multiple questionnaires, whether order of administration will be start a set of the start and the star	7; 11; Ta Figure 3
14	Estimated number of participants needed to achieve study objectives and how it was determined, including clinical and statistical assumptions supporting any sample size calculations	SPIRIT-14- PRO Extension	When a PRO is the primary and point, state the required sample it is the primary and point, was determined) and recruiting it is the power of the primary of	8
15	Strategies for achieving adequate participant enrolment to reach target sample size		and similar t	7
gnment of	interventions (for controlled trials)		echnc	
			9 10, 2	NA
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	13 14 15 gnment of	13 Time schedule of enrolment, interventions (including any run-ins and washouts), assessments, and visits for participants. A schematic diagram is highly recommended (see Figure) 14 Estimated number of participants needed to achieve study objectives and how it was determined, including clinical and statistical assumptions supporting any sample size calculations 15 Strategies for achieving adequate participant enrolment to reach target sample size gmment of interventions (for controlled trials)	13 Time schedule of enrolment, interventions (including any run-ins and washouts), assessments, and visits for participants. A schematic diagram is highly recommended (see Figure) SPIRIT-13- PRO 14 Estimated number of participants needed to achieve study objectives and how it was determined, including clinical and statistical assumptions supporting any sample size calculations SPIRIT-14- PRO 15 Strategies for achieving adequate participant enrolment to reach target sample size Image: Comparison of interventions (for controlled trials) 16 Image: Comparison of interventions (for controlled trials) Image: Comparison of interventions (for controlled trials)	BMJ Open BMJ Open BMJ Open 13 Time schedule of enrolment, interventions (including any run-ins and washouts), assessments, and visits for participants. A schematic diagram is highly recommended (see Figure) Include a schedule of PRE providing a rationale for the initial assessments, and justifying if the initial assessments is not prerandomized, whether PRO called from its mindows, whether PRO called from its multiple questionnaires, whether PRO administration will be strange for order of administration will be strange for order of administration will be strange for administration will be strange any sample size calculations 14 Estimated number of participants needed to achieve study objectives and how it was determined, including clinical and statistical assumptions supporting any sample size calculations SPIRIT-14- PRO When a PRO is the primate for the state the required sample for administration will be strange (and how it was determined) and recognition and statistical assumptions supporting any sample size calculations SPIRIT-14- PRO 15 Strategies for achieving adequate participant enrolment to reach target sample size analyses. 15 Strategies for achieving adequate participant enrolment to reach target sample size analyses. 16 Interventions (for controlled trials) Interventions (for controlled trials)

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Sequence generation	16a	Method of generating the allocation sequence (eg, computer-generated random numbers), and list of any factors for stratification. To reduce predictability of a random sequence, details of any planned restriction (eg, blocking) should be provided in a separate document that is unavailable to those who enrol participants or assign interventions		ncluding for uses related to text and d	-029565 on 20 August 2019. Download Erasmushogescho	NA
Allocation concealment mechanism	16b	Mechanism of implementing the allocation sequence (eg, central telephone; sequentially numbered, opaque, sealed envelopes), describing any steps to conceal the sequence until interventions are assigned		ata mining, Al training,	led from http://bmjopen	NA
Implementation	16c	Who will generate the allocation sequence, who will enrol participants, and who will assign participants to interventions		and similar tech	.bmj.com/ on Ju	NA
Blinding (masking)	17a	Who will be blinded after assignment to interventions (eg, trial participants, care providers, outcome assessors, data analysts), and how		nologies.	ne 10, 2025 at De	NA
					partment GEZ-LTA	

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	17b	If blinded, circumstances under which unblinding is permissible, and procedure for revealing a participant's allocated intervention during the trial		029565 on 20 A cluding for use	NA
Methods: Data	collection,	management, and analysis		s relat	
Data collection methods	18a	Plans for assessment and collection of outcome, baseline, and other trial data, including any related processes to promote data quality (eg, duplicate measurements, training of assessors) and a description of study instruments (eg, questionnaires, laboratory tests) along with their reliability and validity, if known. Reference to where data collection forms can be found, if not in the protocol	SPIRIT-18a (i)-PRO Extension	Justify the PRO instrument by e used and describe domains, not by e used and describe domains, not by e used and describe domains, not by e used and scoring (eg, range and direction of scores indicating a good or poor but the provided of scores indicating a good of poor but the provided of scores indicating a good of poor but the provided of scores indicating a good of poor but the provided of scores indicating a good of poor but the provided of scores indicating a good of poor but the provided of scores indicating a good of poor but the poor but t	8; 11-
			SPIRIT-18a (ii)-PRO Extension	Include a data collection gangutlining the permitted mode(s) of gamunistration (eg, paper, telephone, elegitropic, other) and setting (eg, clinic, home, gather).	7
			SPIRIT-18a (iii)-PRO Extension	Specify whether more than 1 wersion will be used and state whether translated versions have beer developed using currently recommended methods.	Total manus

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			SPIRIT-18a (iv)-PRO Extension	When the trial context requires someone other than a trial participant to answer on his or her behalf (a proxy reported outcome), state and justify the use of a proxy respondent. Provide or the evidence of the validity of the use assessment if available.	NA
	18b	Plans to promote participant retention and complete follow-up, including list of any outcome data to be collected for participants who discontinue or deviate from intervention protocols	SPIRIT-18b (i)-PRO Extension	Specify PRO data collect and management strategies for management avoidable missing data. at a mining avoidable missing data. at a mining	16
			SPIRIT-18b (ii)-PRO Elaboration	Describe the process of PRO assessment for participants who discontinue or deviate from the assigned intervention protocol.	7; 16
Data management	19	Plans for data entry, coding, security, and storage, including any related processes to promote data quality (eg, double data entry; range checks for data values). Reference to where details of data management procedures can be found, if not in the protocol		nj.com/ on June 10, 2025 a d similar technologies.	11-12; 15-16
		For peer review only - http://b	mjopen.bmj.con	t Department GEZ-LTA n/site/about/guidelines.xhtml	

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Statistical	20a	Statistical methods for analysing	SPIRIT- 20a-	State PRO analysis methed signal	16-19
methods		primary and secondary outcomes. Reference to where other details of the statistical analysis plan can be found, if not in the protocol	PRO Elaboration	any plans for addressing and fultiplicity/type I (α) error.	
	20b	Methods for any additional analyses (eg, subgroup and adjusted analyses)		st 2019. D ated to te	16; 18-19
	20c	Definition of analysis population relating to protocol non-adherence (eg, as randomised analysis), and any statistical methods to handle missing data (eg, multiple imputation)	SPIRIT- 20c- PRO Elaboration	State how missing data will be described and outline the methods for bendling missing items or entire as the methods (eg, approach to imputation and sonsitivity analyses).	16
Methods: Monite	oring			p://br Al tra	
Data monitoring	21a	Composition of data monitoring committee (DMC); summary of its role and reporting structure; statement of whether it is independent from the sponsor and competing interests; and reference to where further details about its charter can be found, if not in the protocol. Alternatively, an explanation of why a DMC is not needed		mjopen.bmj.com/ on June 10, 2025 a aining, and similar technologies.	NA

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	21b	Description of any interim analyses and stopping guidelines, including who will have access to these interim results and make the final decision to terminate the trial			-029565 on 20 Augus	NA
Harms	22	Plans for collecting, assessing, reporting, and managing solicited and spontaneously reported adverse events and other unintended effects of trial interventions or trial conduct	SPIRIT- 22- PRO Extension	State whether or not PRC monitored during the stud clinical care of individual and, if so, how this will be standardized way. Descr process will be explained eg, in the participant info and consent form.	attended in a attended in a attend	NA 11
Auditing	23	Frequency and procedures for auditing trial conduct, if any, and whether the process will be independent from investigators and the sponsor			://bmjopen.bmj.	NA
Ethics and disse	mination			0	com/ c imilar	
Research ethics approval	24	Plans for seeking research ethics committee/institutional review board (REC/IRB) approval			on June 10, 20 technologies	19
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Protocol amendments	25	Plans for communicating important protocol modifications (eg, changes to eligibility criteria, outcomes, analyses) to relevant parties (eg, investigators, REC/IRBs, trial participants, trial registries, journals, regulators)			Eras	9-029565 on 20 August 20	NA
Consent or assent	26a	Who will obtain informed consent or assent from potential trial participants or authorised surrogates, and how (see Item 32)			mushogeschool I to text and data	019. Downloaded	7
	26b	Additional consent provisions for collection and use of participant data and biological specimens in ancillary studies, if applicable	101		. mining, Al train	from http://bmi	NA
Confidentiality	27	How personal information about potential and enrolled participants will be collected, shared, and maintained in order to protect confidentiality before, during, and after the trial		en on	ing, and similar tec	open.bmi.com/ on J	11-12
Declaration of interests	28	Financial and other competing interests for principal investigators for the overall trial and each study site			hnologies.	une 10. 2025	20
Access to data	29	Statement of who will have access to the final trial dataset, and disclosure of contractual agreements that limit such access for investigators				at Department	11-12

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Ancillary and post- trial care	30	Provisions, if any, for ancillary and post- trial care, and for compensation to those who suffer harm from trial participation			19-029565 on 20	NA
Dissemination policy	31a	Plans for investigators and sponsor to communicate trial results to participants, healthcare professionals, the public, and other relevant groups (eg, via publication, reporting in results databases, or other data sharing arrangements), including any publication restrictions			August 2019. Downloaded fro Erasmushogeschool . Ises related to text and data mi	8; 19-20
	31b	Authorship eligibility guidelines and any intended use of professional writers	ro.		om http://	19
	31c	Plans, if any, for granting public access to the full protocol, participant-level dataset, and statistical code		24,	omjopen.bm, raining, and	20
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Informed consent materials	32	Model consent form and other related documentation given to participants and authorised surrogates			on June 10, ; r technologie	12
Biological specimens	33	Plans for collection, laboratory evaluation, and storage of biological specimens for genetic or molecular analysis in the current trial and for future use in ancillary studies, if applicable			2025 at Department	NA

BMJ Open Abbreviations: SPIRIT, Standard Protocol Items: Recommendations for Interventional Trials; PRO, patient-reported outcome. *It is strongly recommended that this checklist be read in conjunction with the SPIRIT 2013 Explanation 💈 E boration for important clarification on The strongly recommended that this checklist be read in conjunction with the SPIRIT 2013 Explanation **B** Esplanation **Commercial-NoDerivs 3.0 Unported**" license and is reproduced with permission. The spirit over and attribution the total and the terms of te the items. Amendments to the protocol should be tracked and dated. The SPIRIT checklist is copyrighter by the SPIRIT Group under the Creative

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Supplementary File 2

Except good to excellent psychometric properties, the selection of outcome measures depended also on patient involvement during their development, patient acceptability of the tools or recommendations from relevant governmental organisations or MS Societies. The MusiQol was developed using individual semistructured interviews of 1992 PwMS in five countries, followed by qualitative content analysis. The MS-EDGE Outcome Measure Task Force reviewed and recommended the MusiQol for use in PwMS[1]. Similarly, both the German long and short versions of the Resilience Scale are based on face-to-face interviews of 2031 people. The GSE was exposed to repeated validation studies in thousands of participants where excellent validity and reliability were confirmed. A study explored the validity of the HADS in PwMS using interviews by an interviewer who was blind to the HADS scores [2]. The acceptability of the German HADS to various patient populations was tested and confirmed [3]. The UK National Institute for Health and Care Excellence (NICE) 2018 Guidelines for MS[4] recommended the HADS anxiety subscale as a sensitive and specific anxiety screening tool, based on a review[5]. The NFI-MS was developed through a two-stage process in 635 PwMS: guantitative validation and qualitative interviews with patients. The NFI-MS was reviewed and recommended by the MS International Federation (MSIF)[6].

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GERMAN TRANSLATION, CULTURAL ADAPTION AND VALIDATION OF THE UNIDIMENSIONAL SELF-EFFICACY SCALE FOR MULTIPLE SCLEROSIS: STUDY PROTOCOL

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GERMAN TRANSLATION, CULTURAL ADAPTION AND VALIDATION OF THE UNIDIMENSIONAL SELF-EFFICACY SCALE FOR MULTIPLE SCLEROSIS: STUDY PROTOCOL

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ABSTRACT

Introduction: Self-efficacy refers to individuals' confidence in their ability to perform relevant tasks to accomplish desired goals. This is independent of their actual abilities. In people with multiple sclerosis (MS), self-efficacy has been shown to powerfully influence motivation and health-related behaviour, such as adherence to prescribed treatment or physical activity. So far, a rigorously tested German language self-efficacy questionnaire for people with MS is missing.

Methods: The purpose of this study is to translate the original Unidimensional Self-Efficacy Scale for Multiple Sclerosis (USE-MS) into German and to validate the German USE-MS (USE-MS-G). Based on Bandura's concept of self-efficacy and international guidelines for questionnaire development, the patient-led development of the pre-final German version will involve a forward-backward translation process, synthesis of translations, expert committee review and consensus with the original test developers. At two centres in Tyrol, Austria, content and face validity and cultural adaption for Austria will be established using face-to-face semi-structured cognitive interviews of 30 people with MS. A further 292 people with MS with minimal to severe disability will be tested at two time-points to validate the USE-MS-G.

Results: Mixed methods analyses will be applied. Interviews will be transcribed and analysed employing qualitative content analysis. External validity will be explored using Spearman's Rank correlation coefficients of the USE-MS-G with the 13-item Resilience Scale, General Self-Efficacy Scale, Multiple Sclerosis International Quality of Life questionnaire, Hospital Anxiety and Depression Scale and MS-specific Neurological Fatigue Index. Test-retest reliability, internal consistency and floor and ceiling effects will be evaluated. Internal validity will be examined using Rasch analysis.

Ethics and dissemination: Ethical approval was received from the Ethics Committee of the Medical University of Innsbruck, Austria (reference number EK1260/2018; 13.12.2018). Results from this study will be disseminated to the participants and MS Societies, and to clinicians and researchers through peer-reviewed publications and conferences.

Study registration: ISRCTN Registry; trial ID ISRCTN14843579; prospectively registered on 02. 01. 2019; http://www.isrctn.com/ISRCTN14843579

Keywords: Multiple Sclerosis; Self Efficacy; Patient Reported Outcome Austria; Cross-Cultural Comparison; Validation Studies as Topic.	Study pro	tocol, revision 1, 26.6.2019
	Keywords	s: Multiple Sclerosis; Self Efficacy; Patient Reported Outcome ross-Cultural Comparison: Validation Studies as Tonic

ARTICLE SUMMARY

Strengths and limitations of this study

- This study protocol describes the German translation of the original English language Unidimensional Self-efficacy Scale for Multiple Sclerosis (USE-MS), upon permission of the scale developers and applying international recommendations.

- Consistent with the conceptual framework of the English USE-MS, Bandura's concept of self-efficacy will be adhered to.

- Employing a patient-led process in phase 1, 30 people with MS (PwMS) will be interviewed about the pre-final German USE-MS, to establish face and content validity and cultural adaption for PwMS in Austria.

- In phase 2, the German USE-MS will be validated in a larger sample of 292 PwMS.

- Applying classical test theory and Rasch analysis approaches, internal and external validity, internal consistency and test retest reliability will be explored.

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INTRODUCTION

Multiple sclerosis (MS) is one of the most common neurological diseases in young adults worldwide, with increasing prevalence¹. MS is characterised by a wide variety of symptoms and different disease courses². Despite the development of novel disease modifying drugs and neurorehabilitation strategies, the unpredictability of the disease with psychological distress, losses in social contact and quality of life (QoL) are concerning for PwMS. However, individuals' self-knowledge can modulate their approach to day-to-day activities. According to Bandura's social cognitive theory, psychosocial functioning is regulated by reciprocal interactions between behaviour, personal factors and environmental conditions³. Self-regulation and intrinsic motivation enable individuals to set and pursue their own goals, observe and evaluate themselves in relation to attained goals⁴. Bandura defined self-efficacy as individuals' beliefs regarding their capability to perform significant tasks, to achieve goals that are meaningful for their daily lives³. Self-efficacy beliefs considerably influence people's feelings, thoughts and motivation⁵ while, notably, being independent of their physical performance⁵. Such a concept appears important for people with disabilities because it may shape their motivation to initiate and adhere to treatment, particularly when facing side effects.

Perceived self-efficacy influences health-related behaviour such as adhering to medication⁶ or engaging in physical activity in PwMS⁷. Health status evaluations of responses to rehabilitation and steroid treatment after an MS relapse can be predicted by self-efficacy levels⁸. Also, higher self-efficacy levels are associated with better long-term perceived cognitive functioning⁹ and QoL^{10 11}. PwMS who report higher perceived self-efficacy also state lower levels of fatigue, depression and anxiety¹². Recent evidence has provided insight into the importance of self-management and intrinsic motivation for motor learning¹³. Recognising the relevance of self-efficacy especially for people with disabilities, valid and reliable measurement tools are still needed for its assessment. Three generic self-efficacy scales were found in the literature^{7 14-16}. However, generic questionnaires may not adequately cover the construct of self-efficacy in a chronic neurological disease like MS. The initial impact of a diagnosis of MS, in addition to the manifold symptoms and necessity of managing a progressive disease may affect individuals' self-efficacy perceptions. Studies demonstrated that the

capability to effectively solve problems, consistent with higher self-efficacy levels, is strongly associated with PwMS' psychological adaptation to their disability¹⁷, supporting the choice of a disease-specific over a generic self-efficacy questionnaire. MS-specific self-efficacy scales include the Liverpool Self-efficacy Scale (LSES)¹⁸, Multiple Sclerosis Self-Efficacy Scale (MSSS)¹⁹, MS Self-Efficacy Scale (MSSE)²⁰, Unidimensional Self-Efficacy Scale for Multiple Sclerosis (USE-MS)²¹ and University of Washington Self-Efficacy Scale for people with disabilities²².

Following current guidelines, patients should be involved in the translation and development process of disease specific questionnaires, to ensure the scale reflects their experiences²³. LSES and MSSS development used in-depth patient interviews while the USE-MS consists of items from both the LSES and MSSS. Bandura's concept of self-efficacy is reflected in the wording of all three questionnaires. The USE-MS study sample was the largest thereof (N=303), and only the USE-MS was exposed to Rasch²⁴ ²⁵ analysis assessing internal construct validity, in addition to conventional external construct validity and reliability testing. Fit to the Rasch model was demonstrated, and good external validity and reliability²¹. Consequently, the USE-MS appears to be appropriate for use in clinical practice and research. However, so far no validated German language version of the USE-MS is available. The purpose of this study will therefore be to translate the USE-MS into German and validate the German language version in a larger sample of PwMS.

METHODS

Study aims

The first aim of this patient led study is to translate the original English USE-MS, developed by Young et al. (2012) into German, based on international guidelines.

The second aim is to establish face and content validity and cultural adaption of the German version for PwMS in Austria, using individual semi-structured cognitive interviews.

The third aim is to evaluate internal and external validity, internal consistency and test-retest reliability of the German USE-MS (USE-MS-G), using classical test theory and Rasch analysis .

Study design

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This will be a bi-centre prospective cross-sectional translation and validation study with repeated measures, consisting of Phase 1 and Phase 2. The SPIRIT 2013 and SPIRIT-PRO Extension checklist for study protocols²⁶ is presented as Supplementary File 1.

Study setting and timeline

Locations will be the outpatient MS-Clinic of the Clinical Department of Neurology, Medical University of Innsbruck, Austria and Department of Neurology, Clinic for Rehabilitation Münster, Austria.

The expected overall study duration is 33 months, from 01.02.2019 to 31.10. 2021.

Participants and recruitment

A random cross-sectional cohort of patients with clinically definite MS will be recruited from the two centres. Adult (\geq 18 years) people of any ethnicity and with any MS phenotype according to the McDonald's criteria²⁷⁻²⁹ version valid at the time of diagnosis will be included in the study. Their disability status score on the Expanded Disability Status Scale (EDSS)³⁰ may range from 0 (no disability) to 9.0 (severe disability). Patients will be included if they are able to speak and understand German language. Exclusion criteria are concomitant diseases which may affect subjective self-efficacy ratings (e.g. malignant diseases, other neurological or psychiatric disorders), a relapse of MS within the last two months or any medication change within four weeks prior to the study. A relapse between testing 2 and 3 would necessitate the exclusion of the participant.

The study will be advertised in the MS-Clinic, the Rehabilitation Centre and on the Austrian MS Society website. Further interested PwMS will be examined for eligibility by neurologists at the two study locations. Severely disabled PwMS (EDSS \geq 8) will be offered home visits to enable their participation. Written informed consent will be obtained by the first author (BS) who is not involved in the treatment of the patients. Participants may withdraw from the study at any time and for any reasons without prejudice. Outpatient participants will be reimbursed for travel expenses only.

Patient and public involvement

In Phase 1, patients will be lay members of the expert committee to consolidate all the translations and back translations of the USE-MS. Their role regarding the item and response option wording and sentence structure will be crucial, as the final

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questionnaire should be understood by PwMS. Patients will also be involved using faceto-face cognitive interviewing, to gain insight into their views about the clarity of the wording, meaning and completeness of the questions of the pre-final USE-MS-G. The Austrian MS (recruitment) and MS Research Societies (funding) will be involved in this study, with whom the findings will be shared as soon as available (patient magazine, meetings). The findings will also be disseminated to the UK MS Society and MS Trust.

Sample size

Phase 1

Patients will be recruited until saturation is achieved. Saturation is a standard term in qualitative methodology to signify the point when the analysis of data from new participants reveals no further emergent qualitative themes. Saturation is typically achieved after 10-30 people have been interviewed but is determined by the nature of the analysis and the participants themselves³¹.

Phase 2

Rasch analysis sample size requirements are predicated upon the degree of precision required for estimating item and person difficulties. Regardless of targeting, one can be 99% confident that a sample size of 243 participants is adequately large to obtain a (high) precision of ± 0.5 log odd units (logits). Good targeting provided, a sample size of 108 people would be sufficient^{21 32}. Using the formula N=n/(1-(z/100)) where n is the calculated number of participants and z the expected attrition rate of 15-20%, a total sample size of 286-304 participants will be aimed at in this study.

Outcomes and data collection

Assessments used in this study were developed using patient involvement and/or recommended by governmental or patient organisations (Supplementary File 2). Study outcomes and methods for their assessment are presented in Figure 1. Participant characteristics and assessments used at all time-points are shown in Table 1.

Figure 1 around here

Figure 1 Study outcomes and their assessment

Figure legend: GSE: General Self-Efficacy Scale; RS-13: Resilience Scale, short version; MusiQol: Multiple Sclerosis International Quality of Life questionnaire; HADS: 8

 Hospital Anxiety and Depression Scale; NFI-MS: Neurological Fatigue Index.

 Table 1 Participant characteristics and assessments used in this study

Participant characteristics and assessments	Phase 1	Pha	se 2
(assessments will be collected in a random order to	T1	T2	T3
avoid order effect)			
Participant identifier (ID)	X	X	X
Age	X	X	
Gender	X	X	
MS phenotype ¹	X	Х	
Disease duration	X	X	
Expanded Disability Status Scale (EDSS) ²	X	X	
Disease modifying treatment (DMT) ³	X	X	X
(Pre-final) German version of Unidimensional	X	X	X
Self-Efficacy Scale for Multiple Sclerosis			
Qualitative cognitive interview	X		
Resilience Scale, short version		X	X
General Self-Efficacy Scale		X	X
Multiple Sclerosis International Quality of Life		X	X
questionnaire			
Hospital Anxiety and Depression Scale		X	X
Neurological Fatigue Index	4	X	X

¹Relapsing-remitting; primary progressive; secondary progressive multiple sclerosis³³

²EDSS groups: 0-4.0; 4.5-6.5; 7.0-7.5; 8.0-9.0³⁰

³(a) No DMTs; (b) low effective DMTs: interferon-b 1a, interferon-b 1a, interferon-b 1b, pegylated interferon-b 1a, glatiramer acetate, dimethyl fumarate, teriflunomide, azathioprin, intravenous immunoglobulins; (c) high effective DMTs: alemtuzumab, cladribine, fingolimod, natalizumab, ocrelizumab, cyclophosphamide, mitoxantrone, rituximab^{34 35}

At recruitment, disability will be assessed by neurologists (FD, CB or RE) using the EDSS, ranging from 0 to 10, with higher scores representing higher levels of disability³⁰.

Although psychometric validation studies criticised its low responsiveness to changes, the EDSS has no floor or ceiling effects³⁶, has been shown to be valid and reliable³⁷ and is therefore recommended for use in clinical studies³⁸.

Excellent internal and external validity and reliability of the original USE-MS has been shown²¹. Scoring of the USE-MS draws results from all 12 items while items 5, 7, 8, 9 and 11 are reversed scored. Higher numbers represent stronger self-efficacy beliefs in participants²¹. The USE-MS includes a 4-point Likert scale (0= strongly disagree to 3=strongly agree).

To assess external construct validity, the following questionnaires will be administered:

The validated German version³⁹ of the 10-item General Self-Efficacy Scale (GSE)¹⁵ is a self-administered 4-point Likert scale with a summary score ranging from "not at all true" to "exactly true". The total GSE score ranges between 10 and 40, higher scores signifying greater self-efficacy. Psychometric testing demonstrated high internal consistency, moderate concurrent validity and unidimensionality¹⁵.

The validated German version⁴⁰ of the 13-item Resilience Scale (RS-13)⁴¹, based on the 25-item Resilience Scale⁴² will be used. RS-13 item scores from a 7-point Likert scale are added up, indicating low (13-66 points), moderate (67-72 points) or high (73-91 points) resilience⁴¹. The German RS-13 showed high internal consistency and moderate test-retest reliability. Confirmatory factor analysis indicated an acceptable model fit⁴¹.

The validated German version⁴³ of the 31-item Multiple Sclerosis International Quality of Life (MusiQol) questionnaire⁴⁴ will be employed. Response options use a 6-point Likert scale, from 1= "never/not at all" to 5= "always/very much" and 6= "not applicable". Negatively worded item scores are reversed, and for each participant mean scores for each dimension of the item scores are calculated. All nine dimension scores are linearly transformed to a 0-100 scale, their mean representing the global index score, 0 indicating the worst level of health-related QoL and 100 the best. Psychometric testing showed satisfactory internal and external validity and acceptable reliability for all MusiQol dimensions⁴⁴.

The validated German version⁴⁵ of the 14-item Hospital Anxiety Depression Scale

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(HADS)⁴⁶ will be used. The HADS is a self-report questionnaire with a 4-point Likert scale and a 42 point maximum, higher scores representing higher levels of anxiety or depression. Items 2, 4, 7, 9, 12 and 14 are reversed scored, odd items are added to score the anxiety subscale (0-21 points) and even items are added to generate the depression subscale (0-21 points). Testing of the German version demonstrated good internal consistency and acceptable test-retest reliability ⁴⁵. The two-factor structure of the scale was confirmed⁴⁵.

The validated German version⁴⁷ of the 23-item Neurological Fatigue Index (NFI-MS) will be used⁴⁸. Four factors of the NFI-MS were confirmed by principal component analysis and explained 62% of the variance. The four subscales and total scale showed acceptable responsiveness⁴⁹, good test-retest reliability, moderate convergent validity and fit to Rasch model expectations⁴⁸. Items are scored on a 4-point Likert scale from 0= "strongly disagree" to 3= "strongly agree". For scoring, the following item values are added: 1-8= "physical subscale"; 9-12= "cognitive subscale"; 13-18= "relief by diurnal sleep or rest subscale"; 19-23= "abnormal nocturnal sleep and sleepiness subscale"; and 1-7, 9, 11-12 = "physical and cognitive summary score"⁴⁸.

Assessments will be performed by trained physiotherapists holding a Master's (SK) and PhD degree (BS) and a clinical neuropsychologist (LZ). The number of participants who decline to participate or drop out will be recorded, together with reasons (CONSORT flow chart). Any health problems will be recorded.

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Phase 1: data will be collected at one time-point (Testing 1, T1), with an expected duration of 45-60 minutes.

Phase 2: for the test-retest reliability assessment, data will be collected at two timepoints and will last 60-90 minutes: Testing 2 (T2) and Testing 3 (T3), 14-21 days after T2^{48 50}.

Data management

With regard to confidentiality, the Austrian and Tyrolean Data Protection Acts will be adhered to. Double data entry and range checks for data values will be used. For qualitative content analysis, double coding of the data set will be performed. Only the research team will have access to the data. All personal data will be codified by a participant ID. Data and files will be saved on a password protected computer, will not be

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transferred via emails and will be only used for the purposes for which they were collected. Participants will be informed about their right to disclosure for their own data even if these data lack clinical utility. Codified data will be kept for 15 years following completion of the study. Blank data collection forms can be requested from the corresponding author.

Study procedures

This study will follow the Beaton et al. guidelines for the cross-cultural adaptation of patient reported outcomes⁵¹ and its enhanced version from the University of Leeds, UK.

Phase 1

Stage 1: Forward translation of the items, response options, instructions and scoring information into German will be performed by three independent translators; translator 1 is a medical professional and informed about self-efficacy, while translators 2 and 3 have no medical knowledge and are "naïve" to self-efficacy. Translators are bilingual German native speakers and will create a written report for all translations (T1, T2 and T3), which will then be compared, to distinguish any wording differences or ambiguities⁵².

Stage 2 will be a synthesis of T1-3 into T-123. Involving a fourth, unbiased person, the three versions will be discussed with the translators and any discrepancies solved by consensus. A revised questionnaire and comprehensive report will be produced⁵¹.

Stage 3: Backward translation of T-123 into English will be done by three bilingual English native speakers who are blind to the original version. Translators are "naïve" to self-efficacy and medicine, to minimise bias⁵². Vague wording, obvious inconsistencies or theoretical errors in the translations shall be detected. A report for each version, TB1, TB2 and TB3, will be written by the translators. To maximise comprehension, language will be used which can be understood by a 12 year old^{53 54}, indicated by a Flesch reading-ease score of 80-90⁵⁵. The German Flesch value=180-ASL-(58,5*ASW), where ASL=average sentence length and ASW=average number of syllables per word⁵⁵.

Stage 4: Considering written documentations, an Expert Committee will review and integrate all versions of the questionnaire, involving instructions and scoring documentation, and develop the pre-final version of the USE-MS-G. The Expert Committee will consist of three neurologists, two physiotherapists, a neuropsychologist,

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a methodologist, two language professionals, the translators, three lay PwMS and the translation synthesis recorder. The Expert Committee will be in close contact with the original USE-MS developers. A written report of the consensus process will be created. Decision-making will be based on guidelines to accomplish cross-cultural equivalence between the original and German versions in four areas⁵², shown in Figure 2.

Figure 2 around here

Figure 2 Cross-cultural equivalence areas to be achieved between original and German USE-MS (USE-MS-G); adapted from⁵²

Stage 5: Pretesting of the pre-final USE-MS-G will be performed in 30 PwMS, involving completion of the scale and face-to-face cognitive interviews. Cognitive interviewing will be used to evaluate whether survey questions are easily comprehended, response categories match natural responses, and if people are motivated to respond truthfully and accurately⁵⁶⁻⁵⁸. Leading questions will be avoided to minimise bias. Enquiries for comprehension and meaning will be used, and repetition of content by patients^{56 58}. Probing will be applied to explore cognitive processes such as memory, underlying reasons for certain responses and overall level of difficulty or confidence⁵⁷. Verbal probes, following Willis' model, will be used immediately after the questions⁵⁹: (a) standardised, anticipated probes: scripted; (b) standardised, conditional probes: scripted, but will be used only if activated by certain participant behaviors such as hesitation⁶⁰; (c) non-standardised, emergent probes: applied in reaction to participant behaviour⁶¹. The interview guide is presented in Table 2. Recording and field notes will be used, reviewed for inconsistencies or gaps shortly before the end of the interview.

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Table 2 Questions used for semi-structured interview

Parti MS.	cipants will be given sufficient time to complete the pre-final German USE-
1.	Having read the questions in the questionnaire, what are your thoughts about them?
2.	Would you please repeat this question in your own words?
3.	What do you think this question is asking?
4.	What do you think about that particular question?
5.	What do you think about the wording of this question, in terms of its clarity?
6.	How easy or hard was this to answer?
7.	How sure are you of your answer?
8.	Could you talk me through your answers in more detail?
9.	What were you thinking of when you answered this question?
10.	Do you have any other comments?
11.	If responses from participants are somewhat unclear, the interviewer asks: "Why so?"
12.	Should a participant hesitate, the interviewer conveys: "You spent some time answering that question - what were you thinking about?"
Adapt	ted from ⁵⁷ and ⁶²

An overview of study procedures is presented in Figure 3.

Figure 3 around here

Figure 3 Flowchart of the study procedures

Figure legend: MS: multiple sclerosis; T1 (2; 3): testing 1 (2; 3).

Phase 2

The USE-MS-G will be validated in a larger sample of 292 PwMS who will complete the above described questionnaires at T1 and T2.

Data analyses

Mixed methods data analyses will be used.

Phase 1- Qualitative analyses

Interviews will be transcribed and analysed using qualitative content analysis (QCA)^{63 64} using QDA MINER LITE software (Provalis Research, Montreal, Canada) and adhering to the Consolidated Criteria for Reporting Qualitative Research (COREQ)⁶⁵. Analysis steps will be performed as follows⁶⁶⁻⁶⁹:

- Data organisation based on the research question
- Identification of recurring ideas, concepts, themes and words
- Development of a coding frame (requirements: unidimensionality, mutual exclusivity of subcategories within dimensions, exhaustiveness of subcategories and saturation, where each subcategory is used at least once)

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- Selection of relevant material, structuring, marking and segmentation of text sections, based on Bandura's concept of self-efficacy and the original USE-MS, to identify main and subcategories
- Definition, naming and characterisation of categories and decision rules, to enable consistent assignment of data segments
- Illustration of categories and subcategories using citations
- Creation of a data matrix, followed by quantitative data analysis (descriptive statistics, e.g. frequencies)
- Report
- Rigor and credibility will be maximised by⁷⁰⁻⁷²

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- revision and expansion of the coding frame
- double coding of the whole dataset by two independent researchers (10-14 days after initial coding)
- checking for researcher effects (reflexivity)73

Phase 2 - Quantitative analyses

Descriptive statistics and reliability estimates will be performed using IBM SPSS software, release 25.0 (IBM Corporation, Armonk, NY, USA). Rasch Analysis will be conducted with RUMM2030 software⁷⁴. Statistical significance is defined as two-tailed p value <0.05.

Missing data will be treated as follows:

- 1) Missing data should be avoided by checking questionnaires for missing item responses and asking participants for completion.
- Rasch analysis calculates an estimate from all available data and does not require a complete data set⁷⁵.

Test-retest reliability

Test-retest reliability will be evaluated using Lin's concordance correlation coefficient (r_c) between T2 and T3 $(0-1)^{76}$ ⁷⁷. R_cs will be calculated with their 95% confidence intervals (CI). Values of <0 will be considered to indicate poor, 0–0.20 slight, 0.21–0.40 fair, 0.41–0.60 moderate, 0.61–0.80 substantial and 0.81–1 almost perfect agreement⁷⁸. The data will be racked for the analysis of the concordance correlation coefficient, and stacked for differential item functioning (DIF) by time point.

External validity

It is hypothesised that scores on the German USE-MS will demonstrate moderate to high positive correlations with scales assessing conceptually similar constructs (convergent validity; with the GSE, RS-13 and MusiQoI) and moderate to high negative correlations with scales measuring divergent constructs (divergent validity; with the HADS and NFI-MS); Spearman's Rank correlation coefficients of 0.3-0.49 being considered low, 0.5-0.69 moderate and \geq 0.7 strong⁷⁹.

Internal validity - Rasch analysis

Rasch analysis²⁵ assumes the probability of a person endorsing an item is a logistic function of the difference between the "person ability" (perceived self-efficacy) and the "item difficulty" (level of self-efficacy) expressed²⁴. Item characteristic curves, arranged on the log-odds units (logit) scale, will be used to visualise the probability of a person's correct response in relation to the item difficulty⁸⁰.

The polytomous Rasch model will be chosen for this study, suitable for scales with multiple response categories for their items⁸¹. A significant likelihood ratio test signifying inconsistent distance between response category thresholds would require the use of Masters' unrestricted (partial credit) model⁸², otherwise Andrich's rating scale model⁸³. Category thresholds are located centrally between two adjacent categories where either response is equally likely^{81 84}. The 4-point USE-MS includes three thresholds.

Ordered item category thresholds

Category probability curves will be inspected, checking regular distribution and monotonic advance of measures across categories⁸⁴.

Targeting

Targeting refers to the degree to which the scale captures the full range of self-efficacy. Inspecting person-item threshold distribution maps, the mean location score for the respondents will be compared with the default items zero value. A well-targeted scale is centred around zero logits (±0.5 logits), corresponding to the scale's item of mean difficulty⁸⁵.

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The proportion of floor and ceiling effects will be monitored, considered noteworthy if >5%⁸⁶.

Local independence

Local independence means there should be no associations between the items. Inspection of the correlation matrix of item standardised residuals should show Pearson's correlations of <0.2 above the mean value of the matrix as a whole.

Unidimensionality

Unidimensionality as a Rasch model requirement allows a summary score

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measurement of a single construct. Using a PCA of the residuals, positively and negatively loadings of the first component will be identified, generating two subsets and separate person estimates. Independent t-tests will explore significant differences⁸⁷. If less than 5% of t-tests are significant or the lower bound of the binominal CI overlaps 5%, unidimensionality is supported^{88 89}.

Fit to the Rasch model

Different fit statistics will seek to determine if the assumption of a probabilistic ordering of items is satisfied:

(a) Summary Chi-square interaction statistics and individual item Chi-square statistics are expected to be non-significant (Bonferroni-adjusted p-values for the number of items)⁴⁸.

(b) Individual person and item fit residuals are expected to be between ±2.5 (99% CI)⁹⁰.

(c) Person and summary item fit residuals reflect perfect model fit if their mean and standard deviation are close to 0 and 1, respectively^{91 92}.

Reliability

Reliability is indicated by the person separation index (PSI; range 0-1)⁹³ and Cronbach's alpha (missing data excluded), which should be ≥ 0.85 for individual use or 0.70 for group use^{48 94}.

Invariance, differential item functioning and differential test functioning

Invariance means that all persons completing a questionnaire, regardless of their ability (or self-efficacy), recognise the difficulty in identical items⁹³. Any likelihood of differently scored items between the groups violates the assumption of invariance, called DIF^{95 96}. The data will be pooled with a data set from the UK development sample and tested for invariance by language to equate the language versions. Absence of DIF will be tested in gender (female; male), age (quartile groups), disease duration (quartile groups), language (English, German), time point (retest) and centre and indicated by a non-significant ANOVA of the residuals (5% alpha with Bonferroni correction) where the group is the main factor^{96 97}. Any observed DIF will be examined to know whether it cancels out at the test level⁹⁵. If there are many items displaying DIF by language, Differential Test Functioning (DTF) will be performed.
If model fit is not achieved, an iterative stepwise procedure will be initiated, involving strategies for combining response categories, stepwise deletion of the worst fitting item, testlet (superitem) construction and adjusting for DIF as appropriate⁹⁸.

Ethics approval, permissions and dissemination plan

Ethical approval for both centres was received from the Ethics Committee of the Medical University of Innsbruck, Austria (reference number EK1260/2018; 13.12.2018). Due to the absence of an intervention, no insurance policy is required for this study and no harm to participants is expected.

Permission to translate into German and validate the original USE-MS²¹ was provided by the test developers who hold the copyright for the USE-MS-G.

Results from this study will be disseminated to the participants via mail and MS Societies (Austria, UK). Findings will be disseminated to clinicians and researchers through peer-reviewed publications and conferences.

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Author contributions

All authors critically and substantially revised the manuscript and approved the current version to be submitted for publication. BS devised and designed the study and drafted the manuscript. RM provided relevant advice on the Rasch analysis. RK developed the qualitative data analysis plan. LZ substantially contributed to the conception and design of the study. MR provided input on the study methodology and quantitative analysis. SK substantially contributed to the development of the study protocol. FD is a study manager at his centre and substantially contributed to the development of the study protocol. CB is a study manager at his centre and contributed to the design of the study protocol. RE is a study manager at his centre and substantially contributed to the design and development of the study protocol.

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Competing Interests

None declared.

Data sharing statement

Data generated by this research that support any publications will be made available upon reasonable request as soon as possible. It will be considered submitting these data to the Open Science initiative once future analyses related to this data set are completed. The informed consent form will include the consent to controlled data sharing.

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170x173mm (300 x 300 DPI)

Semantic

Meaning of words including subtle meaning differences of words within context

Grammar and sentence structure should be considered

Idiomatic

Idioms and colloquialisms are a type of informal language

They cannot be translated literally

Equivalent expressions or item replacement with comparable meaning need to be found

EQUIVALENCE

Experiential

Situations and daily life activities described by the translated questionnaire should match Austrian cultural context

Conceptual

The concept of self-efficacy and events described by the questionnaire items may be differently perceived and experienced by people with MS in different countries or cultures.

For example, the concept of family, friends, fatigue or the condition of MS itself

Figure 2

254x190mm (250 x 250 DPI)

BMJ Open: first published as 10.1136/bmjopen-2019-029565 on 20 August 2019. Downloaded from http://bmjopen.bmj.com/ on June 10, 2025 at Department GEZ-LTA Erasmushogeschool . Protected by copyright, including for uses related to text and data mining, Al training, and similar technologies.



Forward translation into German Contact and consensus with the original test developers from Synthesis of forward translations The Walton Centre for Neurology and Neurosurgery Backward translation into English Liverpool, UK and University of Leeds, Leeds, UK Expert committee consensus at all stages of the on pre-final German version translation process STUDY Participant recruitment of 30 people with MS PHASE 1 Testing of pre-final version by participants l Questionnaire Cognitive Qualitative completion (T1) interview content analysis V Contact with original test developers (summary checklist) Face and content validity, cultural adaption for Austria Creation of final German USE-MS-G, based on analysis Validation of the USE-MS-G in 292 people with MS \downarrow Recruitment Data Data collection (T2) collection (T3) V Quantitative data analysis, including Rasch analysis STUDY PHASE 2 Internal & external validity and reliability of USE-MS-G Consensus with the original test developers **Dissemination of findings**



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Calvert M, Kyte D, Mercieca-Bebber R, et al. Guidelines for Inclusion of Patient-Reported Outcomes in Gigital Trial Protocols: The SPIRIT-PRO Extension. JAMA : the journal of the American Medical Association 2018;319(5):483-94 doi: 10.1001/jan22017.21903[published Online First: Epub Date])

Section/item	ItemNo	Description	SPIRIT-PRO Item No.	SPIRIT-PRO Extension of Item Description	ander Evaboration Graduata n fr	Addressed on Page No.
Administrative in	formation				rom h	
Title	1	Descriptive title identifying the study design, population, interventions, and, if applicable, trial acronym	er		tp://bmjoper	1
Trial registration	2a	Trial identifier and registry name. If not yet registered, name of intended registry		~~~~~~~~~~~~~~~~~~~~~~~~~~~~~~~~~~~~~~	bmj.com/ ol and similar t	2
	2b	All items from the World Health Organization Trial Registration Data Set			n June 10 echnolog	2: described in registry
Protocol version	3	Date and version identifier			, 2025 ies.	3
Funding	4	Sources and types of financial, material, and other support			at Depar	20
					tment GEZ-L1	

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Roles and responsibilities	5a	Names, affiliations, and roles of protocol contributors		including f	1; 20
	5b	Name and contact information for the trial sponsor	SPIRIT-5a- PRO Elaboration	Specify the individual(s) respectively the individual(s) respectively the for the PRO content of the trivel protocol.	BS
	5c	Role of study sponsor and funders, if any, in study design; collection, management, analysis, and interpretation of data; writing of the report; and the decision to submit the report for publication, including whether they will have ultimate authority over any of these activities	to.	2019. Downloaded from http:// smushogeschool . ed to text and data mining, Al t	20
	5d	Composition, roles, and responsibilities of the coordinating centre, steering committee, endpoint adjudication committee, data management team, and other individuals or groups overseeing the trial, if applicable (see Item 21a for data monitoring committee)		omjopen.bmj.com/ on June raining, and similar technol	7; 11-13
Introduction				10, 20 ogies	
Background and rationale	6a	Description of research question and justification for undertaking the trial, including summary of relevant studies (published and unpublished) examining benefits and harms for each intervention	SPIRIT-6a- PRO Extension	Describe the PRO-specific research question and rationale for PRO assessment and summarize RRO findings in relevant studies.	5-6

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	6b	Explanation for choice of comparators		including for u	10-11; Suppleme File 2
Objectives	7	Specific objectives or hypotheses	SPIRIT-7- PRO Extension	State specific PRO objectives or hypotheses (including relevants PRO concepts/domains).	6
Trial design	8	Description of trial design including type of trial (eg, parallel group, crossover, factorial, single group), allocation ratio, and framework (eg, superiority, equivalence, noninferiority, exploratory)		9. Downloaded from ushogeschool . o text and data mini	6-7
Methods: Partici	pants, in	terventions, and outcomes		http: ng, Al	
Study setting	9	Description of study settings (eg, community clinic, academic hospital) and list of countries where data will be collected. Reference to where list of study sites can be obtained	C/	//bmjopen.bmj.com/ training, and simila	7
Eligibility criteria	10	Inclusion and exclusion criteria for participants. If applicable, eligibility criteria for study centres and individuals who will perform the interventions (eg, surgeons, psychotherapists)	SPIRIT-10- PRO Extension	Specify any PRO-specific blightility criteria (eg, language/reading requirements or prerandonization completion of PRO). If PROS will not be collected from the entire study provide a rationale and describe the method for obtaining the PRO	7

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Interventions	11a	Interventions for each group with sufficient detail to allow replication, including how and when they will be administered		9-029565 on 20 Au including for uses	NA
	11b	Criteria for discontinuing or modifying allocated interventions for a given trial participant (eg, drug dose change in response to harms, participant request, or improving/worsening disease)		igust 2019. Downloa Erasmushogesch related to text and	7-8 (concerning assessments) Interventions: NA
	11c	Strategies to improve adherence to intervention protocols, and any procedures for monitoring adherence (eg, drug tablet return, laboratory tests)		aded from http:// tool . data mining, Al t	7; 13
	11d	Relevant concomitant care and interventions that are permitted or prohibited during the trial		bmjopen.bm	7
Outcomes	12	Primary, secondary, and other outcomes, including the specific measurement variable (eg, systolic blood pressure), analysis metric (eg, change from baseline, final value, time to event), method of aggregation (eg, median, proportion), and time point for each outcome. Explanation of the clinical relevance of chosen efficacy and harm outcomes is strongly recommended	SPIRIT-12- PRO Extension	Specify the PRO concepted dog nains used to evaluate the intervention (eg, overall health-related quality of light, specific domain, specific symptom areal, for each one, the analysis metric (eg, change from baseline, final value, time to egent) and the principal time point or period of interest.	8-11; Figure 1

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13	Time schedule of enrolment, interventions (including any run-ins and washouts), assessments, and visits for participants. A schematic diagram is highly recommended (see Figure)	SPIRIT-13- PRO Extension	Include a schedule of PR assessments, providing a rationale for the time points, and justifying if the initial assessment is not prerandomization. Specify time windows, whether PRO collection is prior to clinical assessments, and using multiple questionnaires, whether order of administration will be start and a start	7; 11; Ta Figure 3
14	Estimated number of participants needed to achieve study objectives and how it was determined, including clinical and statistical assumptions supporting any sample size calculations	SPIRIT-14- PRO Extension	When a PRO is the primary and point, state the required sample it is the primary and point, was determined) and recruiting it is the power of the primary of	8
15	Strategies for achieving adequate participant enrolment to reach target sample size		and similar t	7
gnment of	interventions (for controlled trials)		echnc	
			9 10, 2	NA
)25 at Department GEZ-LTA	
	13 14 15 gnment of	13 Time schedule of enrolment, interventions (including any run-ins and washouts), assessments, and visits for participants. A schematic diagram is highly recommended (see Figure) 14 Estimated number of participants needed to achieve study objectives and how it was determined, including clinical and statistical assumptions supporting any sample size calculations 15 Strategies for achieving adequate participant enrolment to reach target sample size gmment of interventions (for controlled trials)	13 Time schedule of enrolment, interventions (including any run-ins and washouts), assessments, and visits for participants. A schematic diagram is highly recommended (see Figure) SPIRIT-13- PRO 14 Estimated number of participants needed to achieve study objectives and how it was determined, including clinical and statistical assumptions supporting any sample size calculations SPIRIT-14- PRO 15 Strategies for achieving adequate participant enrolment to reach target sample size Image: Comparison of interventions (for controlled trials) 16 Image: Comparison of interventions (for controlled trials) Image: Comparison of interventions (for controlled trials)	BMJ Open BMJ Open BMJ Open 13 Time schedule of enrolment, interventions (including any run-ins and washouts), assessments, and visits for participants. A schematic diagram is highly recommended (see Figure) Include a schedule of PRE providing a rationale for the initial assessments, and justifying if the initial assessments is not prerandomized, whether PRO called from its mindows, whether PRO called from its multiple questionnaires, whether PRO administration will be stand the stand how it was determined, including clinical and statistical assumptions supporting any sample size calculations SPIRIT-14- PRO When a PRO is the primate of the initial state the required sample as is not statistical based on the PRO end point, state the required sample as is not statistical analyses. 15 Strategies for achieving adequate participant enrolment to reach target sample size Init erventions (for controlled trials) 16 Interventions (for controlled trials) Interventions (for controlled trials)

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Sequence generation	16a	Method of generating the allocation sequence (eg, computer-generated random numbers), and list of any factors for stratification. To reduce predictability of a random sequence, details of any planned restriction (eg, blocking) should be provided in a separate document that is unavailable to those who enrol participants or assign interventions		ncluding for uses related to text and d	-029565 on 20 August 2019. Download Erasmushogescho	NA
Allocation concealment mechanism	16b	Mechanism of implementing the allocation sequence (eg, central telephone; sequentially numbered, opaque, sealed envelopes), describing any steps to conceal the sequence until interventions are assigned		ata mining, Al training,	led from http://bmjopen	NA
Implementation	16c	Who will generate the allocation sequence, who will enrol participants, and who will assign participants to interventions		and similar tech	.bmj.com/ on Ju	NA
Blinding (masking)	17a	Who will be blinded after assignment to interventions (eg, trial participants, care providers, outcome assessors, data analysts), and how		nologies.	ne 10, 2025 at De	NA
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				ight, in	
	17b	If blinded, circumstances under which unblinding is permissible, and procedure for revealing a participant's allocated intervention during the trial		029565 on 20 A cluding for use	NA
Methods: Data	collection,	management, and analysis		s relat	
Data collection methods	18a	Plans for assessment and collection of outcome, baseline, and other trial data, including any related processes to promote data quality (eg, duplicate measurements, training of assessors) and a description of study instruments (eg, questionnaires, laboratory tests) along with their reliability and validity, if known. Reference to where data collection forms can be found, if not in the protocol	SPIRIT-18a (i)-PRO Extension	Justify the PRO instrument by e used and describe domains, not by e used and describe domains, not by e used and describe domains, not by e used and scoring (eg, range and direction of scores indicating a good or poor but the provided of scores indicating a good of poor but the provided of scores indicating a good of poor but the provided of scores indicating a good of poor but the provided of scores indicating a good of poor but the provided of scores indicating a good of poor but the provided of scores indicating a good of poor but the poor but t	8; 11-
			SPIRIT-18a (ii)-PRO Extension	Include a data collection gangutlining the permitted mode(s) of gamunistration (eg, paper, telephone, elegitropic, other) and setting (eg, clinic, home, gather).	7
			SPIRIT-18a (iii)-PRO Extension	Specify whether more than 1 wersion will be used and state whether translated versions have beer developed using currently recommended methods.	Total manus

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			SPIRIT-18a (iv)-PRO Extension	When the trial context requires someone other than a trial participant to answer on his or her behalf (a proxy reported outcome), state and justify the use of a proxy respondent. Provide or the evidence of the validity of the use assessment if available.	NA
	18b	Plans to promote participant retention and complete follow-up, including list of any outcome data to be collected for participants who discontinue or deviate from intervention protocols	SPIRIT-18b (i)-PRO Extension	Specify PRO data collect	16
			SPIRIT-18b (ii)-PRO Elaboration	Describe the process of PRO assessment for participants who discontinue or deviate from the assigned intervention protocol.	7; 16
Data management	19	Plans for data entry, coding, security, and storage, including any related processes to promote data quality (eg, double data entry; range checks for data values). Reference to where details of data management procedures can be found, if not in the protocol		nj.com/ on June 10, 2025 a d similar technologies.	11-12; 15-16
		For peer review only - http://b	mjopen.bmj.con	t Department GEZ-LTA n/site/about/guidelines.xhtml	

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Statistical	20a	Statistical methods for analysing	SPIRIT- 20a-	State PRO analysis methed signal	16-19
methods		primary and secondary outcomes. Reference to where other details of the statistical analysis plan can be found, if not in the protocol	PRO Elaboration	any plans for addressing and fultiplicity/type I (α) error.	
	20b	Methods for any additional analyses (eg, subgroup and adjusted analyses)		st 2019. D ated to te	16; 18-19
	20c	Definition of analysis population relating to protocol non-adherence (eg, as randomised analysis), and any statistical methods to handle missing data (eg, multiple imputation)	SPIRIT- 20c- PRO Elaboration	State how missing data will be described and outline the methods for bendling missing items or entire as the methods (eg, approach to imputation and sonsitivity analyses).	16
Methods: Monite	oring			p://br Al tra	
Data monitoring	21a	Composition of data monitoring committee (DMC); summary of its role and reporting structure; statement of whether it is independent from the sponsor and competing interests; and reference to where further details about its charter can be found, if not in the protocol. Alternatively, an explanation of why a DMC is not needed		mjopen.bmj.com/ on June 10, 2025 a aining, and similar technologies.	NA

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	21b	Description of any interim analyses and stopping guidelines, including who will have access to these interim results and make the final decision to terminate the trial			-029565 on 20 Augus Including for uses rel	NA
Harms	22	Plans for collecting, assessing, reporting, and managing solicited and spontaneously reported adverse events and other unintended effects of trial interventions or trial conduct	SPIRIT- 22- PRO Extension	State whether or not PRC monitored during the stud clinical care of individual and, if so, how this will be standardized way. Descr process will be explained eg, in the participant info and consent form.	attention attention	NA 11
Auditing	23	Frequency and procedures for auditing trial conduct, if any, and whether the process will be independent from investigators and the sponsor			://bmjopen.bmj. l training, and s	NA
Ethics and disse	mination			0	tom/ c imilar	
Research ethics approval	24	Plans for seeking research ethics committee/institutional review board (REC/IRB) approval			n June 10, 20 technologies	19
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Protocol amendments	25	Plans for communicating important protocol modifications (eg, changes to eligibility criteria, outcomes, analyses) to relevant parties (eg, investigators, REC/IRBs, trial participants, trial registries, journals, regulators)			Eras	9-029565 on 20 August 20	NA
Consent or assent	26a	Who will obtain informed consent or assent from potential trial participants or authorised surrogates, and how (see Item 32)			mushogeschool I to text and data	019. Downloaded	7
	26b	Additional consent provisions for collection and use of participant data and biological specimens in ancillary studies, if applicable	re		. mining, Al train	from http://bmi	NA
Confidentiality	27	How personal information about potential and enrolled participants will be collected, shared, and maintained in order to protect confidentiality before, during, and after the trial		en 00/	ing, and similar tec	open.bmi.com/ on J	11-12
Declaration of interests	28	Financial and other competing interests for principal investigators for the overall trial and each study site			hnologies.	une 10. 2025	20
Access to data	29	Statement of who will have access to the final trial dataset, and disclosure of contractual agreements that limit such access for investigators	_			at Department	11-12

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Ancillary and post- trial care	30	Provisions, if any, for ancillary and post- trial care, and for compensation to those who suffer harm from trial participation			19-029565 on 20	NA
Dissemination policy	31a	Plans for investigators and sponsor to communicate trial results to participants, healthcare professionals, the public, and other relevant groups (eg, via publication, reporting in results databases, or other data sharing arrangements), including any publication restrictions			 August 2019. Downloaded fro Erasmushogeschool . ses related to text and data mi 	8; 19-20
	31b	Authorship eligibility guidelines and any intended use of professional writers	ro.		om http://	19
	31c	Plans, if any, for granting public access to the full protocol, participant-level dataset, and statistical code		24,	bmjopen.bm) raining, and	20
Appendices				06	.com/ simila	
Informed consent materials	32	Model consent form and other related documentation given to participants and authorised surrogates			on June 10, ; r technologie	12
Biological specimens	33	Plans for collection, laboratory evaluation, and storage of biological specimens for genetic or molecular analysis in the current trial and for future use in ancillary studies, if applicable			2025 at Department	NA

BMJ Open Abbreviations: SPIRIT, Standard Protocol Items: Recommendations for Interventional Trials; PRO, patient-reported outcome. *It is strongly recommended that this checklist be read in conjunction with the SPIRIT 2013 Explanation 💈 E boration for important clarification on The strongly recommended that this checklist be read in conjunction with the SPIRIT 2013 Explanation **B** Esplanation **Commercial-NoDerivs 3.0 Unported**" license and is reproduced with permission. The spirit over and attribution the total and the terms of te the items. Amendments to the protocol should be tracked and dated. The SPIRIT checklist is copyrighter by the SPIRIT Group under the Creative

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Supplementary File 2

Except good to excellent psychometric properties, the selection of outcome measures depended also on patient involvement during their development, patient acceptability of the tools or recommendations from relevant governmental organisations or MS Societies. The MusiQol was developed using individual semistructured interviews of 1992 PwMS in five countries, followed by qualitative content analysis. The MS-EDGE Outcome Measure Task Force reviewed and recommended the MusiQol for use in PwMS[1]. Similarly, both the German long and short versions of the Resilience Scale are based on face-to-face interviews of 2031 people. The GSE was exposed to repeated validation studies in thousands of participants where excellent validity and reliability were confirmed. A study explored the validity of the HADS in PwMS using interviews by an interviewer who was blind to the HADS scores [2]. The acceptability of the German HADS to various patient populations was tested and confirmed [3]. The UK National Institute for Health and Care Excellence (NICE) 2018 Guidelines for MS[4] recommended the HADS anxiety subscale as a sensitive and specific anxiety screening tool, based on a review[5]. The NFI-MS was developed through a two-stage process in 635 PwMS: guantitative validation and qualitative interviews with patients. The NFI-MS was reviewed and recommended by the MS International Federation (MSIF)[6].

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- Surveillance of Multiple sclerosis in adults: management (NICE guideline CG186). Secondary Surveillance of Multiple sclerosis in adults: management (NICE guideline CG186) 2018.

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7 8	3. Litsler B, Hest Riv, Patteri 3B, et al. Screening Tools for Anxiety in People with
9	Multiple Sclerosis: A Systematic Review. Int J MS Care 2016; 18 (6):273-81 doi:
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