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Stigma and Quality of Life in People with Neurodegenerative Conditions
featuring Visible Motor Symptoms: A systematic review

AND

Self-Stigma, Psychological Flexibility, Emotional Distress and Quality of Life
in People living with Multiple Sclerosis.



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Thesis Abstract

The aim of the systematic review was to assess whether stigma was associated with anxiety, depression and quality of life (QoL) in individuals living with neurodegenerative conditions featuring visible motor symptoms. The neurodegenerative conditions included were Multiple Sclerosis, Parkinson's Disease, Motor Neurones Disease (MND) /Amyotrophic Lateral Sclerosis (ALS) and Huntington's Disease, which each contain symptoms such as movement difficulties that may be visible to others and result in negative stigmatising experiences. The empirical study aimed to investigate whether psychological flexibility and emotional distress independently and serially mediated the association between *self-stigma* and QoL in people with Multiple Sclerosis (pwMS). A systematic review was conducted between the years of each journal inception until December 2022, with an updated search completed in March 2023. A cross sectional study examined whether psychological flexibility and emotional distress (anxiety, depression) independently and serially mediated the association between *self-stigma* and QoL. A series of online measures were completed by 208 pwMS. Twenty three studies were included in this review which highlighted that the strength of the link between stigma and anxiety ($r= 0.15- 0.58$), depression ($r=0.29-0.64$) and QoL ($r= 0.39- 0.82$) varied across studies. Psychological flexibility and emotional distress independently and serially mediated the association between *self-stigma* and QoL amongst pwMS. The systematic review highlighted that attention towards the role of stigma is necessary in order to promote health outcomes amongst individuals with neurodegenerative conditions. The empirical study highlighted the importance of attending to psychological flexibility and emotional distress as potential treatment mechanisms to protect against the harmful effects of *self-stigma* on QoL in pwMS.

Lay Summary

It is estimated that over one million people in the UK live with a neurodegenerative condition. Four neurodegenerative conditions including Multiple Sclerosis (MS), Parkinson's Disease (PD), Motor Neurons Disease/Amyotrophic Lateral Sclerosis (MND) and Huntington's Disease (HD) feature symptoms which may be visible to others. The progression of symptoms across the aforementioned conditions can be unpredictable and may include difficulties with walking, abnormal muscle tone or stiffness and writhing jerks. Individuals may require more support with daily tasks which may also be visible to others. Stigma occurs when individuals are treated negatively by others on the basis of being viewed to be different. Individuals with these conditions can be treated negatively by people in society and stigmatised due to perceived differences. Stigma experiences can lead to loss of job opportunities, isolation and disengagement from health services and may impact on individuals' mood levels and affect their overall quality of life. However, the stigma experiences of people with neurodegenerative conditions have rarely been studied. Therefore, we thought it may be useful to address this research gap in order to improve health outcomes for this client group.

Section I of this thesis project was a systematic review which aimed to ascertain whether stigma experiences across people with MS, PD, MND or HD impacted on anxiety and depression levels and affected quality of life. The systematic review also aimed to determine if there were relationships between variables such as age, gender or the number of years that individuals had lived with each condition and stigma. The results indicated that stigma was associated with anxiety and depression levels across conditions and was also

related to poorer quality of life. There was some suggestion that factors such as age, gender and diagnosis duration may be related to stigma, however these results should be replicated using designs which collect data at more than one time point. It emerged that different types of stigma might vary in how they affect the mood levels and quality of life of individuals with neurodegenerative conditions.

On this basis, section II of the thesis project (an empirical research project) was conducted. This project explored whether *self-stigma* (when individuals' incorporate negative attitudes about them onto their identity) was related to quality of life, amongst people with Multiple Sclerosis. Acceptance and Commitment Therapy (ACT) has popularised the term 'psychological flexibility' which relates to the development of acceptance skills, contacting the present moment and behaving in ways which are in line with an individuals' values. It was proposed that psychological flexibility and emotional distress might account for some of the association between stigma and quality of life.

In order to test this proposal, a series of online questionnaires were completed by 208 people with MS. These measured stigma experiences, emotional distress, psychological flexibility and quality of life. The results indicated that psychological flexibility and emotional distress were relevant factors in the association between *self-stigma* and quality of life. Therefore these factors may be useful to monitor in clinical settings to prevent any harmful effects of *self-stigma* on the quality of life of people living with MS. An example of a treatment that may be useful in direct work to promote psychological flexibility amongst people with MS is Acceptance and Commitment Therapy (ACT). However, wider

interventions which focus on the awareness of the detrimental effects of stigma may also be advantageous.

In summary, stigma experiences are important to account for when assessing and working with people with neurodegenerative conditions. Future research should continue to investigate stigma experiences amongst people with neurodegenerative conditions and aim to collect data at more than one time point. This might help to understand further how stigma affects individuals across the duration of their condition. Efforts to reduce the burden of *self-stigma* amongst people with MS are necessary in order to improve their quality of life.

Stigma and Quality of Life in People with Neurodegenerative Conditions featuring Visible Motor Symptoms: A systematic review.

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(see Appendix A for author guidelines).

1.0 Abstract

This systematic review aimed to investigate whether stigma was associated with anxiety, depression and quality of life in individuals living with Multiple Sclerosis, Parkinson's Disease, Motor Neurones Disease and Huntington's Disease. As each of these neurodegenerative conditions possess physical symptoms that may be evident to others, this review explores relevant correlates of stigma including condition specificity, age and disease progression amongst individuals with neurodegenerative conditions. A systematic review was carried out from the beginning of each journal inception until December 2022, across five electronic databases. An updated search was completed in March 2023. Cross-sectional and longitudinal studies were included in the review, in line with studies which met inclusion criteria. Twenty three relevant studies were identified, which indicated that stigma was related to anxiety ($r= 0.15- 0.58$), depression ($r= 0.29-0.64$) and quality of life ($r= 0.39- 0.82$) amongst individuals living with neurodegenerative conditions. However, the strength of the association between stigma, anxiety, depression and quality of life varied across studies. It may be useful for future research to focus on addressing stigma experiences at both an individual and wider public level, in order to up skill society towards the detrimental impact of stigma on health outcomes amongst individuals with neurodegenerative conditions.

Keywords: Stigma, Anxiety, Depression, Quality of Life, Multiple Sclerosis, Parkinsons Disease, Motor Neurones Disease, Huntingtons Disease, Neurodegenerative Conditions

2.0 Introduction

Neurodegenerative conditions such as Multiple Sclerosis (MS), Parkinson's Disease (PD), Motor Neurone's Disease (MND) /ALS and Huntington's disease (HD) involve progressive deterioration of brain function, which can result in a significant decrease of quality of life for individuals and their families (Batista & Pereira., 2016) and although treatments exist to slow decline or provide symptomatic management of symptoms, these conditions are currently incurable. Processing problems of the central nervous system and peripheral weakness are common characteristics of neurodegenerative conditions (Jones et al., 2012). MS is a progressive inflammatory disorder, characterised by demyelination in the central nervous system (CNS) (Wallis et al., 2020). Unpredictable symptoms can manifest such as fatigue, muscle stiffness, gait difficulties, visual problems and cognitive issues (Benito-Lyon et al, 2003; Hyarat et al., 2019). Similarly in PD, symptoms such as tremor, rigidity, gait disturbances, speech difficulties, akinesia (movement initiation difficulties) and bradykinesia (slowed movement) can occur (Moustafa et al., 2016). In MND or ALS, symptoms follow a period of rapid muscle wasting and body paralysis (Aoun et al., 2020), whilst HD, an inherited neurodegenerative disease, is characterised by uncontrolled motor symptoms and cognitive and emotional deficits (Finkbeiner, 2011).

The quality of life (QoL) of people living with neurodegenerative conditions can be significantly impacted as people endeavour to cope with the unpredictability of symptom progression (Hodayuni et al., 2021; Yalachov et al., 2019; Meyers et al., 2000). QoL often referred to and measured clinically as health-related quality of life (HRQoL), relates to the cognitive appraisal that a person makes in connection to how their condition is affecting their

life (Upton & Upton, 2015). HRQoL can be described as a person's capacity to draw satisfaction from their thoughts and behaviour despite the impact of the condition (Meyers et al., 2000). Some individuals with neurodegenerative conditions such as HD may experience dissatisfaction with life through the physical limitations of their condition which can be visible to others (Broersma et al., 2018). Despite there being no pharmaceutical cures for neurodegenerative conditions, individuals are living longer with such conditions (Collins, 2022). Studies often describe quality of life and health-related quality of life interchangeably, which is a common occurrence throughout the literature which leads to inherent confusion due to distinct differences between both concepts (Karimi & Brazier., 2016). As QoL can be argued to extend beyond health status (Tennant & Mckenna, 1995), this review will adopt the term QoL in an effort to understand related factors which may inform treatment and care provisions for this client group. Traditional stigma frameworks have focused on single health conditions and investigated psychological processes amongst individuals, which has encouraged a siloed approach towards stigma research (Stangl et al., 2019). It is hoped that a review of how stigma relates to QoL across neurodegenerative conditions may help to address this issue.

2.1 Stigma & Neurodegenerative conditions

One factor which may impact on QoL amongst such conditions is stigma. Stigma is defined as an “attribute that is deeply discrediting” whereby a stigmatised person is reduced “from a whole and usual person to a tainted, discounted one” (Goffman., 1963, p.3). The visible physical properties of neurodegenerative conditions may lead individuals to be treated unfairly by others, on the basis of their perceived differentness (Eccles et al., 2022). Certain

characteristics which may be visible to others can potentially result in stigmatising social experiences (Joachim & Acorn, 2000). Neurodegenerative conditions are frequently associated with visible physical symptoms including gait disturbances, muscle spasticity and muscle weakness in MS (Gustavsen et al., 2021), facial masking (reduced facial expression) and tremors in PD (Ma et al., 2019), abrupt, irregular and unpredictable movements, characterised as chorea in HD (Roos, 2010) and bulbar dysfunction in MND (Schluter et al., 2018). When an individual experiences discrimination on the grounds of their perceived inferiority or unacceptability, this is termed as *enacted stigma* (Scambler & Hopkins, 1986). An example of *enacted stigma* (often referred to as *public stigma*) would relate to denying an individual a job opportunity who had a mental illness (Fox et al., 2018).

Once an individual fears or expects *enacted stigma*, this can be conceptualised as *perceived stigma* or *felt stigma*, with both terms often used interchangeably throughout studies (Corrigan et al., 2006a). When *enacted stigma* is recognised by an individual, *internalised stigma* or *self- stigma* can ensue. *Self- stigma* relates to internalising public attitudes onto ones' identity which can lead to the distortion of self- image, increase negative emotions and influence the concealment of conditions (Corrigan & Rao, 2012). Given that stigma can influence the propensity of condition concealment and lead to isolation and disengagement from services (Corrigan, 2004), it is important to explore stigma amongst individuals with neurodegenerative conditions to optimise health outcomes for this client group.

2.2 Stigma, Emotional distress and QoL

Stigma can influence health outcomes by worsening and undermining social relationships, stress, resource availability and adaptive psychological and behavioural responses (Hatzenbuehler et al., 2013). Anxiety and depression are prevalent in conditions where people experience stigma (Sikkema et al., 2000). There is a high prevalence of anxiety (Wallis et al., 2019) and depression (Cadden et al., 2018) amongst people living with MS. In PD, a recent meta-analysis of 129 studies reported a 38% prevalence rate of depression (Cong et al., 2022), whilst anxiety and depression have also been reported to be high amongst individuals with MND as the rapid, progressive condition is associated with a short survival time and low survival rate (Cui et al., 2015). In HD, depression is also considered to be a principal feature of the condition (Gubert et al., 2020). Such emotional distress can occur at any point in the trajectory of illness and negatively impact the quality of life of individuals and their caregivers (Baquero & Martin, 2015). Anxiety and depression have been demonstrated to influence quality of life in people living with MND (Edge et al., 2020) and PD (Dissanayaka et al., 2010). Increased stigma experiences have been associated with poorer quality of life amongst individuals with Multiple Sclerosis (Valvano et al., 2016) and PD (Ma et al., 2019), while a longitudinal study of individuals with HD reported an association between perceived stigma and emotional health (Boileau et al., 2020).

Few studies to date have also explored how stigma relates to quality of life in neurodegenerative conditions with visible motor symptoms. Whilst psychological symptoms such as depression and anxiety have been previously conceptualised as distress in a previous

study of people with Multiple Sclerosis (pwMS) (Meek et al., 2022), they will be referred to as emotional distress in this review to promote clarity.

2.3 Stigma and clinical, demographic/social factors

Stigma is a widespread public health issue which may affect individuals from across the lifespan. The prevalence of stigmatic experiences have been reported to vary between demographic groups, with younger men with PD reporting increased incidences of *perceived stigma* (Skorvanek et al., 2015). Younger men diagnosed with PD may have alterations to their perceptions and expectations regarding the fulfilment of social, occupational and family roles at an earlier stage of life (Salazar et al., 2019). In addition, research in a healthy population of men and women reported that men who held more *self-stigma* towards psychological support seeking were more likely to hold negative views towards seeking such support (Topkaya, 2014). Few studies have addressed gender, stigma and clinical factors amongst neurodegenerative conditions.

The visible motor symptoms of neurodegenerative conditions may be evident to others and result in negative social experiences. The Stereotype Content Model suggests that individuals engage in stereotyping behaviour which is based on the perceived warmth and competence of social groups (Fiske et al., 2002). Amongst pwPD, facial masking has been rated as more detrimental towards negative impressions of older women (Hemmesch, 2014), which could be related to their perceived lack of warmth related to their reduced facial expression. In another study, *felt-stigma* was associated with higher motor symptom severity and depressive symptoms amongst females compared to males, while *felt-stigma* was

correlated with disease duration in males only (Hou et al., 2021). Stigma experiences have also been reported in working age adults, as a recent systematic review highlighted that pwMS with more severe symptoms were more likely to be stigmatized and less likely to communicate their diagnosis to others in the workplace (Vitturi et al., 2022). Consideration of the role of demographic and social factors such as gender and employment status may be useful to inform stigma related interventions targeted at particular demographic groups.

A review of research to date which addresses whether stigma is associated with emotional distress and QoL may allow for a deeper understanding of how stigma relates to health-outcomes amongst the aforementioned conditions. It is also important to acknowledge the wider factors which may influence stigma experiences and QoL, so that future interventions might address the role of relevant variables in informing care considerations across neurodegenerative conditions. Research comparing meaningful daily activity across MS, PD & MND for example, has identified the need to target the physical and social consequences of symptoms, with the amelioration of stigma suggested to be a fundamental step (Morley et al., 2018).

2.4 Aims

The aim of this systematic review was to collate data pertaining to stigma experiences and levels of emotional distress (anxiety, depression) amongst individuals living with neurodegenerative conditions with visible motor symptoms. The review also aimed to ascertain whether stigma experiences were related to demographic and social factors across conditions. This research is important as the findings may be useful in order to inform

decisions around resource allocation (Morley et al., 2018) and improve potential treatment pathways for individuals living with MS, PD, MND & HD. To address the aims of this review the following research questions were identified:

2.1 Research Questions

- 1) Is stigma associated with anxiety, depression and quality of life across neurodegenerative conditions with visible motor symptoms?
- 2) Is stigma correlated with other factors such as age, gender, disease duration or progression amongst people living with neurodegenerative conditions with visible motor symptoms?

3.0 Methods

The systematic review was registered on PROSPERO (CRD42022375129). A study protocol was developed with guidance from the Centre for Reviews and Dissemination (CRD; 2009). A meta-analysis was not performed due to the heterogeneity of statistical data and outcome measures utilised throughout studies, therefore a narrative approach towards data synthesis was chosen.

3.1 Inclusion and Exclusion Criteria

The PIO ‘Population, Intervention/Issue, Outcome’ framework was used to guide the inclusion and exclusion criteria. Studies that met the inclusion criteria featured: (i) Quantitative Methodology; (ii) Participants with a diagnosis of Parkinson’s Disease (PD), Motor Neurone’s Disease (MND) or Amyotrophic Lateral Sclerosis (ALS), Multiple Sclerosis (MS) or Huntingtons (HD); (iii) Participants aged 18+; (iv) Studies that measured stigma and correlated this with clinical factors e.g. anxiety or depression, or social/ demographic variables. Studies that met exclusion criteria included: (i) Qualitative studies; (ii) Intervention studies, as they were out with the scope of this review; (iii) Studies where stigma was measured but not correlated with demographic, social or clinical factors; (iv) Studies that focused on individuals with the HD gene, but at the pre-symptomatic phase as at this point there would be no visible symptoms; (v) Studies that included co-morbid neurological conditions; (vi) Studies that lack a confirmed diagnosis e.g. clinically isolated syndrome; (vii) Papers that are not written in English.

3.2 Search Strategy

Preliminary scoping exercises were conducted using the Database of Abstracts for Reviews and Effects (DARE), Cochrane Database of Systematic Reviews (CDSR), Web of Science, PROSPERO and Google Scholar. ProQuest Dissertations and Theses Global and the Electronic Theses Online Service (ETHOS) were accessed using the keywords ‘Stigma, Multiple Sclerosis and Neurological conditions’. Following consultation with an expert academic librarian, search terms were widened to include synonyms of each

neurodegenerative condition in addition to the search terms “stigma*”, “quality of life”, “anxi*” and “depress*” (Table 1).

Table 1

Ovid Search Sequence Used in Systematic Review

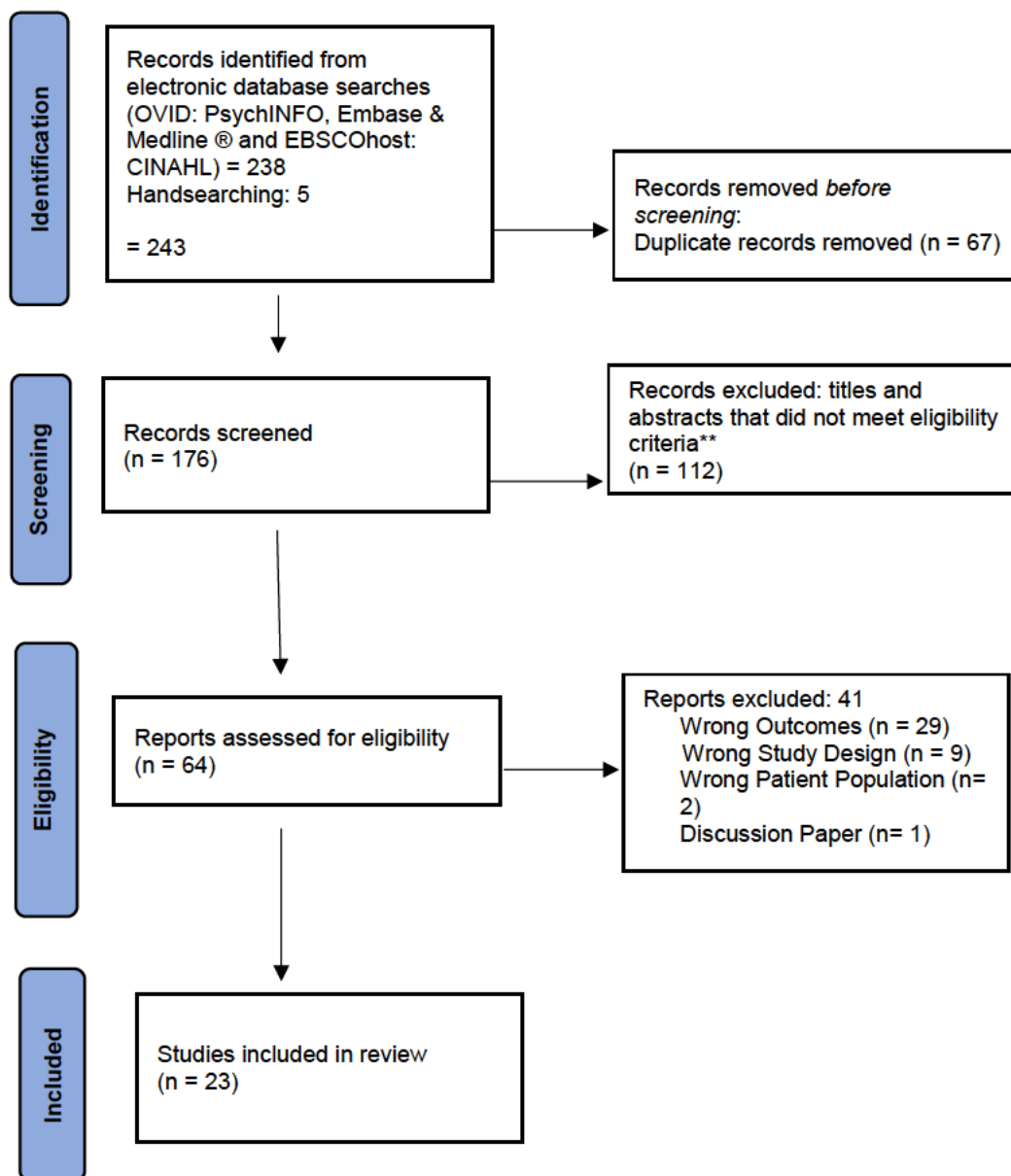
-
1. "disseminated sclerosis" or "ms (multiple sclerosis)" or "multiple sclerosis multiple sclerosis, acute fulminating" or "sclerosis, disseminated" or "sclerosis, multiple"
 2. “anterior horn cell disease" or "familial motor neuron disease" or "lateral scleroses" or "lateral scleroses, primary" or "lateral sclerosis" or "lateral sclerosis, primary" or "lower motor neuron disease" or "motor neuron disease" or "motor neuron disease, familial" or "motor neuron disease, lower" or "motor neuron disease, secondary" or "motor neuron disease, upper" or "motor neuron diseases" or "motor system disease" or "motor system diseases" or "neuron disease, motor" or "neuron diseases, motor" or "primary lateral scleroses" or "primary lateral sclerosis" or "scleroses, lateral" or "scleroses, primary lateral" or "sclerosis, lateral" or "sclerosis, primary lateral" or "secondary motor neuron disease" or "upper motor neuron disease"
 3. "Parkinson*" or "Parkinson Disease" or "Parkinson Disease, Secondary"
 4. "huntington*" or "huntington disease"
 5. “stigma*"
 6. "quality of life"
 7. 1 or 2 or 3 or 4
 8. "anxi*" or "depress*"
 9. (5 and 6) or (5 and 8)
 10. 7 and 9
-

The OVID gateway was used to search PsychINFO, EMBASE, MEDLINE (R) and the Cumulative Index to Nursing and Allied Health Literature (CINAHL) due to their clinical and health focus. The online databases were searched between the years of each journal inception until December 2022. The search was repeated in March 2023 and one additional relevant study was identified. The studies considered relevant for inclusion were selected due

to their clinical, psychological and health focus. The PRISMA guidelines were followed throughout the selection and reporting process (see Figure 1).

Figure 1

PRISMA Flow Diagram



3.3 Data Extraction

In total, the electronic database searches retrieved 243 articles. After duplicates were removed, 176 records were screened. Information relating to the study, country, study type, sample and setting, respondents' gender, age, Stigma & QoL Measure, results summary and additional comments were collated in a data extraction table in Covidence, in order to extract relevant data from studies which met the inclusion criteria (see Table 1). The data was extracted to provide information about the study characteristics and collect relevant information in relation to how the studies were conducted. In order to ensure internal consistency, a second rater (NS) screened a random sample of articles (20%) with full agreement achieved between raters.

3.4 Quality Assessment

According to the CRD (2009), quality criteria should encompass assessment of: the risk of bias; choice of outcome measures; statistical issues; external validity; quality of reporting; and quality of the intervention. The Newcastle Ottawa Quality Assessment Scale (Wells et al., 2000) was utilised to assess the quality of cross-sectional studies and The Newcastle Ottawa Scale for Cohort studies (Wells et al., 2014) was used to assess the quality of longitudinal studies. The second rater (NS) also reviewed 20% of the total number of studies in order to assess for risk of bias. One disagreement was resolved internally and there was full agreement between the raters that all of the sampled studies met eligibility for inclusion.

4.0 Results

The titles and abstracts of all of the identified articles (n= 176) were screened after duplicates were removed. Following a full text review of 64 eligible studies, 23 met criteria for inclusion.

4.1 Characteristics of Included Studies

Out of the reviewed studies, 10 included MS samples (Anagnostouli et al., 2019; Broersma et al., 2018; Cook et al., 2016, Eldridge- Smith et al., 2021; Grothe et al., 2022; Penwell-Waines et al., 2017; Perez-Mirallez et al., 2017; Stepleman et al., 2017; Tworek et al., 2023 & Valvano et., 2016) and 9 studies included Parkinson's patients (da Silva et al., 2020; Eccles et al., 2022; Hechtner et al., 2014; Hou et al., 2021; Islam et al., 2022; Ma et al., 2016; Ma et al., 2019; Meng et al., 2022 & Verity et al., 2020). In addition, two studies addressed MND (Leigh et al., 2021 & Schluter et al., 2018) and two studies explored a sample of patients with HD (Boileau et al., 2020; Thorley et al., 2018). In total, the overall participant sample was 11,220: 7999 patients with MS (71.3%), 2027 with Parkinson's (18.1%), 635 patients with MND (5.7%) and 559 patients with HD (5%). Seven thousand one hundred and ninety respondents across studies were female and 4028 were male. Two respondents did not disclose their gender identity. The aforementioned studies spanned 10 countries, including the USA (n= 11), UK (n=3), Spain (n=1), Greece (n=1), The Netherlands (n=1), China (n=2), Germany (n=1), Italy and Brazil (n=1). One longitudinal study also included a sample of respondents from France, Germany, Italy, Spain and the UK. The total mean of the average ages reported across 23 studies was calculated to be 55.9 years. Out of

the identified studies 17 were recruited from clinical health settings and six were online studies.

Table 2*Characteristics of Included Studies*

Study	Country	Study Type	Sample & Setting	Respondents (Gender)	Age	Stigma/ QoL Measure	Results Summary	Comments
Anagnostouli et al., 2016	Greece	Cross-sectional study	Multiple Sclerosis (MS) patients from an outpatient medical school	342 (231 Females, 111 Males)	43.06 (Mean) 11.35 (SD)	The Stigma Scale for Chronic Illness (24-item) (SSC-I). MSQoL- 54.	Disability levels were the strongest predictor of stigma (standardised b= 0.42, p < 0.001)	
Boileau et al., 2020	USA	Longitudinal Cohort Study (baseline, 12months & 24 months)	Huntington's Disease (HD) patients through HD clinics	479 (Female (58%, Male 42% Females) 315 at 12 months and 277 at 24 months.	49.1 (Mean) 13.1 (SD)	The Neuro-QoL Stigma Computer Adapted Test (CAT) The Quality of Life in Neurological Disorders (Neuro=QoL)	Stigma scores were significantly lower from premanifest symptom participants (M=45.3; SD= 7.4) compared to both early HD (M=51.2, SD= 7.9) and late HD (M=53.2, SD = 9.7; F [2,476] = 39.69; P <.0001). Baseline scores for stigma (M=49.2, SD =8.7) did not differ for either 12 month (M=48.9, SD =8.8) or 24- month scores (M =48.9, SD =8.6. F[2,1068]=0.28; P=.76).	Significant associations between speech difficulties and perceived stigma. Late HD participants were more likely to drop out, limiting generalizability of results.
Broersma et al., 2018	The Netherlands	Cross-sectional study	MS Patients attending MS Centre at University.	185 (125 Females, 59 Males)	60 (Mean) 10.8 (SD) 33-88 (range)	SSCI-I (24 Item, Dutch version). World Health Organisation Quality of Life-abbreviated version.	Individuals who reported more stigma experienced poorer quality of life. Self-stigma was related to all quality of life domains: physical health (B = -0.073, P < 0.01), psychological health (B = -0.089, P < 0.01), social relationships (B	

							= -0.124, $P < 0.01$) and environmental aspects ($B = -0.052$, $P < 0.01$). Enacted stigma was negatively related only to physical health ($B = -0.085$, $P < 0.10$).	
Cook et al., (2016)	USA	Cross-sectional study	Multiple Sclerosis patients completed an online survey.	53 (79% Female)	45.8 (Mean) 11.92 (SD) 23 to 71 (range)	16 items (SSCI), 7 items (Cataldo Lung Cancer Stigma Scale), 5 items (The HIV stigma scale), 4 items (unnamed scale, Earnshaw and Quinn), 2 items (the chronic illness anticipated stigma scale), 2 items (the internalised stigma of mental illness scale. No QOL scale.	Anticipated stigma was associated with greater concealment ($b = 0.51$, $t_{46} = 2.70$, $P = .01$, partial $\eta^2 = 0.14$, 95% confidence interval [CI], 0.13 to 0.89). Internalized stigma was associated with marginally higher levels of concealment ($b = 0.27$, $t_{46} = 1.87$, $P = .07$, partial $\eta^2 = 0.07$, 95% CI, -0.02 to 0.55).	29% of the variance in concealment was predicted by stigma scales.
da Silva et al., 2020	Brazil	Cross-sectional study	Parkinsons Disease patients in Hospital setting	54 (63% Male)	58 (mean) 7.4 (SD)	4 stigma items from the 39 item Parkinson's Disease Questionnaire (PDQ-39)	Greater difficulty in ADL was independently associated with higher self-stigma ($r^2 = 0.12$, b coefficient = 0.42, (95% CI, 0.09-0.76), $P < 0.01$)	Measures of ADL and stigma are both domains of PDQ-39, multicollinearity may be an issue.
Eccles et al., 2022	UK	Cross-sectional study	People with Parkinson's disease, online questionnaire	130 (74 Females, 56 Males)	64.68 (mean) 9.42 (SD) 19-27 (range)	SSCI- 24 item. No QOL Measure.	Significant correlations between felt stigma and depression, anxiety and stress. Felt stigma mediated the link between self-compassion and psychological distress (anxiety [standardised effect size of -0.281], depression [standardised indirect effect of -0.200].	Self-reported diagnosis of Parkinson's disease.

Eldridge-Smith et al., (2021)	USA	Cross-sectional study	MS patients attending an academic medical centre in South Eastern US.	137 (84.7% Females)	45.5 (mean) 10.8 (SD)	Reece Stigma Scale (RSS-MS). Leeds MS Quality of Life Scale (8 item)	Felt stigma was significantly correlated with anxiety and depression ($r = 0.46$), $p < 0.001$ and quality of life LMSQoL ($r = 0.54$, $p < 0.000$). Stigma results significantly varied by type of MS subtype reported, ($F(2,132) = 3.61$, $p = 0.0$).	
Grothe et al., 2022	Germany	Cross-sectional study	Multiple Sclerosis patients, German Outpatient department	101 (74 Female)	46.1 years (mean)	Stigma Scale for Chronic Illness-8 item. Multiple Sclerosis International Quality of Life Questionnaire (MusiQoL).	Enacted stigmatization experience ($b = -0.627$) was a significant predictor of QoL. In a mediation analysis, the direct path between stigmatization and QoL remained significant ($b = -0.8080$, $t = 3.95$, $p < 0.001$).	Mean disease duration of 10.72 years.
Hechtner et al., 2014	France, Germany, Italy, Spain, UK	Cross-sectional study	Parkinson's Disease patients in clinical practice settings via neurologists and geriatricians (in the UK)	817 PD patients 439 Males (53.8%)	66.5 (mean) 9.7 (SD) Italian patients were the youngest, 63.7 (mean), 8.3 (SD), shortest disease duration of a median of 2.2 yrs. UK patients were the oldest, 69.6 (mean), 9.7 (SD). French patients had the longest median disease duration (5.5 yrs)	Parkinson's Disease Questionnaire- 39 (PDQ- 39)	Biphasic dyskinesias (physical symptoms) had a strong detrimental effect on stigma ($B = 12.6$, $p = 0.01$). Strong significant association between PDQ-39 (stigma) and QOL dimension in France only, in relation to on-off fluctuations with mobility and ADL.	Poorer QOL observed in France, Italy and Spain. No strict selection criteria regarding the standardisation of PD diagnosis.
Hou et al., 2021	China	Cross-sectional study	Parkinson's disease patients in a clinical setting.	276 (135 Females and 141 Males)	62.5 (mean) 8.5 (SD)	SSCI- 24 No QoL Measure.	Felt stigma was significantly associated with motor severity, depression and disease duration. The model	Results focused on one hospital sample. Correlated factors

							accounted for 67.3% of the variance. For enacted stigma, the significant correlates were motor severity, tremor dominance and age, which accounted for 23.2% of the variance.	mainly related to disease progression and emotional disorders.
Islam et al., 2022	USA	Cross-sectional study	Parkinson's Disease patients, online survey	196 (113 women, 81 men, 2 unspecified)	64.8 (mean) 8.7 (SD) 43-84 (range)	SSCI PDQ-39 Stigma Scale The Mental Health Consumers' Experience of Stigma Scale	Correlations between the stigma composite score and the individual stigma scales were strong: SSCI ($r=0.74$, $p<0.01$), PDQ stigma subscale ($r=0.68$, $p<0.001$). Depression endorsement correlated with worse QoL more stigma perception per composite score and two stigma scales: SSCI and PDQ stigma scale (all p values <0.001).	79% of sample had additional health conditions which impacted the perception of stigma and QoL. Higher number of health conditions was significantly related to higher stigma perception and lower QoL.
Leigh et al., 2021	United Kingdom	Cross-sectional study	Individuals with a diagnosis of Motor Neurone disease, online survey.	77 (34 Females and 43 Males)	59.1 (mean) 10.6 (SD) 36-83 (range)	SSCI-24 No QoL measure.	Correlations were identified between social support and felt stigma ($r=-.483^{**}$), enacted stigma ($r=-.433^{**}$) and psychological distress (stress, $r=-3.85^{**}$, anxiety, $r=-.399^{**}$ and depression, $r=-.437^{**}$)	Small sample size. Participants average time since diagnosis was 3.5 years
Ma et al., 2016	USA	3 year prospective cohort study (Baseline Data reported only)	Parkinson's Disease patients, Movement Disorder Clinic, Boston University	73 (29 Females, 44 Males)	65.72 (mean age)	SSCI-24. PDQ-39.	Participants who experienced more stigma were more advanced in their disease ($r=.339^*$), more depressed ($r=.667^*$) and had motor difficulties of daily living ($r=.672^*$). Problematic QoL was associated with females, more severe PD, depressive symptoms, more motor	

							difficulties of daily living and more experienced stigma.	
Ma et al., 2019	USA	3 year prospective cohort study (Baseline data reported only)	Parkinson's patients from movement disorder clinics	90 (34 Females, 56 Men)	65.49 (mean), (9.72 SD)	SSCI- 24. PDQ-39.	The indirect effect was marginally stronger in women than in men for both stigma and depression mediation (stigma: coefficient 3.70, 90% CI [.29, 7.44], p =.10; depression: coefficient 1.42, 90% CI [.07, 3.32], p .10). Strong relationship identified between stigma and QoL (r = .82).	
Meng et al., 2021	China	Cross-sectional study	Parkinson's disease patients, Beijing Rehabilitation Hospital	162 (92 Females and 70 Males)	Females: 59.60 (mean), 7.24 (SD) Males: 60.41 (mean), 9.23 (SD)	PDQ-39 (stigma items)	Significant differences between males and females on Parkinson's disease (PD) patient's experiences of stigma impact on quality of life according to the PDQ-39, with females reporting higher stigma prevalence (31.52) compared to males (22.77), p <0.05.	Authors suggest that anxiety may account for the difference in QoL reported amongst males and females and that treatment provisions should vary according to gender.
Penwell-Waines et al., 2017	USA	Cross-sectional study	Multiple Sclerosis patients at Augusta MS Centre	121, 85% Female.	45.4 years (mean) 10.65 (SD)	MS-related stigma scale. Leeds MS quality of life scale (LMSQoL).	Stigma was strongly related to quality of life ($\beta = .206^*$, p < .01).	
Perez-Miralles et al., (2019)	Spain	Cross-sectional study	Primary Progressive Multiple Sclerosis from 11 MS units	55 (56.4% Male)	55.8 (mean) 9.5 (SD)	Stigma Scale for Chronic Illness-8 (SSCI-8) Multiple Sclerosis Impact Scale (MSIS-29).	Stigma scores were correlated with physical ($\rho = 0.464$, p < 0.001) and psychological ($\rho = 0.358$, p = 0.007) MS impact scale sub scores. Stigma predicted concurrent depression (odds ratio = 1.13, p = 0.046).	Findings may be different due to type of MS, administration of different instruments to measure stigma and selection

								processes to recruit patients.
Schluter et al., 2018	UK	Cross-sectional study	MND patients in ALS clinics across the UK.	559, 343 Males (61.4%)	64.72 (mean) 10.66 (SD)	SSCI-8. No QoL measure.	Stigma showed the largest effect on social withdrawal (0.34 (95% CI: 0.27, 0.42).	11.79 (mean) years since diagnosis.
Stempleman et al., 2017	USA	Cross-sectional study	MS Patients MS Outpatient centre	137 (116 Females)	45.5 (mean) 10.8 (SD) 20-74 (range)	MS- related stigma scale. Leeds MS Quality of Life Scale (MSQoL)	"Sustained Identity" was strongly correlated (p < .01) with MS-Related Stigma, MRS (r = -0.55) and MSQoL scale (r= -0.61). "Reactionary identity" was weakly correlated with MRS (r = .27, p <.01 and HADS Depression, r =.20, p < .05).	Study designed to test initial validation of a theory-driven quantitative measure of identity reconstruction in patients with MS. Internal validity issues of measure due to small sample size.
Thorley et al., (2018)	USA	Cross-sectional study	Huntington disease (HD) patients)/online survey	80 individuals with HD (75% Female)	47 years (mean age) HD patients	HDQLIFE/PROMIS Measurement System	A multivariate linear regression model indicated that as stigma scores increased, chorea scores also increased (P = 0.0031). Significantly higher stigma (P < 0.0001) and anxiety (P = 0.0423) were observed among respondents with high chorea than those with low chorea.	Individuals with more advanced disease had more severe problems, with higher levels of anxiety and perceived stigma causing decreased HRQoL.
Tworek et al., (2023)	USA	Retrospective Review	MS Patients, Cleveland Clinic Mellen Centre for MS. Review of data from the Quality of Life in Neurological Disorders (Neuro-QoL). PROMIS Global Health scale (PROMIS-GH)	6760 MS Patients (27.7% Male)	60.2 years (mean age), 8.9 (SD)	Neuro-QoL Stigma Scale. PROMIS Global Health scale.	Stigma was significantly related to physical (beta - 0.390, 95% CI [-0.411, - 0.368]; p< 0.001) and mental health (beta=-0.595, 95% CI [-0.624, -0.566]; p <0.001) domains of the PROMIS global health scale. Anxiety and Depression partially	Patients with the highest NeuroQoL stigma T-score were younger, more likely to be black, single and younger at symptom onset

							mediated the link between Neuro-QoL Stigma and PROMIS-GH Physical and Mental Health.	and had more progressive forms of MS, higher disability levels and lower scores on the PROMIS Global health scale.
Verity et al., (2020)	United Kingdom	Cross-sectional study	229 patients with Parkinson's Disease (PD), UK based PD Charity (online study)	229 (116 Female, 113 Males)	65 (mean) 8 (SD) 29-90 (range)	SSCI- 24 PDQ-8	Stigma was significantly correlated with anxiety (r = 0.49**), stress (r = 0.54**), health-related quality of life (r = 0.68**) and positive affect (r = -0.41**) (** p < .01)	Online recruitment, findings may be different for people who are not highly literate.
Valvano et al., 2016	USA	Cross-sectional study	Multiple Sclerosis Patients, MS Centre	85% Female (108)	45.5 (mean), 10.8 (SD)	MS-related stigma scale. Leeds MS Quality of Life Scale (MSQoL).	Stigma was significantly correlated with cognitive fusion (r = 0.41**), p < 0.01, depression (r= 0.39**), p < 0.01, anxiety (r = 0.388**), p < 0.01 and Quality of life (r = -0.52**), p < 0.01. Stigma indirectly influenced depression (r2= .33), anxiety (r2=.59) and quality of life (r2=.46) through its effect on cognitive fusion.	Focused on only 1 sub-process of psychological flexibility.

Twenty studies included a cross-sectional design and three studies adopted a longitudinal approach. However, two of the longitudinal studies included only baseline data. Twenty-one studies measured overall stigma experiences. Studies varied in relation to terminology used to describe stigma towards the self and quality of life. Two studies referred to *self-stigma* (Grothe et al., 2020; Broersma et al., 2018), another study described *internalised stigma* (Anagnostouli et al., 2019) and in one study, *perceived stigmatic experiences* were reported (Boileau et al., 2020). Four studies reported *felt-stigma* and correlates of neurodegenerative conditions (Eccles et al., 2022; Hou et al., 2021; Ma et al., 2016; Leigh et al., 2021). Of the studies listed, seven also measured *enacted stigma* (Anagnostouli et al., 2019; Broersma et al., 2018; Grothe et al., 2020; Eccles et al., 2022; Hou et al., 2021; Ma-Huing et al., 2016; Leigh et al., 2021). Twelve studies measured QoL (Anagnostouli et al., 2019; Broersma et al., 2018; Eldridge-Smith et al., 2021; Grothe et al., 2022; Hechtner et al., 2014; Ma et al., 2019; Islam et al., 2022; Meng et al., 2022; Pennwell - Waines et al., 2017; Stepleman et al., 2017; Tworek et al., 2023; Valvano et al., 2016) and five studies measured HRQoL (Boileau et al., 2020; Ma et al., 2016; Perez-Mirallez et al., 2019; Thorley et al., 2018; Verity et al., 2020). Two studies measured activities of daily living (ADL's) which can be considered indicators of quality of life in pwPD (Eccles et al., 2022; da-Silva et al., 2020). Other studies investigated disease concealment (Cook et al., 2016), social support (Verity et al., 2020) and social withdrawal (Schluter et al., 2018) in neurodegenerative conditions.

Fourteen studies used correlation and regression analyses to explore links between variables, one study used correlation analyses only and eight studies utilised regression methods only. Eleven studies used the Stigma Scale for Chronic Illness (SSCI) (Rao et al., 2009; Molina et al., 2013) as the primary stigma measure. Five studies utilised the PDQ-39

(Peto et al., 1995), four studies used the MS-related stigma scale (Reece, 2003). Two studies used the SSCI and additional stigma measures, including the PDQ-39 (da Silva et al., 2020) and other stigma measures drawn from various stigma domains across numerous measures including the ‘Cataldo Lung Cancer Stigma Scale’ (Cataldo et al., 2011), the ‘HIV stigma scale’ (Berger et al., 2001), ‘The Chronic Illness Anticipated stigma scale’ (Earnshaw et al., 2013) and the ‘Internalised stigma of mental illness scale’ (Ritsher et al., 2003). One additional study used a stigma measure from the Patient reported outcomes measurement system; PROMIS (Thorley et al., 2018). Cronbach’s alpha was reported in only five studies. Various measures were used throughout across studies to measure anxiety and depression, which included the Hospital Anxiety and Depression Scale (HADS) (Snaith & Zigmond., 1986), Depression Anxiety & Stress Scale (DASS-21) (Anthony et al., 1998), Geriatric Depression Scale (GDS) (Almeida & Almeida.,1999), Beck Depression Inventory (BDI) (Beck et al.,1996) & Beck Anxiety Inventory (BAI) (Beck et al., 1992), Hamilton Anxiety Scale (HAMA) (Hamilton, 1959) and Hamilton Depression Scale (HAMD) (Williams, 1988). The reliability and validity of questionnaires were referenced in the majority of studies.

4.2 Quality Appraisal of Included Studies

The assessment of the quality of the included studies is detailed in Table 2. Five studies were rated as good with reference to their level of quality and 17 were rated as satisfactory. Out of the identified longitudinal studies, only one study (Boileau et al., 2020) reported data across multiple time points. The other two studies reported baseline data from a three year longitudinal study, therefore their quality appraisal was rated against The Newcastle Ottawa Quality Assessment Scale (Wells et al., 2000). There were consistent

strengths across studies, including representativeness of each condition sample, controlling for confounding variables and appropriate statistical reporting in most studies using psychometrically adequate measures in terms of specificity and reliability. The second rater, (NS), reviewed four studies of which two were rated as ‘very good’ (Eccles et al., 2022; Stepleman et al., 2017) and ‘good’ (Perez-Miralles et al., 2019; Schluter et al., 2017) respectively. Whilst all studies reported sample size information, only four studies reported power analysis (Eccles et al., 2022; Islam et al., 2022; Leigh et al., 2021; Verity et al., 2020), limiting the selection quality rating scores across studies. All studies reported p values and r values with only one study reporting odds-ratios (Perez-Miralles 2019). Whilst only two studies reported effect sizes (Islam et al., 2022; Leigh et al., 2021), nine studies reported standardized effect sizes (r^2) (Broersma et al., 2018; Cook et al., 2018; da Silva et al., 2020; Grothe et al., 2022; Ma et al., 2016; Pennwell-Waines et al., 2017; Schluter et al., 2018; Thorley et al., 2018; Valvano et al., 2016).

Table 2

Newcastle Ottawa Quality Assessment Table for Cross-sectional studies (Wells et al., 2000).

Studies	Selection	Comparability	Outcome	Total Score
Anagnostouli et al., 2016	*	**	***	6*
Broersma et al., 2018	**	**	***	7*
Cook, Germano & Stadler., 2016	*	**	**	5*
Da Silva et al., 2020	**	**	***	7*
Eccles et al., 2022	**	**	***	7*
Eldridge- Smith, et al., 2021		**	***	5*
Grothe et al., 2022	*	**	***	6*
Hechtner et al., 2014	**	**	***	7*
Hou et al., 2021		**	***	5*

Islam et al., 2022	*	**	***	6*
Leigh, Simpson & Eccles., 2021	*	**	***	6*
Ma et al., 2016	*	**	***	6*
Ma et al., 2019	*	**	***	6*
Meng et al., 2022	*	**	***	6*
Penwell-Waines et al., 2017		**	***	5*
Perez- Miralles (2019)		**	***	5*
Schluter et al., 2018	*	**	***	6*
Stepleman et al., 2017	**	**	***	7*
Thorley et al., 2018	*	**	**	5*
Tworek et al., 2023	**	**	**	6*
Valvano et al., 2016		**	***	5*
Verity et al., 2020	*	**	***	6*

*NB: Selection max * =5, Comparability max *=2, Outcome max *=3. Study Quality Key: Very Good Studies: 9-10 stars; Good Studies: 7-8 stars; Satisfactory Studies: 5-6 stars; Unsatisfactory Studies: 0 to 4 stars.*

Table 3

Newcastle Ottawa Quality Assessment Table for Cohort Studies (Wells et al., 2014)

Studies	Selection	Comparability	Outcome	Total Score
Boileau et al., 2020	**	**	**	6*

*Note: Good quality: 3 or 4 stars in selection domain AND 1 or 2 stars in comparability domain AND 2 or 3 stars in outcome/exposure domain
Fair quality: 2 stars in selection domain AND 1 or 2 stars in comparability domain AND 2 or 3 stars in outcome/exposure domain
Poor quality: 0 or 1 star in selection domain OR 0 stars in comparability domain OR 0 or 1 stars in outcome/exposure domain.*

4.3 Main study findings

All of the included studies measured stigma and depression, anxiety and/ or QoL across neurodegenerative conditions with visible motor symptoms, reporting significant correlations between the variables of interest. Six studies reported a stronger association between stigma and depression ($r = 0.29, -0.64$), and two studies reported a stronger association between stigma and anxiety ($r = 0.15, -0.58$). Four studies reported associations between stigma and depression only, whilst eight studies did not measure associations between stigma, depression and anxiety, reporting on QoL instead ($r = 0.39, -0.82$). In two studies stigma was correlated with psychological health, but it was unclear whether anxiety and depression underpinned this variable. The strength of the association between stigma, anxiety, depression and QoL varied across studies.

4.3.1 Is Stigma associated with Anxiety and Depression across neurodegenerative conditions?

Each of the 23 included studies reported significant links between stigma, anxiety and depression across neurodegenerative conditions with visible motor symptoms. Amongst pwMS, a strong negative correlation between stigma and the mental health composite score of the MSQoL-54 was reported (Anagnostouli et al., 2019). One study found stigma to be weakly correlated with both anxiety ($r=0.38$) and depression ($r= 0.39$) amongst pwMS (Valvano et al., 2016). Another study reported a negative association between *self-stigma* and

psychological health, however it was not clear whether anxiety and depression underpinned psychological health (Broersma et al., 2018). One study reported a weak link between self-stigma and psychological well-being (Grothe et al., 2022), however, psychological well-being was poorly defined. In another study, stigma was found to positively predict depression (OR = 1.13, 95% CI: 1.00–1.28; p=0.046) (Perez-Miralles et al., 2019). One study of *perceived stigma* in pwMS reported equal correlation coefficients between felt stigma and anxiety and depression ($r = 0.46$) (Eldridge-Smith et al., 2021). Tworek et al., (2023) grouped stigma T-scores into low (T score < 40), average (T score 40-60) and high levels of stigma (T score > 60) against anxiety and depression T scores by each group. Scores were assessed through the Neuro-QoL. Cut off values were chosen, comparing individuals who scored within one standard deviation (10) of the general population mean of 50, to those who scored more than one standard deviation from the general population mean. Higher T-scores were reported for anxiety than depression for pwMS with low, average and high levels of stigma, however anxiety t- scores were only marginally higher than depression t-scores.

In pwPD, stigma was reported to be weakly correlated with depression through the Hospital and Anxiety Depression Scale (HADS), with da Silva et al. (2020) reporting a small standardised effect of stigma on depression ($r^2=0.06$). Two studies reported moderate associations between stigma, anxiety and depression amongst pwPD (Islam et al., 2022; Verity et al., 2020), and overall stigma and *felt-stigma* were strongly correlated with depression (Ma et al., 2016). In another study, *felt-stigma* was reported to be moderately correlated with anxiety (0.58) and depression ($r = 0.64$) (Eccles et al., 2022) In addition, enacted stigma was also moderately correlated with anxiety ($r=0.47$) and depression ($r = 0.41$) in the aforementioned study. In a study of stigma experiences in pwPD from five countries, anxiety/depression rates were reported collectively in their study with the majority of

respondents experiencing ‘some symptoms’ on the generic EuroQol 5-dimension (EQ-5D) questionnaire (Hechtner et al., 2014). Islam et al., (2022) reported a stigma composite score difference of 0.38, $p < .001$ between stigma and depression, with depression assessed through the Beck Depression Inventory- II (Beck et al., 1996). Anxiety was measured through the Beck Anxiety inventory (BAI) (Beck et al., 1993) and Parkinson’s Anxiety Scale (PAI) (Leentjens et al., 2014) with composite scores reported between stigma and anxiety on the BAI (0.35, $p < .001$) and PAI (0.31, $p < .001$).

In research amongst individuals with MND, a small direct effect of stigma on depression was highlighted (Schluter et al., 2018). Leigh et al. (2020) reported that self-stigma was moderately associated with anxiety ($r = 0.53$) and depression ($r = 0.69$), assessed through the Stigma Scale for Chronic Illness. However, a smaller correlation was reported for *enacted stigma* and anxiety ($r=0.24$) and depression ($r= 0.47$). In relation to total stigma score, stigma and anxiety were reported to be moderately correlated ($r= 0.45$) and depression was strongly correlated ($r= 0.67$). In people living with HD, perceived stigma was associated with depression ($\beta = 0.29$) and anxiety ($\beta = 0.15$) (Boileau et al., 2020).

4.3.2 Is Stigma associated with Quality of Life across neurodegenerative conditions?

Stigma was moderately correlated with QoL amongst pwMS in five studies (Anagnostouli et al., 2019; Perez-Mirallez et al., 2019 & Eldridge- Smith et al., 2021;

Valvano et al., 2016) and pwMS who suffered from a higher level of physical limitations reported experiencing more *enacted stigma* (Broersma et al., 2018). Each study used a variety of QoL measures, including the MSQoL-54, MS Impact Scale, Leeds MS QoL scale and the World Health Organisation QoL scale. One study highlighted that *enacted stigma* was a significant predictor of QoL amongst pwMS ($r^2 = 0.74$) (Grothe et al., 2022). Few studies sampling pwMS explored different facets of stigma in relation to QoL. *Self-stigma* was reported to be negatively related to all domains of QoL (physical health, psychological health, social relations and environmental aspects) in pwMS with a small negative effect of self-stigma on social relationships ($b = -0.12$) highlighted through their regression model (Broersma et al., 2018). Eldridge- Smith et al., (2021) reported a t-test which highlighted that stigma scores varied on the basis of marital status/ relationship status ($F(2, 134) = 5.64, p = .004$) and less *perceived stigma* was reported in married or cohabiting individuals ($M = 13.62, SD = 5.48$) than single ($M = 18.04, SD = 11.12$) or separated, divorced or widowed individuals ($M = 18.13, SD = 8.60$).

In pwPD, one study reported that higher stigma was associated with poorer QoL on the PDQ-39 (Ma et al., 2016). A small standardised effect of stigma on activities of daily living in pwPD was reported ($r^2 = 0.12$) (da Silva et al., 2020). Other research highlighted a strong positive correlation between stigma and QoL amongst pwPD assessed through the PDQ-8 ($r = 0.68$) (Verity et al., 2020). A study of pwPD across five countries using the PDQ-39 reported a strong positive correlation between stigma and QoL in France only (Hechtner et al., 2014), whilst stigma and QoL were not correlated in Germany, Italy, Spain or the UK.. Other research reported that stigma was strongly related with QoL ($r = 0.82$) (Ma et al., 2019) and motor-activities of daily living ($r = 0.53$) (Ma et al., 2016). Stigma was associated with greater difficulty in activities of daily living ($r = 0.34$) amongst pwPD (da Silva et al., 2020),

while *felt-stigma* was also associated with greater difficulties in activities in daily living ($r=0.64$) (Ma et al., 2016)

Two studies which explored stigma in samples of people with MND did not report on measures relating to QoL specifically, but reported on indicators of QoL. Schluter et al., (2018) reported that stigma showed the largest effect on social withdrawal (0.34, 95% CI: 0.27, 0.42). Leigh et al., (2022) reported moderate correlations between *enacted stigma* and social support ($r = -0.43$) and *self-stigma* and social support ($r = 0.48$). In people living with HD, *perceived stigma* was reported to be weakly related to QoL in the domain of cognitive function ($\beta = -0.12$, assessed through the Neuro-QoL (Boileau et al., 2020)). *Perceived stigma* was related to physical function/ chorea ($\beta= 0.18$) and speech difficulties ($\beta=0.15$) through the HDQLIFE. In other research in individuals with HD, higher stigma scores were associated with an increasing impact of chorea symptoms (i.e. abnormal, abrupt, unpredictable, non-stereotyped movements) on QoL ($r=0.66$) (Thorley et al., 2018).

4.3.3 Is Stigma correlated with other factors such as age, gender, disease duration or progression?

4.3.3.1 Age

In pwMS, one study reported a weak positive correlation between *internalised stigma* and age ($r =0.19$) (Anagnostouli et al., 2016). Amongst pwPD, two studies reported no

significant associated between stigma and age. However, other studies reported significant negative correlations between *enacted stigma* and age ($r = -0.38$), *self-stigma* and age ($r = -0.31$) (Eccles et al., 2022) and overall stigma and age ($\beta = -0.17$, $p < .001$) (Hou et al., 2021). *Enacted stigma* was also associated with age in females only ($\beta = -0.26$, $p = .009$) (Hou et al., 2021). Islam et al. (2022) reported the stigma composite score was significantly affected by more stigma at a younger age ($r = -0.27$) in a sample of pwPD. In MS, MND and HD samples, no significant associations between stigma and age were reported.

4.3.3.2 Gender

In pwMS, two studies reported non-significant differences between males and females in the reporting of stigma scores (Anagnostouli et al., 2019; Eldridge-Smith et al., 2021). In pwPD, *felt-stigma* was associated with lower motor function ($\beta = 0.47$, $p < .001$) and higher depression scores ($\beta = 0.28$, $p = .002$) in females (Hou et al., 2021). In another study, Ma et al., (2019) reported that facial masking (reduced facial expression) was strongly associated with stigma in females ($r = .70$) and moderately in males ($r = .45$), while another study analysed data by a Mann-Whitney-Wilcoxon test, reporting gender differences between males and females in relation to stigma experiences and QoL through the PDQ-39 (Meng et al., 2022). The data from the Mann-Whitney-Wilcoxon was presented as means and standard deviations. Stigma was reported to impact quality of life to a greater extent in females [$M = 31.52$, $SD = 25.29$] compared to males [$M = 22.77$, $SD = 19.91$], ($p < .008$) in PD (Meng et al., 2022). One of the identified MND studies provided information relating to gender and stigma, however the correlation was non-significant. None of the included studies of HD reported gender differences in stigma prevalence.

4.3.3.3 Disease Duration and Progression

Amongst pwMS, Anagnostouli et al. (2019) reported that overall stigma ($\beta = 3.42$), *internalised stigma* ($\beta = 1.54$) and *enacted stigma* ($\beta = 1.3$) were all associated with higher disability levels. One study of pwPD reported that men who had been lived with PD for a longer duration reported higher instances of *felt-stigma* (Hou et al., 2021). In another study, Islam et al., (2022) found that younger disease onset ($r = -0.35$) and longer disease duration ($r = 0.23$) were related to stigma. Ma et al., (2016) reported that *felt-stigma* ($r = .273$), *enacted stigma* ($r = .391$) and overall stigma ($r = .339$) were associated with disease progression. In addition, Verity et al., 2020 reported a small correlation between stigma and disease duration ($r = 0.24$) amongst pwPD. Neither of the identified MND studies highlighted disease duration or progression as stigma correlates.

In HD samples, one longitudinal study identified through a multilevel model that whilst stigma scores were lower earlier in the trajectory of the condition, they did not differ after 12 months or 24 months and stigma scores were not associated with time of diagnosis or disease duration (Boileau et al., 2020). However, another study of a HD sample reported that more advanced stages of the condition were associated with higher levels of *perceived stigma* ($\beta = 0.22$, $p = .003$) (Thorley et al., 2018).

4.3.3.4 Other demographic variables

Other studies amongst pwMS reported significant correlations between stigma and additional demographic variables such as education levels ($r = 0.18$) and marital status ($r = 0.44$) (Anagnostouli et al., 2019), while occupational status was also correlated with overall stigma ($\beta = -3.92$) and *internalised stigma* ($\beta = -2.59$). In a sample of pwMS, Eldridge-Smith et al., (2021) highlighted through a t-test that stigma scores varied based on employment status [$F(2, 134) = 4.88, p = .009$] and unemployed individuals reported more *perceived stigma* ($M = 18.13, SD = 9.92$) than individuals who were employed ($M = 14.35, SD = 6.24$) or retired ($M = 13.22, SD = 3.89$). Social stigma and disease concealment were reported to be positively correlated amongst pwMS ($r = 0.44$) (Cook et al., 2016). In a sample of pwPD, stigma was also reported to be correlated with social support ($r = .416$) (Ma et al., 2016).

5.0 Discussion

This review examined whether stigma was associated with anxiety, depression and QoL across four neurodegenerative conditions with visible motor symptoms. In addition, it sought to establish whether stigma levels were correlated with factors such as condition specificity, age, gender and disease progression across studies. Fifteen studies included in the review demonstrated associations between stigma, anxiety and depression, with six studies suggesting a stronger association between stigma and depression, while two studies reported stronger associations between stigma and anxiety. Stigma was found to be related to QoL across neurodegenerative conditions. However, the two included MND studies reported on indicators of QoL as opposed to measures of QoL specifically, which limits the generalizability of the association of stigma on QoL across all conditions.

The findings of this review are important as they suggest that stigma is related to emotional distress (anxiety, depression) amongst neurodegenerative conditions with visible motor symptoms. Whilst there may be some inevitable feelings of discomfort as individuals cope with the variability and unpredictability of their symptoms across each condition, stigma may influence the overall burden of illness (Elliot et al., 2019). Despite most studies exploring stigma/ *enacted stigma* overall, there is emerging evidence to suggest that different facets of stigma may be differentially related to anxiety, depression and QoL. *Felt-stigma* was found to be moderately correlated with anxiety and depression in one study amongst pwPD (Eccles et al., 2022) and *self-stigma* was negatively correlated with QoL amongst pwMS (Broersma et al., 2018).

Corrigan et al. (2006a) suggest in *The Progressive Model of Self-Stigma* that stigma internalisation operates in four stages involving the awareness of stereotypes, personal agreement with the stereotypes, application of the stigma to oneself and the resulting harm that ensues. The findings of this review support this model, if anxiety and depression are to represent the resulting harm. Anxiety can be considered to be more situational dependent than depression, therefore it may perhaps have a less pervasive effect on QoL than depression (Prisnie et al., 2018). The incorporation of *self-stigma* towards an individuals' identity may affect their psychological functioning and subjective QoL. (Corrigan et al., 2006a; Fung et al., 2008). Although much of the literature to date has focused on the stigmatisation of mental illness, it has provided a useful framework for organising and advancing research (Sheehan et al., 2022). Research of the different facets of stigma amongst neurodegenerative conditions is still in its infancy and further studies are warranted in order to elucidate further how stigma relates to anxiety, depression and QoL across neurodegenerative conditions.

This review also highlighted that there is some preliminary evidence to suggest that stigma is related to variables such as age, gender and disease duration/progression. It may be useful to ascertain whether stigma is related to individual characteristics across neurodegenerative conditions in order to tailor assessments and interventions to address stigma experiences in a timely manner. For example, amongst pwPD, as *felt stigma* was reported to be related to disease duration amongst elderly males (Ma et al., 2021), attending to individual approaches to protect against *self-stigma* and emotional distress may be useful earlier in the disease trajectory. Similarly, individuals with HD may benefit from intervention attempts to buffer against the effects of stigma and emotional distress earlier, before the progression of the condition (Thorley et al., 2018). Few studies considered the association between stigma and occupational status in this review and future research may benefit from considering how stigma relates to wider social factors.

Stigma has long been cited to be a societal issue (Scambler, 1998) which may vary across countries and be influenced by culture and context (Eccles et al., 2022). In a study of pwPD spanning five countries including Germany, Italy, Spain and the UK, only France reported a strong positive correlation between stigma and health-related QoL, the findings of which could not be explained by clinical characteristics or cultural factors (Hechtner et al., 2014). In a Chinese sample of pwPD, Meng et al. (2022) suggested that as females reported poorer QoL and scored lower on questions relating to stigma, bodily discomfort and emotional wellbeing than males, treatment strategies should be tailored to target these areas, according to gender. This is in the context of previous research highlighting gender to be a determinant of QoL amongst a Chinese population (Hu et al., 2018; Song et al., 2014).

Therefore, treatment provisions in relation to stigma may vary on the basis of culture and context.

It is also noteworthy that individual characteristics may determine the level of emotional distress which is related to stigma experiences, irrespective of the neurodegenerative condition. For example, individuals can react to their perception of stigma with indifference or righteous anger (Corrigan and Watson, 2002), and awareness of stigma does not always result in self-directed negative feelings as illustrated in earlier research exploring stigma experiences amongst people with HIV (Herek et al., 2013). This concept is well illustrated in *The Stigma Resistance Model* (Firmin et al., 2017) whereby stigma resistance is described as an ongoing process in which individuals use their lived experiences to reduce the impact of stigma on a personal, peer or public level. Stigma resistance can be achieved through choosing to ignore stigma through to educating others and challenging stigma on a wider platform. However the persistent nature of stigma experiences in the context of visible motor symptoms may make these difficult to ignore and efforts should be made to reduce stigma on a more public sphere.

5.1 Strengths

This is the first known systematic review to assess stigma experiences across four neurodegenerative conditions with visible motor components and to investigate relevant correlates of interest. It also offers an insight into the association between stigma, emotional distress and QoL amongst individuals with neurodegenerative conditions featuring visible motor symptoms. This review is benefited by a wide variety of satisfactory and good quality

studies, across a large sample of people living with neurodegenerative conditions from different countries. Appropriate measures were utilised throughout this review with good reporting of sensitivity and specificity across studies.

5.2 Limitations

This systematic review contains a number of limitations which warrant consideration. Firstly, the cross-sectional nature of the included studies limits the ability to make causal inferences about the findings of this review and whilst three longitudinal studies were included, two studies only included baseline data. It would be beneficial to explore stigma experiences across different time points of each condition as this would result in a better understanding of how stigma experiences affect individuals at varying stages, allowing for more timely and appropriate intervention.

Furthermore, this review is impacted by the vague terminology used throughout studies in relation to variables of interest. There was variability in the measures used to capture patient reported outcomes across studies, with some authors utilising questions from different measures which may have affected the construct validity of the review. The interchangeable use of stigma related concepts across the literature limits consensus as to the mechanisms of change at play when considering stigma experiences and their impact on anxiety, depression and QoL. The wider definition of QoL incorporated in this review was useful in order to encapsulate various factors which may have contributed towards QoL

across studies. However a number of studies did not provide a definition of QoL, whilst studies which do so reported variability in terms of what constituted QoL. Indeed, the lack of a consensus definition of QoL has been identified as a limitation in medical research (Post, 2014). Some of the included studies did not measure QoL but reported on proxy measures such as social isolation and activities of daily living, which may relate to QoL to some extent, but do not capture the broader construct entirely. Thus, further investigation is necessary in order to ascertain which factors specifically relate to QoL across neurodegenerative conditions. Few studies reported effect sizes or odds ratios limiting the ability to pool study results and a meta-analysis was not possible due to the heterogeneity of data.

The combining of neurodegenerative conditions with visible motor symptoms may also be limiting given that there are likely to be differences in relation to how stigma is related to each condition. Individuals in the later stages of HD or MND for example may require support and assistance in daily activities, which may result in labelling and discrimination. Motor symptoms which are not associated with MS (e.g. choreiform movements) may also be perceived more negatively by others. Future research may benefit from synthesising research pertaining to each condition separately once there are enough studies relating to stigma across neurodegenerative conditions to be able to do so.

5.3 Clinical and Social Implications

Given the highlighted association between stigma, anxiety and depression across neurodegenerative conditions, this review indicates the importance of attending to the stigma experiences and emotional distress amongst people with neurodegenerative conditions in

treatment settings. For example, considering the role of stigma experiences and shame in the context of functional impairment and chorea symptoms in HD (Thorley et al., 2018) may be helpful clinically, to appropriately tailor treatment plans to meet the needs of individuals. Similarly, attention drawn to the role of social support in MND (Leigh et al., 2021) may be useful to protect against the effects of self-stigma. There may be factors which aid resilience and prevent enacted stigma experiences from being incorporated onto the identity of the individual; whilst some may argue that stigmatised individuals should not hold the burden of alleviating stigma (Boileau et al., 2020), individual treatment programmes may empower clients with neurodegenerative conditions to foster their natural resilience to provide a buffer against stigmatic experiences. Acceptance and Commitment Therapy (ACT) is an example of an evidence based third wave therapy that aims to increase psychological flexibility, promote present moment awareness and work in the direction of which the clients considers valuable as opposed to modifying dysfunctional thoughts (Hayes et al., 1999). A recent systematic review of ACT to target stigma has demonstrated initial promising results in relation to targeting stigmatic experiences (Gómez 2020). Furthermore, a critical review of *self-stigma* reduction amongst people with mental illness documented the efficacy of individual interventions that aim to alter maladaptive beliefs, enhance skills development and improve self-esteem and coping skills (Mittal et al., 2012). Few studies have addressed *self-stigma* amongst neurodegenerative conditions and research to address this gap is warranted.

This review has highlighted the presence of *enacted stigma* across studies, suggesting educational provisions targeting stigma at a societal level may also be beneficial to help to teach individuals towards how stigma relates to emotional distress and QoL. Yet recent advancements in theoretical stigma frameworks (e.g. The Health Stigma and Discrimination Framework; Stangl et al., 2019) suggest that the field of stigma research move away from

viewing stigma as something an individual imposes on others, towards emphasising the broader social, economic forces that drive stigma. Other research argues that due to the negative and ingrained effects of structural stigma (whereby experiences of those who stigmatise others and those who are stigmatised combine to influence illness prejudice in a broader cultural sphere), stigma reduction efforts should target population based approaches (Corrigan and Fong, 2014). A primary goal of this framework is to allow stigma researchers across disciplines to standardise measures and build effective interventions (Stangl et al., 2019). Given that a goal of the intersectoral global action plan on epilepsy and other neurological conditions 2022-2031 is to reduce the stigma, impact and burden of neurological disorders and to improve QoL of people with neurological disorders and their families (WHO, 2022), it is clear that stigma reduction is a recognised global issue and continued efforts to critically review relevant studies as they emerge are warranted.

5.4 Conclusions

This systematic review identified that stigma is associated with anxiety, depression and QoL across four neurodegenerative conditions with visible motor components. These findings highlight the clinical importance of addressing and measuring stigma experiences during routine health consultations. Future studies would do well to incorporate more longitudinal designs, investigating different facets of stigma and links between demographic and social factors, which may aid in the development of effective coping tools to inform individualised interventions. Governing bodies may benefit from the prioritisation of stigma research and enactment of policies which target the reduction of stigma on a wider level. Meanwhile, in order to promote positive health outcomes amongst individuals living with

neurodegenerative conditions, individual approaches may be warranted in order to protect against the harmful effects of stigma.

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Self-Stigma, Psychological Flexibility, Emotional Distress and Quality of Life in People living with Multiple Sclerosis.

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1.0 Abstract

This study investigated whether psychological flexibility and emotional distress serially mediated the association between *self-stigma* and quality of life (QoL) in people with Multiple sclerosis (pwMS). A cross-sectional, quantitative design was utilised, with an online survey distributed to 208 people with Multiple Sclerosis. Participants completed the Hospital Anxiety and Depression scale (HADS), the CompACT, the Leeds MS Quality of Life Scale and the Stigma Scale for Chronic Illness Eight item (SSCI- 8). Correlational, Regression and Mediation Analyses were used to investigate variables of interest. The results highlighted statistically significant associations between *self-stigma*, psychological flexibility and QoL, with higher levels of *self-stigma* associated with lower psychological flexibility, higher levels of emotional distress and poorer QoL. Psychological flexibility and emotional distress independently and serially mediated the link between *self-stigma* and QoL amongst people with MS. This study highlighted that *self-stigma*, psychological flexibility and emotional distress predicted QoL. Interventions which aim to target psychological flexibility and emotional distress levels may be useful to combat the effects of *self-stigma* on QoL amongst pwMS.

Keywords: Multiple Sclerosis, Stigma, Psychological Flexibility, Emotional Distress, Quality of Life

2.0 Introduction

Multiple Sclerosis (MS) is a chronic, incurable, neurodegenerative disorder of the central nervous system (CNS) (Wallis et al., 2020) which has been reported to affect 2.3 million people worldwide (Browne et al., 2014). The symptoms of MS can be unpredictable and broad, covering a wide range including vision impairment, sensorimotor deficits, paralysis, balance problems and bladder and intestinal dysfunction (Ajdacic-Gross et al., 2021). Physical symptoms which are visible to others can result in individuals experiencing stigma; this is defined as an “attribute that is deeply discrediting”, whereby a stigmatised person is reduced “from a whole and usual person to a tainted, discounted one” (Goffman., 1963, p.3). Such adverse social experiences may result in *internalised stigma* or *self-stigma* which relates to negative stereotypes about illness which are accepted and incorporated into the identity of an individual (Corrigan et al., 2006a). *Self-stigma* may lead to negative feelings and emotional reactions as the individual agrees with the negative stereotype and apply it towards themselves. For example, people with schizophrenia have been found to self-stigmatise through their internalisation of the negative views of others (Fung et al., 2008). When an individual internalises stigma this can lead to feelings of hopelessness and determine the *why try effect*, where the pursuit of life goals in relation to work, education or relationships are given up (Corrigan et al., 2009). Stigma (Anagnostouli et al., 2019; Perez-Mirallez et al., 2019 & Eldridge- Smith et al., 2021; Valvano et al., 2016) and *self-stigma* (Broersma et al., 2018) have been found to correlate with quality of life (QoL) amongst people with Multiple Sclerosis (pwMS), however, the mechanisms by which *self-stigma* relates to QoL amongst pwMS are currently unknown. *Self-stigma* has been previously related to deteriorations in QoL and increased symptom severity and suicidality in schizophrenia (Vrbova et al., 2018). It can represent a substantial barrier to recovery as

people refrain from seeking help and engaging with mental health services (Yanos et al., 2008; Corrigan, 2016).

One mechanism which may link *self-stigma* and QoL is psychological flexibility. Acceptance is a core component of psychological flexibility, which relates to the ability to notice and accept the presence of thoughts and feelings, engaging in the present moment and acting in line with personal values (Cherry et al., 2021). Psychological flexibility is a fundamental determinant of health due to the major contribution it makes towards the maintenance of psychological health and wellbeing (Kashdan & Rottenberg, 2010). Previous research of psychological flexibility amongst pwMS focused on a single component of psychological flexibility (cognitive fusion) (Valvano et al., 2016), although the authors did not measure the broader psychological flexibility process, omitting behavioural awareness or valued action.

The broader psychological flexibility process was studied more recently amongst individuals' with secondary progressive MS, with greater psychological flexibility associated with lower distress and higher QoL (Meek et al., 2022). The association between *self-stigma* and psychological flexibility has yet to be explored amongst pwMS. The purpose of this current study was to investigate whether psychological flexibility and emotional distress (depression, anxiety) represented indirect mechanisms by which *self-stigma* predicted QoL amongst pwMS. Depression and anxiety have been highlighted to be risk factors for lower QoL amongst pwMS (Gil-Gonzalez et al., 2020; Gaynes et al., 2002) and may warrant consideration as underlying mechanisms in the link between *self-stigma* and QoL. A clearer

understanding of the factors which predict QoL amongst pwMS may help to inform individual treatment interventions and influence wider service provisions.

2.1 Quality of Life (QoL)

The World Health Organisation define *quality of life* (QoL) as an individual's perception of life in the context of their culture and value system, in relation to their goals, expectations, standards and concerns (WHO, 1993). Whilst this term has been commonly adopted by researchers, a myriad of QoL definitions exist, with the literature limited by a lack of a consensus definition (Post, 2013). A similar concept, *Health-related quality of life* (HRQoL), refers to an individuals' account of their functioning and well-being in physical, mental and social aspects of life (Kaplan & Hays, 2022), while HRQoL is said to represent the link between QoL and individual health status (Ysraelit et al., 2018). QoL is often compared and used interchangeably with HRQoL in the literature which can spark confusion, particularly as QoL is argued to extend beyond health-status (Karimi & Brazier, 2016; Tennant & McKenna, 1995). For the purposes of clarity this study will adopt the term QoL.

The extent to which QoL measures are client centred and accurately measure the QoL of individuals has been highlighted, with the development of individualised measures of QoL considered to be best practice (Carr & Higginson, 2001). The challenge of QoL measurement is both the subjective measure of life satisfaction by clients and objective measure of health status by physicians (Boyer et al., 2014). QoL measurement is useful as patient reported outcomes enable communication between the client and physician in response to treatment (Black, 2013). Determinants of QoL can vary between clinicians and clients, with neurologists placing greater emphasis on the importance of physical function and role

limitations in QoL compared to pwMS who prioritise vitality as an indicator of QoL (Ysrraelit et al., 2018). This study will adopt an MS related QoL scale, which was devised through co-production with pwMS in order to capture the relevant construct of interest.

2.2 QoL in Multiple Sclerosis

PwMS often report poorer QoL in comparison to the general population (Chruzander et al., 2014; Pittock et al., 2004). MS is typically diagnosed around the ages of 20-40 years old (Rejdak et al., 2010), a time which often coincides with key developments in an individual's professional and personal life. The effects of MS can be diverse, impacting on financial stability, employment, family relationships, life goals and well-being (Dennison et al., 2009). Recent research has explored predictors of QoL, with factors such as unemployment associated with lower QoL amongst pwMS (Schmidt & Jöstingmeyer., 2019). In a global survey of 1075 pwMS, over 50% reported daily activity limitations due to fatigue, muscle problems and physical weakness which impacted on factors including self-esteem, maintaining relationships and career progression (Bass et al., 2020). It is noteworthy that not all pwMS report poor QoL and some individuals experience relatively good QoL over the course of their lives (Pittock et al., 2004), while others can experience pervasively poorer QoL over many years (Chruzander et al., 2014). Overall, it is not MS in isolation which impacts on QoL, but rather the interplay of biological, psychological and social factors that affect an individual's functioning (Homayuni et al., 2021; Strober 2018). As such it is necessary to investigate modifiable mechanisms which may underpin QoL in order to tailor appropriate treatment provisions to provide optimal care for pwMS.

2.3 Stigma & QoL

Stigma represents an understudied psychosocial factor which may be relevant to QoL amongst pwMS. There are various different types of stigma which provide a useful framework for the organisation of research (Sheehan et al., 2022). The Social Cognitive Model posits that stigma is a complex phenomenon which is comprised of stereotypes, prejudices and discrimination (Sheehan et al., 2017) and stigma can cause an individual to be seen as unworthy, undesirable or sometimes dangerous (Engebretson, 2013). *Enacted stigma* occurs when an individual experiences discrimination on the grounds of their perceived inferiority or unacceptability (Scambler & Hopkins, 1986) and such experiences may lead individuals to undertake manipulative behavioural strategies in order to cope with the effects of discrimination (Grytten & Maseide, 2005). In a cross-sectional survey exploring stigma amongst pwMS (n=101), 57.3% reported experiencing *enacted stigma* at least once (Grothe et al., 2021). Stigma was reported to be highly prevalent in cross-sectional research amongst pwMS and related to poorer QoL (Broersma et al., 2018; Valvano et al., 2016). Stigma may influence the propensity towards disease concealment, with stigmatised people tend to seek isolation, which may make them less likely to access health services, which may lead to poorer health-outcomes and poorer QoL (Cook et al., 2016; Fung et al., 2008). However, at present, the exact mechanisms which link stigma and QoL amongst pwMS are currently unknown and warrant further assessment.

2.4 Self-stigma & QoL

One facet of stigma which may relate specifically to QoL amongst pwMS is *self-stigma*. Individuals who identify as belonging to a stigmatised group may begin to direct negative attitudes towards themselves in the form of *self-stigma* (Krafft et al., 2018). *Self-stigma* relates to the “shame, evaluative thoughts, and fear of enacted stigma that results from an individual's identification with a stigmatized group that serves as a barrier to the pursuit of valued life goals” (Luoma et al., 2008, p.150). The Progressive Model of Self-Stigma proposes that *self-stigma* operates in stages, whereby individuals become aware of enacted stigma, agree with the negative stereotypes, apply them towards themselves in form of *self-stigmatization* which results in emotional harm (Corrigan & Rao, 2006a).

A preliminary study exploring differential properties of HIV stigma mechanisms, found that *self-stigma* was associated with affective and behavioural health and wellbeing, in relation to people's acceptance of HIV, their helplessness regarding their diagnosis and perceived benefits of their condition (Earnshaw et al., 2013). *Self-stigma* has also been a targeted treatment mechanism in reducing the negative impacts associated with substance use disorder (da Silveira et al., 2018) and obesity (Palmeira et al., 2016). PwMS with higher levels of *self-stigma* have reported to experience poorer QoL (Grothe et al., 2021; Broersma et al., 2018). Anagnostouli et al. (2016) reported lower QoL scores amongst pwMS because of fear of discrimination and shame, rather than actual experiences of discrimination experiences. *Self-stigma* is a potentially important factor in treatment to research due to its damaging effects on self-esteem and self-efficacy (Corrigan, 2016). *Self-stigma* is argued to be more amenable towards intervention than *enacted stigma* since *self-stigma* is more robustly linked to more intrinsic mechanisms that may be modifiable in treatment (Latalova et al., 2014). Therefore the current study will focus on examining the mechanisms that are associated with *self-stigma* and QoL.

2.5 Self-stigma & Psychological Flexibility

Psychological flexibility may represent an intrinsic mechanism which links self-stigma and QoL. Psychological flexibility can be defined as an individual's “ability to contact the present moment more fully as a conscious human being, and to change or persist in behaviour when doing so serves valued ends” (Hayes et al., 2006, p. 6). It has proved to be a difficult concept to define, with 23 flexibility constructs reported across 203 studies, which has led to a lack of conceptual clarity and varied research across disciplines (Cherry et al., 2021).

Psychological flexibility is the main construct of Acceptance and Commitment Therapy (ACT) which conceptualises human psychological suffering as a function of attempts to avoid unwanted private experiences and aims to promote value consistent behaviour and increase an individual's psychological flexibility (Hayes et al., 1999). Psychological flexibility is not unique to ACT, having been studied in neuropsychology research since the mid-20th century (Schultz & Searleman, 2002), however, the study of previous psychological flexibility processes did not account for how an individual flexibly responds to emotional experiences in line with their valued goals (Doorley et al., 2020). A plethora of ACT research in recent years has popularised psychological flexibility and incorporated the adoption of an individual's behaviour in line with values, thus the definition by Hayes et al. (2006) will be adopted herein.

Clients experiencing *self-stigma* are taught to engage mindfully with feelings of shame and diffuse from them without trying to change or avoid difficult feelings (Krafft et al., 2018). In a meta-analysis across 16 studies, Krafft et al., (2018) identified that stigma had a relatively large and stable correlation with psychological flexibility. However, the few studies which targeted *self-stigma* were limited by small sample sizes and no study involved pwMS.

Previous psychological flexibility measurement across studies has been reported to be inadequate, with measures such as the Acceptance and Action Questionnaire II (AAQ-II) (Bond et al., 2011), targeting sub processes as opposed to psychological flexibility overall. (Cherry et al., 2021). The compACT (Francis et al., 2016) has been developed in order to address some of the limitations of existing psychological flexibility measures and it demonstrates good psychometric properties. This study will attempt to provide a more comprehensive measure of psychological flexibility through utilising the compACT.

Whilst psychological flexibility is an important mechanism to consider in relation to the pathway between *self-stigma* and QoL amongst pwMS, other psychological factors may also warrant consideration.

2.6 Self-Stigma & Emotional Distress

Emotional Distress (anxiety and depression) may also be pertinent in the link between *self-stigma* and QoL. Anxiety and depression levels, described in this study collectively as *emotional distress*, have been vaguely conceptualised as distress in previous literature in an MS sample (Meek et al., 2022). There is a 50% lifetime prevalence rate of depression

amongst pwMS, which is 2-3 times higher than the general population (Cadden et al., 2018; Patten et al., 2017). Longitudinal research has suggested that depression predicted poorer QoL up to ten years later amongst pwMS. (Chruzander et al., 2014). Recent research has highlighted that elevated depression and anxiety levels are associated with more frequent reporting of internalised shame amongst pwMS (Barta & Kiropoulos., 2023). Perceived discrimination has also been associated with earlier onset of disease and depressive symptoms (Ochoa-Morales et al., 2021). For example, in a recent sample of 6760 pwMS, overall stigma was associated with increased depression levels and lower QoL (Tworek et al., 2023), however the study did not account for the self-stigma experiences of pwMS.

Anxiety is also common amongst pwMS with a reported prevalence rate of 45% (Wallis et al., 2020; Wood et al., 2012). Anxiety can be considered to be more situational dependent than depression and may have a less pervasive effect on QoL (Prisnie et al., 2018). This is particularly true of panic symptoms which may fluctuate during the course of the assessment. Nonetheless, individuals who self-stigmatise may experience elevated anticipatory anxiety because they fear rejection (Vauth et al., 2007). *Self-stigma* is correlated with anxiety in previous studies amongst individuals with anxiety disorders and schizophrenia (Ociskova et al., 2015; Vauth et al., 2007; Corrigan et al., 2009), however none of these studies focused on pwMS. *Perceived stigma* has been associated depression and anxiety amongst pwMS (Eldridge-Smith et al., 2021), however the study used a measure of stigma that had not previously been validated. Other research amongst pwMS has reported a negative *association* between self-stigma and psychological health, although psychological health was derived from a subscale of Dutch Quality of Life scale and it was not clear which psychological health related to anxiety, depression or any other factors (Broersma et al.,

2018). Further research is warranted to examine how *self-stigma* relates to emotional distress amongst pwMS.

2.7 Psychological Flexibility & Emotional Distress

Psychological flexibility and emotional distress may represent mechanisms which link *self-stigma* and QoL amongst pwMS. Individuals with greater psychological flexibility may experience less emotional distress as they engage in less experiential avoidance (attempts to suppress unwanted internal experiences) in an attempt to accept difficult emotions (Krafft et al., 2018; Kashdan & Rottenberg, 2010). Previous cross-sectional studies have reported consistent associations between psychological flexibility and emotional distress in people with moderate to severe anxiety and depression (Forman et al., 2007; Fledderus et al., 2013; Pots et al., 2016), while longitudinal research has highlighted the role of psychological flexibility in reducing depressive symptoms in an outpatient sample, albeit in a group format (Østergaard et al., 2020).

In a recent study of pwMS, increased psychological flexibility was associated with lower emotional distress (depression, anxiety) and greater QoL (Meek et al., 2022). This was the first known study assess psychological flexibility in a sample of pwMS, despite similar findings in a healthy adult sample (Dawson & Golijani- Moghaddam, 2020). In addition, the psychological flexibility subscale ‘openness to experience’ and depression had a strong negative association, which may have been due to the reduction in activity being a symptom of depression, leading to individual’s engaging in less valued activities (Meek et al., 2022). Higher psychological flexibility was associated with lower depressive symptoms, however, this sample related to individuals’ with secondary progressive MS only and did not address

stigma. The mechanisms which link *self-stigma* and QoL in MS are currently unknown and warrant further investigation.

2.8 Project Aims and Hypotheses

In summary, this study aims to explore whether *self-stigma* is associated with lower QoL amongst pwMS. This project aimed to examine the association between *self-stigma*, psychological flexibility, emotional distress and QoL amongst pwMS. It is proposed that higher *self-stigma* will be associated with lower psychological flexibility, higher emotional distress and poorer QoL. Furthermore, this study aimed to assess whether psychological flexibility and emotional distress represented both individual and serial mechanisms from *self-stigma* to QoL in a cross-sectional sample of pwMS. In order to account for the potential influence of *enacted stigma* on the serial mediation model, *enacted stigma* was added as a covariate.

The hypotheses for this study were:

- 1) Higher levels of *self-stigma* will lead to poorer QoL amongst pwMS.
- 2) Psychological flexibility will mediate the link between *self-stigma* and QoL amongst pwMS.
- 3) Emotional Distress (depression, anxiety) will mediate the link between *self-stigma* and QoL amongst pwMS.

- 4) Psychological flexibility and emotional distress will serially mediate the link between *self-stigma* and QoL.

3.0 Methods

3.1 Participants and Recruitment

Participants comprised 208 pwMS. Whilst 262 responses were collected through an online survey, 54 responses were removed due to non-completion. As the survey utilised forced choice entry; there were no missing values. In order to determine the sample size required for the serial mediation study, power analyses were conducted using a Monte Carlo power analysis simulation for indirect effects, available online from: https://schoemanna.shinyapps.io/mc_power_med/ (Schoemann et al., 2017). To the best of the author's knowledge no previous study has used serial mediation analysis to test the indirect effect of the proposed pathways ($a_1d_2b_2$). Therefore, the estimated effect size of each pathway was informed by correlations from previous research (Valvano et al., 2016; Krafft et al., 2018) A meta-analysis found that stigma had a large and stable correlation with psychological flexibility across 16 studies (Krafft et al., 2018). Therefore the estimated correlations of each pathway were entered into the online Monte Carlo power analysis tool, based on a bias corrected bootstrapping estimate of 5000 samples. The results indicated that in order to achieve the target power of 0.8, a sample size of 217 would be required.

Individuals who met eligibility criteria were asked to provide some basic demographic information pertaining to their age, gender, ethnicity, MS subtype (relapse remittent, progressive or secondary progressive), occupation and years since diagnosis.

Table 1 highlights the characteristics of the included sample.

Table 1

MS Sample demographic characteristics

MS Sample	n	%
Female [F]	171	82
Male [M]	37	18
Ethnicity		
White	201	97
Black	1	.5
Asian	1	.5
Other	5	2
MS Subtype		
Relapse remitting	156 135 [F], 21 [M]	86% [F], 13% [M] 75
Primary Progressive	10 7 [F], 3 [M]	70% [F], 30% [M] 5
Secondary Progressive	40 27 [F], 13 [M]	67% [F], 33% [M] 19
Progressive Relapsing	2 2 [F]	1 100% [F]
Employed	114	55

Unemployed	94	45
	<i>Mean</i>	<i>Range</i>
Age	47.9	58 (20-78)
Time Since Diagnosis (years)	10.2	41 (.08-40.8)

97% of the individuals who completed the survey were white caucasian and 82% of respondents were female. The mean age of respondents was 47.9 years and average duration of time living with MS was 10.21 yrs. 75% of respondents lived with relapse remittent MS and 19% lived with secondary progressive MS.

Recruitment was carried out via two pathways. The first pathway related to online engagement where the study link was posted by third sector organisations, including the MS Trust, MS Society & Ann Rowling Clinic. Each organisation was provided with study information to disseminate to eligible respondents via their online support group webpages. The second pathway involved an NHS Scotland Neurology Clinic distributing the study link to pwMS following their routine health appointment. The online survey was hosted on the platform Qualtrics, however, the option of hard copy completion in clinic was provided by MS nurses. The inclusion criteria for eligibility involved respondents being aged 16+ and fluent in English, with a confirmed diagnosis of Multiple Sclerosis (Relapse Remitting, Primary or Secondary Progressive). Participants were excluded from the study if they had another neurological or psychiatric condition such as dementia, epilepsy or schizophrenia. Any individuals who were unable or unwilling to provide informed consent and who had a learning disability were also excluded.

3.2 Procedure

This study utilised a cross-sectional, survey-based design to examine the links between stigma, psychological flexibility, emotional distress and QoL in pwMS. The London Westminster Research Ethics Committee granted ethical approval through IRAS and sponsorship was provided by the University of Edinburgh's College of Arts, Humanities and Social Sciences Research Governance Coordinator (see appendix B). Eligible participants completed a survey which was hosted online via the survey platform 'Qualtrics'. Due to Covid-19 restrictions, in person completion of surveys could not be carried out. Consequently, all respondents completed the online survey only. The survey was live between January-April 2022. Individuals were required to confirm that they had read and understood the participant information sheet and give consent for their data to be used in the research, prior to beginning the survey. They were informed that they were required to provide responses to each question and non-completion was considered to be withdrawal from the study.

3.2.1 Leeds MS Quality of Life (LMSQoL) (Ford et al., 2001)

In order to measure QoL in pwMS, the LMSQoL (Ford et al., 2001) scale 8 item version was selected. In this measure, responses range on a scale from 'Not at all' (0) to 'Most of the time' (3) on a 4 point Likert scale and with higher scores reflecting poorer QoL. The scale has a maximum score of 24. The LMSQoL contains questions designed to assess well-being and aspects of daily functioning such as relationships, energy levels and worry

(Ford et al., 2001). The LMSQoL is easy to administer and has good psychometric properties, demonstrating no floor or ceiling effects, good internal consistency (0.79) and test reliability (0.85) (Ford et al., 2001). The LMSQoL has demonstrated good content validity amongst samples with MS (Ensari, Motl & McAuley., 2016) and it demonstrated good internal consistency in this study ($\alpha = .833$).

3.2.2 Stigma Scale for Chronic Illness ‘SSCI-8’ (Molina et al., 2013)

In order to measure *self-stigma* the ‘Stigma Scale for Chronic Illness’ ‘SSCI-8’ (Molina et al., 2013) was selected. This brief, 8 item version measures stigma across neurological conditions, including MS. Two items relate to *self-stigma*, for example ‘*I felt embarrassed about my illness*’. Of the 8 items, 6 items relate to *enacted stigma*. Response items range across a five point Likert scale from ‘Never (1) to Always (5)’, with a greater summation of scores indicative of more stigmatic experience. The SSCI-8 (Molina et al., 2013) has been found to possess adequate reliability/internal consistency for *enacted* and *self-stigma* (Cronbach’s alpha, $\alpha = 0.85$ and 0.87 respectively) and convergent validity. The reliability for both subscales together in this study was very good ($\alpha = .892$). The *self-stigma* items were utilised as part of the study hypotheses with *enacted stigma* added as a control variable.

3.2.3 CompACT (Francis et al, 2016)

In order to measure psychological flexibility- the ComPACT (Francis, 2016) was selected. This 23 item questionnaire's responses range across a seven point Likert scale from Strongly Disagree (0) to Strongly agree (6), for example *'I can identify the things that really matter to me in life and pursue them'*. Greater responses are indicative of higher psychological flexibility levels. Twelve items are reverse-scored before summation. Scores were derived by the summation of the responses of 3 subscales (Openness to Experience, Behavioural Awareness and Valued Action) the scale as a whole (CompACT total score). The CompACT has been previously used in an MS sample and demonstrated good reliability/internal consistency ($\alpha= 0.90$) and content validity (Meek et al., 2022). The total compACT score was utilised and cronbach's alpha for this study was very good ($\alpha=.892$).

3.2.4 Hospital Anxiety & Depression Scale (HADS) (Zigmond & Snaith, 1983).

In order to assess levels of emotional distress (depression, anxiety) in pwMS, the HADS (Zigmond & Snaith, 1983) was used. The 14 item measure has been previously validated for use in MS populations and demonstrated good sensitivity and specificity (Honormad & Feinstein, 2009; Watson et al., 2014) It is the most common measure of emotional distress amongst MS Populations (Meek et al., 2022). The measure provided two scales: one measured anxiety (HADS-A) and the other scale measured depression (HADS-D) rates (7 items each/ 14 in total). Scores range across a 4 point Likert scale from 0-3, with a cutoff score of 8 or above on either subscale indicating possible emotional distress. The measure has a maximum score of 42 (21 depression/ 21 anxiety). The HADS-T total score

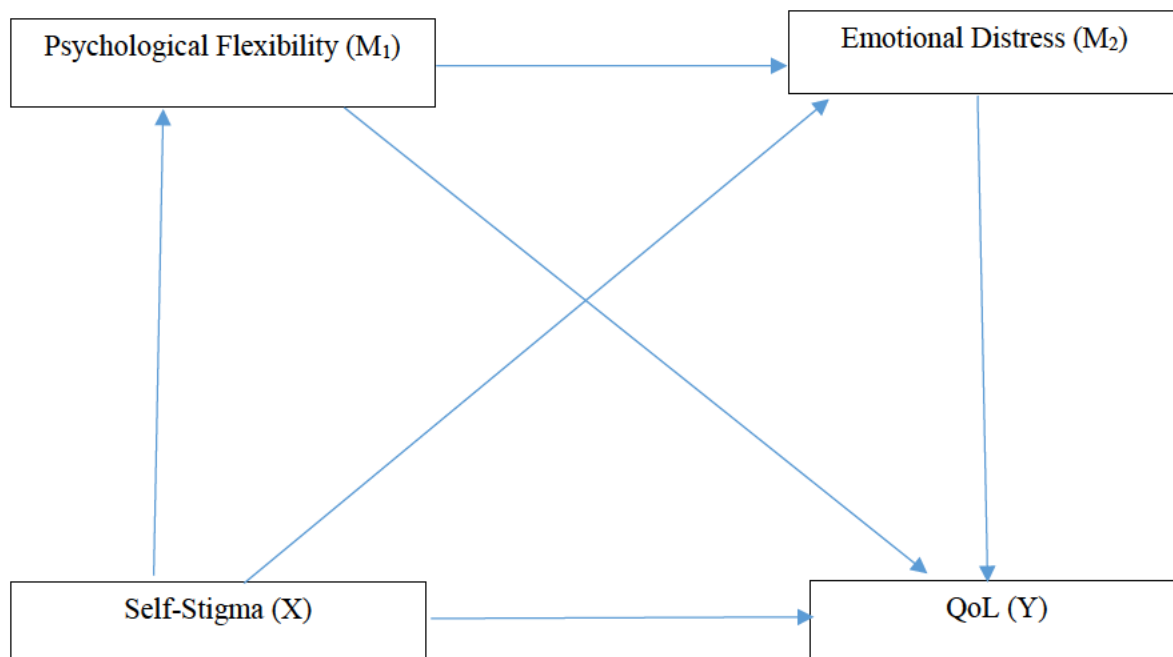
was used as a measure of total emotional distress across the depression and anxiety scale. The HADS demonstrated good internal consistency in this sample ($\alpha = .855$).

3.3 Data Analysis

Statistical analyses were conducted using IBM SPSS Statistics for Windows (Version 25). Preliminary exploration of the data was conducted through the use of descriptive statistics, Pearson correlations and t-tests. Correlational analyses were conducted to ascertain the associations between variables of interest. Regression analysis was utilised to determine whether *self-stigma* predicted QoL. In order to ensure that regression assumptions were not violated, the data was examined for a normative distribution of residuals, homoscedasticity, multicollinearity and to ensure a linear relationship between the predictor and outcome variable (Field, 2013). The dataset was initially examined for outliers, skewness levels and kurtosis. Histograms and boxplots were utilised to detect potential outliers in the dataset for the compACT, LMSQoL, SSCI-8 and HADS scales. It was predicted that higher levels of *self-stigma* would lead to lower levels of psychological flexibility and higher emotional distress which in turn would lead to poorer QoL amongst pwMS. In order to test this predicted model, simple and serial mediation analyses were run using Hayes' Process Tool (model 6) (Hayes, 2022) employing a bias-corrected bootstrap model, based on 5000 samples.

Figure 1

Conceptualised Model of the study Hypotheses



4. Results

4.1 Descriptive Data

The mean, standard deviation, range, skewness and kurtosis scores for the study variables are highlighted in Table 2. On average, pwMS reported moderate levels of self-

stigma, clinically significant levels of emotional distress and relatively poor QoL. There was a wide range of psychological flexibility scores with pwMS displaying an average level of PF of 76.25 in this sample. Cut off score interpretation for the compACT amongst pwMS have been previously reported to be below average if the total score < 59 (Meek et al., 2022). The mean Total Emotional Distress score as assessed by the HADS-T was 18.30 (with cut off scores of >8 on each subscale) and the mean QoL score was 14.68.

Table 2*Descriptive statistics for comparable key data*

Measure	Mean	Standard Deviation (SD)	Range	Skewness z-score	Kurtosis z-score
ComPACT (Psychological Flexibility)	76.25	22.72	101 (23-124)	0.43	-2.27
HADS (T) (Total Emotional Distress)	18.30	8.14	35 (3-28)	1.70	-2.12
<i>HADS-D (Depression)</i>	<i>8.06</i>	<i>4.74</i>	<i>21 (0-21)</i>	<i>1.43</i>	<i>-2.13</i>
<i>HADS- A (Anxiety)</i>	<i>10.24</i>	<i>4.22</i>	<i>10 (2-20)</i>	<i>1.53</i>	<i>-2.27</i>
SSCI (Self-stigma)	5.60	2.37	8 (2-10)	1.7	-2.40
<i>SSCI (Enacted-stigma)</i>	<i>13.50</i>	<i>5.36</i>	<i>24 (6-30)</i>	<i>4.08</i>	<i>0.68</i>
LMSQoL (Quality of Life)	14.68	4.98	15 (2-24)	-1.80	-1.50

Note: higher scores on LMSQoL indicate poorer QoL. HADS-D & HADS-A scores were combined to form HADS-T (Emotional Distress). SSCI (Enacted Stigma) score was added to subsequent mediation model as covariate.

4.2 Correlation Analyses

Table 4 highlights the statistically significant associations found between key variables of interest. Emotional Distress (HADS-T) and Quality of Life (LMSQoL) were found to be highly correlated (.820). In order to address multi-collinearity, a Variance Inflation Factor test was run (see 4.3). Associations were observed between *self-stigma*, psychological flexibility and emotional distress and a strong positive correlation was highlighted between *self-stigma* and QoL. Higher *self-stigma* was associated with lower psychological flexibility, higher emotional distress and poorer QoL. Furthermore MS subtype, employment status, age, gender and time since diagnosis were significantly associated with QoL. Individuals who had a more progressive MS subtype and who were unemployed reported poorer QoL. Similarly, those who were older, male and who had been diagnosed with MS for a longer duration reported poorer QoL. Due to the potentially confounding effect of gender, age, enacted stigma, employment status and MS-subtype were entered as covariates in the subsequent serial mediation analysis.

	1.	2.	3.	4.	5.	6.	7.	8.	9.	10.	11.	12.
1. Psychological Flexibility (compACT)	-											
2. Total Emotional Distress (HADS- T)	-.789**	-										
3. Depression (HADS-D)	-.713**	.919**	-									
4. Anxiety (HADS-A)	-.722**	.897*	.651**	-								
5. Self-Stigma (SSCI-8)	-.448*	.528*	.530**	.425**	-							
6. Enacted Stigma (SSCI-8)	-.448**	.611**	.618**	.485**	.546**	-						
7. Quality of Life (LMSQoL)	-.729**	.820**	.811**	.673**	.638**	.648**	-					
8. MS Subtype	.014	.084	.171*	-.030	.131	.156*	.149*	-				
9. Employment Status	.086	-.179**	-.248**	-.067	-.146*	-.270**	-.163*	-.480**	-			
10. Age	.160*	-.028	.110	-.177*	-.051	.032	-.028	.493**	-.498**	-		
11. Gender	-.087	.078	-.004	.155*	.103	.027	-.061	-.196**	.209**	-.134	-	
12. Time Since Diagnosis (yrs)	.035	.044	.132	-.062	-.030	.059	.011	.425**	-.341**	.525**	-.277**	-

Table 4 Correlations between key study variables

Note: *correlation is significant at 0.05 (one-tailed), ** correlation is significant at 0.01 (one tailed). Based on 1000 bootstrap samples. NB: MS Status coded (1= Relapse Remitting, 2= Progressive Types, Primary Progressive, Secondary Progressive, Progressive Relapsing), Employment Status coded (0= unemployed, 1= employed), gender coded (1=male, 2= female).

4.2 Linear Regression

A linear regression analysis was conducted to investigate whether higher levels of *self-stigma* led to lower QoL amongst pwMS. The model was statistically significant [$R^2 = .407$], $F(1, 206) = 141.55$, $p < .001$. Higher levels of *self-stigma* predicted poorer QoL. Multiple Linear Regression analyses were used to determine whether *self-stigma*, psychological flexibility, and emotional distress significantly predicted QoL (highlighted in Table 5). The overall model was statistically significant [$R^2 = .745$], $F(3, 204) = 199.01$, $p < .001$. *Self-stigma*, psychological flexibility and emotional distress explained 74% of variance in QoL scores.

Table 5

Regression Analysis Summary for Self- Stigma, Psychological Flexibility and Emotional Distress predicting QoL

Variable	Beta	SE	95% CI		β	p
			LL	UL		
Self-stigma	0.58	0.09	0.41	0.75	0.28	<.001
Psychological Flexibility	-0.42	0.01	-0.10	-0.02	-0.20	<.001
Emotional Distress	0.32	0.04	0.25	0.39	0.52	<.001

Note. * $p < .001$

It was found that high levels of *self-stigma* significantly predicted poorer QoL ($\beta_1 = 0.58$, $p < .001$) and lower levels of psychological flexibility also predicted poorer QoL ($\beta_2 = -0.42$, $p < .001$). Furthermore, higher levels of emotional distress predicted poorer QoL ($\beta_3 = 0.32$, $p < .001$). The regression analysis was also used to assess inflation factors. Tests to see

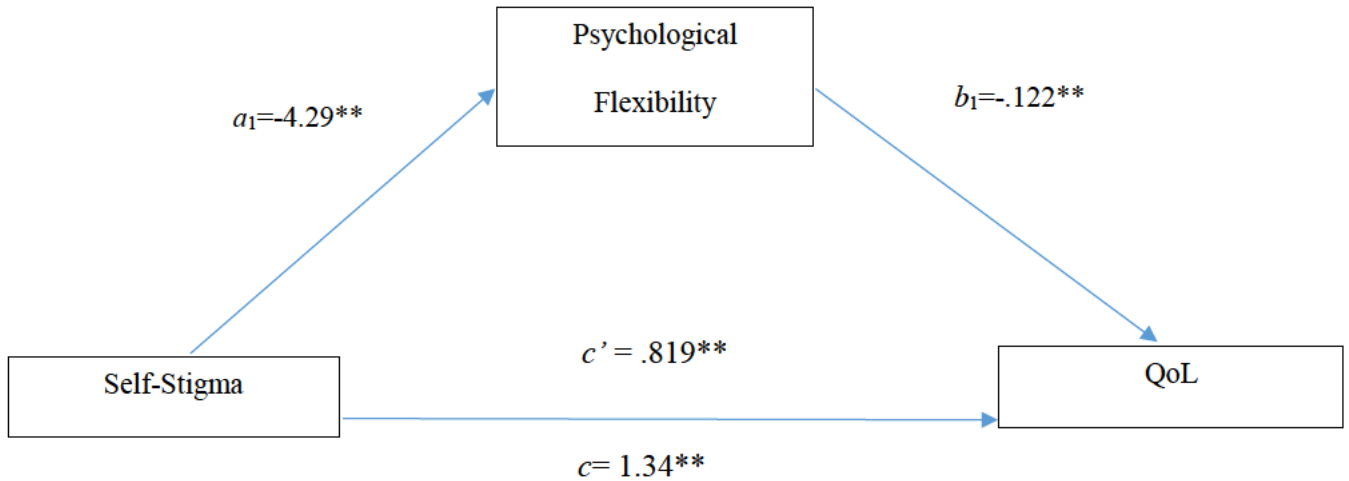
if the data met the assumption of collinearity indicated that multicollinearity was not a concern as the variance inflation factors associated with each of the predictors was < 5 .

4.3 Simple Mediation

In order to explore the study hypotheses, mediation analyses were utilised, using Hayes PROCESS (version 4.1; model 6) (Hayes, 2022) in SPSS. Bootstrapping was based on 5000 samples and significance for the direct and indirect effects was determined if the bootstrapped confidence intervals did not cross zero. Firstly, a simple mediation model was conducted and the outcome variable entered was QoL (Y), the independent variable was *self-stigma* (X), the mediator variable was psychological flexibility (M_1). In support of hypothesis 1, there was a significant direct effect of *self-stigma* on QoL (c') ($b= 1.34, [1.12; 1.56]$). Higher levels of *self-stigma* predicted poorer QoL amongst pwMS (highlighted in Figure 2). *Self-stigma* predicted 41% of the variance in QoL scores in this model.

Figure 2

Self-Stigma as a predictor of QoL, mediated by Psychological Flexibility.



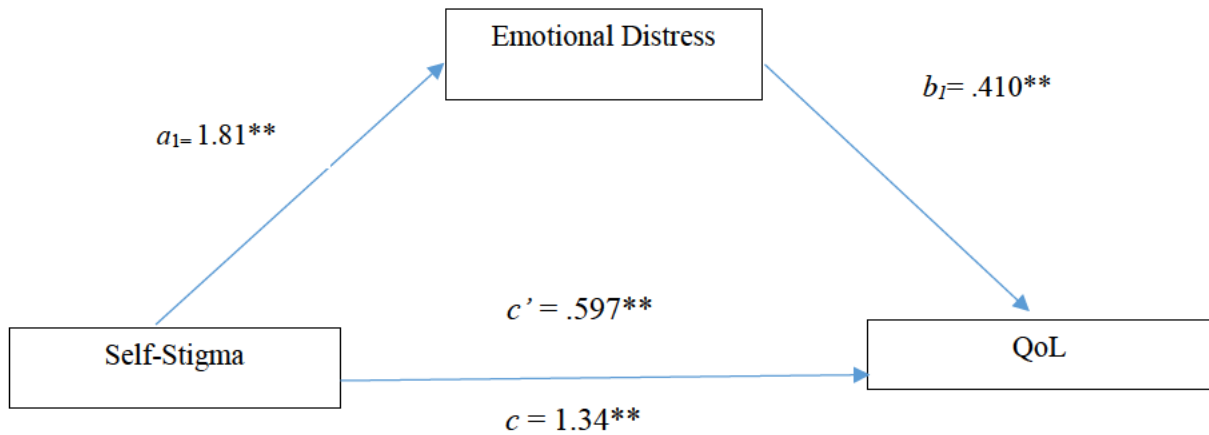
Note: $p < .001$, unstandardised indirect effects; c = the total effect of self-stigma on QoL

In relation to hypothesis 2, Psychological flexibility partially mediated the association between *self-stigma* and QoL. ($b = .521, [.376; .679]$). Higher levels of *self-stigma* predicted lower levels of psychological flexibility (a_1) ($b = -4.29, [-5.47; -3.11]$) and lower levels of psychological flexibility predicted poorer levels of QoL (b_1) ($b = -.122, [-.141; -.102]$). Psychological flexibility 65% of the variance in QoL scores in this model.

A second simple mediation model was conducted in which the mediator variable which was emotional distress (M_2) (highlighted in Figure 3).

Figure 3

Self-Stigma as a predictor of QoL, mediated by Emotional Distress.



Note: $p < .001$, unstandardised indirect effects; c = the total effect of self-stigma on QoL

In relation to hypothesis 3, emotional distress partially mediated the association between *self-stigma* and QoL ($b = .744, [.559; .640]$). Higher levels of *self-stigma* predicted higher levels of emotional distress (a_1) ($b = 1.82 [1.42; 2.22]$) and higher levels of emotional distress predicted poorer QoL (b_1) ($b = .410, [.359; .462]$). Emotional distress predicted 73% of the variance in QoL scores.

4.4 Serial Mediation

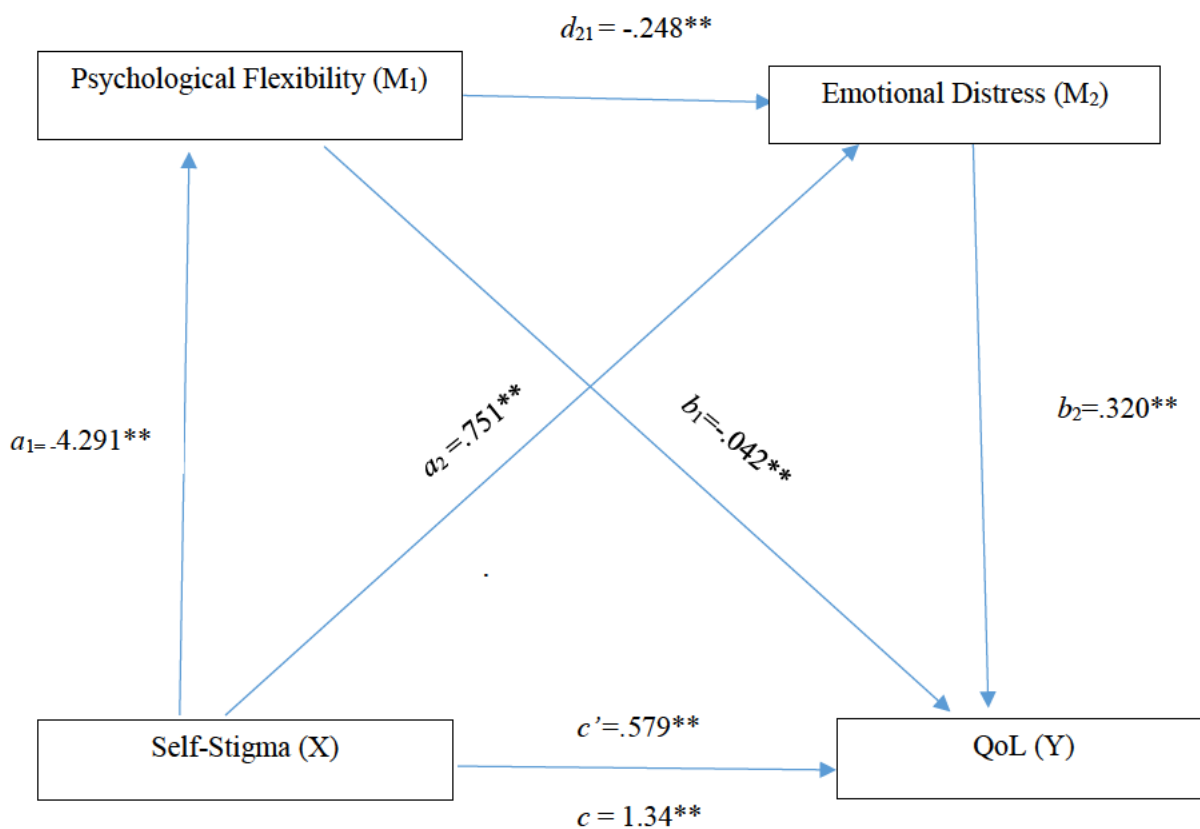
In order to test the fourth study hypothesis, a serial mediation analysis was conducted. Age, gender, enacted stigma, employment status and MS-subtype were entered as covariates due to their significant correlations between key study variables. In Table 6 the model

summary, regression co-efficients, confidence intervals and standard errors are presented.

Figure 2 highlights the serial mediation model.

Figure 2

Serial Mediation Model featuring Psychological Flexibility and Emotional Distress



*note: $^{**} = p < .001$, unstandardised indirect effects; c = the total effect of self-stigma on QoL.*

In support of hypothesis 4, there was a significant indirect effect from *self-stigma* to psychological flexibility (a_1) through emotional distress (d_{21}) on QoL (b_2) ($b = .339$, [.232; .469]). Higher levels of *self-stigma* predicted lower levels of psychological flexibility and lower levels of psychological flexibility predicted higher emotional distress which predicted

poorer QoL. The standardised indirect effect of *self-stigma* on QoL when tested through psychological flexibility and emotional distress was significant ($b = .162, [.114; .218]$). Self-stigma, psychological flexibility and emotional distress predicted 75% of the variance in QoL scores. The total effect of *self-stigma* on QoL was 1.34. Pairwise comparisons of the specific indirect effects revealed that psychological flexibility and emotional distress were not statistically different from any other specific indirect effect as the confidence intervals crossed zero.

When reverse mediation was conducted and the order of the mediator variables was changed, the path between *self-stigma* and psychological flexibility was not statistically significant, as the confidence interval crossed zero, therefore supporting the order of the serial mediation chain. In addition, when *enacted stigma*, employment status, MS Status, gender and age were added to the model as covariates, the association between *self-stigma* and emotional distress remained significant with a point estimate of .332, however this strength of the association was reduced. All other associations remained significant.

Table 6 Regression Coefficients, Standard Errors and Model Summary Information.

		Consequent										
		M ₁ (Psychological Flexibility)			M ₂ (Emotional Distress)			Y (QoL)				
Antecedent		Coeff.	SE	p		Coeff.	SE	p		Coeff	SE	p
X (Self-stigma)	<i>a</i> ₁	-4.291	0.597	<.001	<i>a</i> ₂	0.751	0.157	<.001	<i>c</i> '	0.579	0.087	<.001
M ₁ (Psychological Flexibility)		—	—	—	<i>d</i> ₂₁	-0.248	0.016	<.001	<i>b</i> ₁	-0.042	0.013	<.001
M ₂ (Emotional Distress)		—	—	—		—	—	—	<i>b</i> ₂	0.320	0.037	<.001
Constant	<i>i</i> _{M1}	100.181	3.615	<.001	<i>i</i> _{M2}	32.994	1.84	<.001	<i>i</i> _y	8.837	1.567	<.001
			R ² = 0.201			R ² = 0.661			R ² = 0.745			
			F (1, 206) = 51.7, p <.001			F (2, 205) = 199.57, p <.001			F (3, 204) = 199.01, p <.001			

5.0 Discussion

This study aimed to determine whether higher levels of *self-stigma* led to poorer QoL in pwMS and tested the prediction that psychological flexibility and emotional distress would both independently and serially mediate the link between *self-stigma* and QoL. Consistent with our hypothesis, higher levels of *self-stigma* were associated with lower QoL in pwMS, with higher *self-stigma* associated with lower psychological flexibility, higher emotional distress levels and poorer QoL. This current sample experienced marginally higher levels of emotional distress (depressive and anxiety symptoms) than in previous research amongst people with secondary progressive MS (Meek et al., 2022). Psychological flexibility accounted for some of the variance in the association between self-stigma and QoL, which follows similar findings through research of single facets of psychological flexibility (e.g. cognitive fusion) and stigma in pwMS (Valvano et al., 2016).

The association between *self-stigma* and QoL was also found to operate indirectly through emotional distress. This finding is in line with cross-sectional research involving heroin users (Cheng et al., 2019) and individuals with schizophrenia (Lin et al., 2016). Both psychological flexibility and emotional distress represent mechanisms which indirectly link *self-stigma* and QoL. Specifically, if individuals self-stigmatise they may possess less psychological flexibility, which in turn may lead to higher levels of emotional distress and poorer QoL. These findings highlight the importance of attending to psychological flexibility, and symptoms of emotional distress when addressing self-stigma experiences amongst pwMS.

Whilst this is the first known study to test a serial mediation model relating to self-stigma in a cross-sectional sample of pwMS, the findings are consistent with previous research indicating that pwMS who reported higher levels of *self-stigma* experienced poorer QoL (Broersma et al., 2018). Furthermore, a significant association was observed between psychological flexibility and QoL in this current study, in line with previous research exploring psychological flexibility and QoL in secondary progressive MS (Meek et al., 2022). Whilst Meek et al. (2022) reported greater psychological flexibility to be associated with improved QoL amongst people with secondary progressive MS (Meek et al., 2022), this study found no evidence linking MS-subtype and psychological flexibility. Whilst previous research has also reported a positive association between *self-stigma* and age (Anagnostouli et al., 2016) amongst pwMS, this study did not find evidence in support of their findings.

5.1 Strengths & Limitations

This study contains several limitations that warrant consideration. Firstly, the cross-sectional design prevents conclusions being drawn about the nature of casual or temporal links between variables. Some researchers argue that mediation analysis should not be conducted on cross-sectional data, as a substantial indirect effect can be implied which may otherwise be zero in a longitudinal design (Maxwell & Cole 2007). Future research ought to be conducted on a longitudinal basis, exploring *self-stigma* at two time points amongst pwMS; for example at the initial diagnosis and then two years following, at a time when individuals may be more accepting of their condition. This approach would not only address Maxwell & Cole's concerns but would also allow for further quantification of *self-stigma*,

psychological flexibility, emotional distress and QoL which may reveal insights into predictors of adjustment.

In addition, this study did not ascertain the social support levels or the educational attainment of pwMS. Previous research has shown higher social support to be positively associated with QoL amongst pwMS (Ratajska et al., 2020) and relationship status and educational level have been identified as better predictors of QoL amongst older pwMS (Buhse et al., 2014). Other research has highlighted that individuals who were in a relationship were less likely to report *self-stigma* (Eldridge-Smith et al., 2021; Silverman et al., 2017), highlighting that social support may serve as a protective buffer against self-stigmatisation. Future research may benefit from addressing the role of social support and educational attainment in *self-stigma* experiences.

Due to Covid-19 restrictions at the time of data collection, it was not possible to collect data from a clinical sample of pwMS. Since more people with relapse remitting MS completed this survey, progressive forms of MS subtypes were combined and grouped against relapse remitting MS, allowing MS subtype to be utilised as a control variable in the study analysis. However, this may have been a simplistic approach given the nuances between the different progressive subtypes of MS. The combining of anxiety and depression scores could also be considered to be a limitation, given the distinct differences between these conditions. Future research should attempt to address these issues in a clinical sample of pwMS in order to elucidate how self-stigma impacts on QoL in pwMS with potentially more progressive symptoms.

Despite the aforementioned limitations, this study is benefited by a number of strengths. It is the first study to test a hypothesised link between *self-stigma*, psychological flexibility and emotional distress on QoL in a cross-sectional sample of pwMS. This study recruited a good sample size and it was the first to predominantly focus on *self-stigma*, a factor more intrinsically linked to internal mechanisms such as psychological flexibility and emotional distress. This research offers a novel theoretical model to account for how *self-stigma* relates to QoL amongst pwMS, which may be useful to inform and underpin future research and service provisions.

5.2 Clinical Implications

The current study highlights that pwMS self-stigmatise and that this experience negatively impacts on their QoL. This research suggests the importance of psychological flexibility and emotional distress (depression, anxiety) as mechanisms to consider when addressing the effects of *self-stigma* on QoL in pwMS. Interventions such as ACT, which aim to foster skills to improve psychological flexibility and tolerate difficult emotions may buffer against the effects of *self-stigma*, in line with previous research promoting the use of ACT clinically (Bai et al., 2020; Gloster et al., 2020). Empowering pwMS may be an effective way of reducing self-stigmatisation through encouraging clients to believe that their goals can be achieved (Corrigan & Rao., 2012); if pwMS feel up-skilled and validated, they may be less likely to agree with negative public stereotypes and apply them to themselves, leading to lower levels of emotional distress and improved QoL. However, a recent meta-analysis assessing the effectiveness of ACT for improving QoL in pwMS across six studies reported that there was no significant effect of ACT on reducing anxiety or depression or improving QoL (Thompson et al., 2022). Whilst it is noteworthy that none of the included studies

addressed stigma experiences of pwMS, other interventions may be useful to promote QoL and reduce *self-stigma* amongst pwMS.

Cognitive Behavioural Therapy (CBT) has demonstrated efficacy in helping to reduce symptoms of emotional distress. Two meta-analyses involving a total of 17 studies of pwMS found CBT and mindfulness based therapies to have a small to moderate effect of reducing psychological distress (Ires et al., 2019). Although the study quality was low and the studies did not address stigma, CBT has been reported to help individuals to buffer against the detrimental effects of *self-stigma* through cognitive restructuring to challenge irrational beliefs (Morrison et al., 2013). Furthermore, anxiety and depression symptoms are targeted by developing an individual's coping skills and implementing relaxation techniques (Jones et al., 2013). Group based CBT interventions have highlighted positive effects in the treatment of *self-stigma* in schizophrenia, by promoting readiness for change in relation to problematic behaviours and enhancing self-esteem (Fung et al., 2010) and reducing *perceived stigma* and improving treatment compliance in major depressive disorder (Tong et al., 2020). Group based interventions may also offer a cost effective alternative to individual approaches (Østergaard et al., 2020). Early interventions could be targeted at younger adults, given the positive association between age and psychological flexibility reported here and elsewhere (Meek et al., 2022).

However, it could be argued that as human beings we all have the propensity to stigmatise and be stigmatisers, therefore structural stigma should inform interventions in order to tackle stigma at a wider level (Stangl et al., 2019). Whilst this current study has focused on *self-stigma* and the mechanisms which are associated with QoL, *enacted stigma*

has also been highlighted to impact on link between *self-stigma* and emotional distress herein. Therefore, two-tailed interventions may be useful at both an individual and wider societal level to target the effects of stigmatising behaviour and teach about the influences of stigma experiences on health outcomes amongst marginalised groups. It is noteworthy that ill-informed interventions which address stigma experiences can lead to unintended consequences, which can further fuel enacted stigma (Corrigan., 2016). Therefore, it is important that future interventions are focused on relevant mechanisms such as psychological flexibility and emotional distress in order to educate the wider audience towards the detrimental effects of self-stigmatisation on QoL.

5.3 Conclusion

This present study was the first to examine psychological flexibility and emotional distress, as serial mediators of the association between *self-stigma* and QoL amongst pwMS. The results suggest that psychological flexibility and emotional distress are important mechanisms that contribute towards the positive association between *self-stigma* and QoL. Further research is needed to determine whether these findings are replicated in longitudinal samples with a more heterogeneous profile. Psychological flexibility may be a useful primary mechanism to address through treatment provisions, in order to buffer against the effects of *self-stigma* and emotional distress on QoL amongst pwMS.

6.0 References

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8.0 Appendices

Appendix A: The British Journal of Psychology Author Submission Guidelines

BJP AUTHOR GUIDELINES

Sections

Submission
Aims and Scope
Manuscript Categories and Requirements
Preparing the Submission
Editorial Policies and Ethical Considerations
Author Licensing
Publication Process After Acceptance
Post Publication

1. SUBMISSION

Authors should kindly note that submission implies that the content has not been published or submitted for publication elsewhere except as a brief abstract in the proceedings of a scientific meeting or symposium.

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Keywords;

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Acknowledgments.

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Please provide an abstract of between 100 and 200 words, giving a concise statement of the intention, results or conclusions of the article. The abstract should not include any sub-headings.

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Please provide appropriate keywords.

Acknowledgments

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Stigma and quality of life in people living with neurodegenerative conditions: a systematic review

Craig Mackay, Monja Knoll

Review methods were amended after registration. Please see the revision notes and previous versions for detail.

Citation

Craig Mackay, Monja Knoll. Stigma and quality of life in people living with neurodegenerative conditions: a systematic review. PROSPERO 2022 CRD42022375129 Available from: https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42022375129

Review question [1 change]

To investigate the impact of stigmatic experiences on quality of life amongst people living with neurodegenerative conditions.

- 1) How does stigma impact on wellbeing and quality of life across neurodegenerative conditions?
- 2) Is stigma correlated with other factors such as condition specificity, age, educational level or disease progression amongst people living with neurodegenerative conditions?

Searches [1 change]

Preliminary scoping exercises will be conducted using the Database of Abstracts for Reviews and Effects

(DARE), the Cochrane Database of Systematic Reviews (CDSR), Web of Science, PROSPERO and Google Scholar. ProQuest Dissertations and Theses Global and the Electronic Theses Online Service (ETHOS) will be accessed via the University of Edinburgh website using the keywords 'Stigma, Multiple

Sclerosis and Neurological conditions'. The PsycINFO, EMBASE, MEDLINE (R), the Cumulative Index to Nursing and Allied Health Literature (CINAHL) online databases will be searched. These databases will be selected due to their clinical, psychological and health focus. The searches will be conducted between the start of the database inception until December 2022.

A key word search involving the following terms will be utilised: Stigma*, Multiple Sclerosis", "Parkinsons Disease', 'Huntingtons Disease', Motor Neurons Disease/ Amyotrophic lateral sclerosis, 'anxi* 'depress*' and 'quality of life'.

Additional search strategy information can be found in the attached PDF document (link provided below).

Types of study to be included

Cross-sectional design studies.

Longitudinal studies.

Condition or domain being studied

Stigma experiences and health-related quality of life in people living with neurodegenerative conditions including Multiple Sclerosis, Parkinsons, Amyotrophic Lateral Sclerosis, and Motor Neurones Disease.

Participants/population

Inclusion criteria:

- Quantitative methodology
- Participants with a confirmed diagnosis of Parkinson's Disease (PD), Motor Neurone's Disease (MND)/Amyotrophic Lateral Sclerosis (ALS), Multiple Sclerosis (MS) or Huntington's (HD).
- Participants aged 18+
- Studies that measured stigma and correlated this with demographic variables, e.g. social or clinical factors.

Exclusion criteria:

- Qualitative studies
- Intervention studies
- Studies where stigma was measured but not correlated with demographic, social or clinical factors.
- Studies that focused on individuals with the HD gene but at the pre-symptomatic phase as at this point there would be no easily discernible difference.
- Studies that included co-morbid neurological conditions.
- Studies that lack a confirmed diagnosis e.g. clinically isolated syndrome.
- Papers that are not written in English.

Intervention(s), exposure(s) [1 change]

The primary exposure factor in this review is stigma amongst people living with neurodegenerative conditions including multiple sclerosis, parkinsons, huntington's and motor neurones disease. The outcome variable is the effect of stigma experiences on well-being (anxiety/depression rates) and health-related quality of life.

Comparator(s)

/control Not applicable.

Context

Studies which measure stigma experiences and quality of life in people living with neurodegenerative conditions including Multiple Sclerosis, OR Parkinsons, OR Amyotrophic Lateral Sclerosis OR Motor Neurones Disease.

Main outcome(s) [1 change]

The main outcome will be the effect of stigma on wellbeing and quality of life in people living with neurodegenerative conditions. Studies may demonstrate links between stigma and condition specificity, age, educational level or disease progression. Only validated self-report measures of stigma, anxiety, depression and quality of life will be used.

Measures of effect r values, beta coefficients, statistical significance, confidence intervals, effect sizes.

Additional outcome(s)

[1 change] None.

Measures of effect

Not applicable

Data extraction (selection and coding) [1 change]

The initial search will be conducted via the OVID interface (with duplicates removed), Thesis ProQuest Global, Web of Science, Google Scholar and hand searches. Covidence will assist with the screening and identification of relevant articles for inclusion. Full texts will be identified, with main authors emailed if full texts are not available online. Data from studies which meet the relevant inclusion criteria will be added to an Excel spreadsheet with data relating to:

- Study authors
- Country
- Study type
- Sample and Setting
- Number of respondents/ gender
- Age (range)
- Diagnosis confirmation
- Stigma measure
- Results summary
- Commentary

A sample of 10% of studies extracted will be rated by a second reviewer to allow a consensus to be achieved.

Risk of bias (quality) assessment

Included studies will be examined using the Newcastle-Ottawa Quality Assessment Checklist.

Predominantly the cross-sectional study variation will be used. However, when other studies are identified, e.g. longitudinal studies, then a variation of this checklist will be utilised. An independent reviewer will also rate each study against this checklist.

Strategy for data synthesis [1 change]

In light of potential study design variability, differing outcome measures and statistical approaches a metaanalysis will not be used to synthesise the results. Instead, a formal narrative synthesis will be employed using the guidelines of Popay and colleagues (Popay et al., 2006). The narrative synthesis will be conducted on a minimum of 5 studies involving samples with multiple sclerosis, parkinsons, amyotrophic lateral sclerosis or motor neurone disease. Quantitative results will be extracted from studies which are relevant to each aim of the review. A textual description will be provided of each study summary and relevant data will be grouped in relation to the type of self-report measure, e.g. stigma, well-being (anxiety/depression) or quality of life measures. Significant findings from studies will be reported according to study authors and tabulated. In terms of study quality, the review will

outline methodological limitations and provide recommendations for future research. PRISMA guidelines will be used for writing up and reporting items for the systematic review (Moher et al., 2009).

Analysis of subgroups or subsets None planned.

Contact details for further information

Craig Mackay

Organisational affiliation of the review [1 change]

University of Edinburgh, NHS Lanarkshire

Review team members and their organisational affiliations [1 change]

Mr Craig Mackay. University of Edinburgh, NHS Lanarkshire

Dr Monja Knoll. University of Edinburgh

Type and method of review [1

change] Narrative synthesis,
Systematic review

Anticipated or actual start date [2 changes]

06 December 2022

Anticipated completion date [1 change]

01 May 2023

Funding sources/sponsors

None

Conflicts of interest

None known

Language

English

Country

Scotland

Stage of review

Review
Ongoing

Subject index terms status

Subject indexing assigned by CRD

Subject index terms

Disease Progression; Humans; Neurodegenerative Diseases; Quality of Life; Social Stigma; Stereotyping

Date of registration in PROSPERO

05 December 2022

Date of first submission

21 November 2022

Stage of review at time of this submission [1 change]

Stage completed	Started Com	
Preliminary searches	Yes	Yes
Piloting of the study selection process	Yes	Yes
Formal screening of search results against eligibility criteria	No	No
Data extraction	No	No
Risk of bias (quality) assessment	No	No
Data analysis	No	No

Revision note

period has been updated to include up to December 2022.

The record owner confirms that the information they have supplied for this submission is accurate and complete and they understand that deliberate provision of inaccurate information or omission of data may be construed as scientific misconduct.

The record owner confirms that they will update the status of the review when it is completed and will add publication details in due course.

Appendix C: IRAS Ethics Committee Study Approval

Letter

London - Westminster Research Ethics Committee

Equinox House
City Link
Nottingham
NG2 4LA

29 November 2021

Mr Craig Mackay
0/2, 31 Cartside Street
Glasgow
G42 4TN

Dear Mr Craig Mackay

Study title: Exploring the relationship between stigma, psychological flexibility and health related quality of life in individuals with Multiple Sclerosis.

REC reference: 21/PR/1457

Protocol number: CAHSS2108/09

IRAS project ID: 302953

Thank you for your letter of 15th November 2021, responding to the Proportionate Review Sub-Committee's request for changes to the documentation for the above study.

The revised documentation has been reviewed and approved on behalf of the PR sub-committee.

Confirmation of ethical opinion

On behalf of the Research Ethics Committee (REC), I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised.

Good practice principles and responsibilities

The [UK Policy Framework for Health and Social Care Research](#) sets out principles of good practice in the management and conduct of health and social care research. It also outlines the responsibilities of individuals and organisations, including those related to the four elements of [research transparency](#):

1. [registering research studies](#)

2. [reporting results](#)
3. [informing participants](#)
4. [sharing study data and tissue](#)

Conditions of the favourable opinion

The REC favourable opinion is subject to the following conditions being met prior to the start of

the study.

Confirmation of Capacity and Capability (in England, Northern Ireland and Wales) or NHS management permission (in Scotland) should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise).

Guidance on applying for HRA and HCRW Approval (England and Wales)/ NHS permission for research is available in the Integrated Research Application System.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of management permissions from host organisations.

Registration of Clinical Trials

All research should be registered in a publicly accessible database and we expect all researchers, research sponsors and others to meet this fundamental best practice standard.

It is a condition of the REC favourable opinion that **all clinical trials are registered** on a publicly accessible database within six weeks of recruiting the first research participant. For this purpose, 'clinical trials' are defined as the first four project categories in IRAS project filter question 2. Failure to register is a breach of these approval conditions, unless a deferral has been agreed by or on behalf of the Research Ethics Committee (see here for more information on requesting a deferral: <https://www.hra.nhs.uk/planning-and-improving-research/research-planning/research-registration-research-project-identifiers/>)

If you have not already included registration details in your IRAS application form, you should notify the REC of the registration details as soon as possible.

Publication of Your Research Summary

We will publish your research summary for the above study on the research summaries section of our website, together with your contact details, no earlier than three months from the date of this favourable opinion letter.

Should you wish to provide a substitute contact point, make a request to defer, or require further information, please visit:

<https://www.hra.nhs.uk/planning-and-improving-research/application-summaries/research-summaries/>

N.B. If your study is related to COVID-19 we will aim to publish your research summary within 3 days rather than three months.

During this public health emergency, it is vital that everyone can promptly identify all relevant research related to COVID-19 that is taking place globally. If you haven't already done so, please register your study on a public registry as soon as possible and provide the REC with the registration detail, which will be posted alongside other information relating to your project. We are also asking sponsors not to request deferral of publication of research summary for any projects relating to COVID-19. In addition, to facilitate finding and extracting studies related to COVID-19 from public databases, please enter the WHO official acronym for the coronavirus disease (COVID-19) in the full title of your study. Approved COVID-19 studies can be found at:

<https://www.hra.nhs.uk/covid-19-research/approved-covid-19-research/>

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

After ethical review: Reporting requirements

The attached document "After ethical review – guidance for researchers" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study, including early termination of the study
- Final report
- Reporting results

The latest guidance on these topics can be found at <https://www.hra.nhs.uk/approvals-amendments/managing-your-approval/>.

Ethical review of research sites

The favourable opinion applies to all NHS/HSC sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see “Conditions of the favourable opinion” above).

Approved documents

The documents reviewed and approved by the Committee are:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Copies of materials calling attention of potential participants to the research [Poster, Stigma, PF & MS]	1	05 October 2021
Covering letter on headed paper [Covering Letter]		
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [PI Confirmation]		05 October 2021
IRAS Application Form [IRAS_Form_12102021]		12 October 2021
IRAS Application Form XML file [IRAS_Form_12102021]		12 October 2021
Other [EL Certificate]	1	05 October 2021
Other [response letter v1]	1	12 November 2021
Participant consent form [Consent Form, Stigma, PF & MS]	1	05 October 2021
Participant information sheet (PIS) [PIS, Stigma, PF & MS]	2	12 November 2021
Research protocol or project proposal [Stigma, PF & MS]	1	05 October 2021
Schedule of Events or SoECAT [IRAS Schedule of Events, Stigma, PF & MS]	1	05 October 2021
Summary CV for Chief Investigator (CI) [CV CM]		05 October 2021
Summary CV for student [CV CM]		05 October 2021
Summary CV for supervisor (student research) [CV MK]		05 October 2021
Summary of any applicable exclusions to sponsor insurance (non-NHS sponsors only) [TWIMC]		05 October 2021
Summary, synopsis or diagram (flowchart) of protocol in non technical language [Recruitment Flow Chart, Stigma, PF & MS]	1	05 October 2021
Validated questionnaire [Stigma, PF & MS]		04 October 2021

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received

and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website:

<http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/>

HRA Learning

We are pleased to welcome researchers and research staff to our HRA Learning Events and online learning opportunities– see details at:

<https://www.hra.nhs.uk/planning-and-improving-research/learning/>

IRAS project ID: 302953 correspondence	Please quote this number on all
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With the Committee's best wishes for the success of this project.

Yours sincerely

pp Mr Robert Goldstein Chair

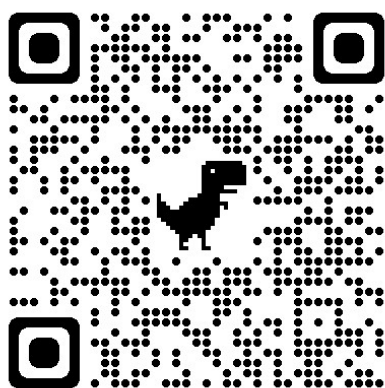


THE UNIVERSITY of EDINBURGH



RECRUITMENT POSTER MULTIPLE SCLEROSIS STUDY

- We are looking for people living with multiple sclerosis (MS) to complete an online questionnaire.
- We want to learn more about stigma, thinking styles and well-being in people living with MS.
- The questionnaire will take no more than 15 minutes to complete
- If you are interested, please scan the QR code, or contact the researcher via email below.



***Craig Mackay (Trainee Clinical Psychologist/ NHS Lanarkshire/
University of Edinburgh) E:***

Appendix E: Participant Information Sheet



THE UNIVERSITY of EDINBURGH



PARTICIPANT INFORMATION SHEET

Exploring the relationship between stigma, psychological flexibility and health related quality of life in individuals with Multiple Sclerosis.

You are being invited to take part in research on Multiple Sclerosis. The primary researcher is Craig Mackay (Trainee Clinical Psychologist) at the University of Edinburgh is leading this research. Before you decide whether to take part it is important you understand why the research is being conducted and what it will involve. Please take time to read the following information carefully.

WHAT IS THE PURPOSE OF THE STUDY?

The purpose of the study is to try to further understand the relationship between stigma, psychological flexibility and quality of life in individuals with MS. The term psychological flexibility relates to the holding of individual thoughts and feelings more lightly and acting on longer term goals and values, as opposed to shorter ones. A questionnaire will be presented which will aim to assess individual's psychological flexibility, their stigmatic experiences, quality of life and mood states. It is well known that stigma can negatively affect people's quality of life. For example, stigma can cause difficulties within social relationships. This research will look deeper into the association between stigma and quality of life in people living with Multiple Sclerosis. It may be that individual levels of psychological flexibility may influence how stigmatic experiences are perceived and how quality of life is rated by people living with MS. This study hopes to inform future service provisions by understanding more about 'Psychological Flexibility' and how it relates to people with MS.

WHY HAVE I BEEN INVITED TO TAKE PART?

You are invited to participate in this study because you are someone who is currently living with Multiple Sclerosis, aged 16 or over and are fluent in English. In some cases, you may have previously expressed an interest in being contacted with regards to MS related research or through the third sector charity of which you are affiliated.

DO I HAVE TO TAKE PART?

No – it is entirely up to you. If you do decide to take part, you are still free to withdraw at any time and without giving a reason. Deciding not to take part or withdrawing from the study will not affect your current or future healthcare in any manner.

Please note that your data will be anonymous therefore it will not be possible to withdraw your data once you submit your responses.

WHAT WILL HAPPEN IF I DECIDE TO TAKE PART?

If you do decide to take part, you will be asked to complete an online Informed Consent Form to show that you understand your rights in relation to the research, and that you are happy to participate.

You will then be asked to complete an online questionnaire where you will be asked a number of questions regarding your demographic information, stigmatic experiences, thinking styles and health related quality of life. The questionnaire will be hosted online through a secure platform called Qualtrics. No personal information will be requested. The questionnaire should take around 15 minutes to complete.

If you wish to complete a paper copy of the questionnaire as opposed to the online version, please email the primary researcher and highlight your preferred method of engagement.

WHAT ARE THE POSSIBLE BENEFITS OF TAKING PART?

There are no direct benefits, but by sharing your experiences with us, you will be helping Craig Mackay and the University to better understand psychological flexibility and stigmatic experiences and how they relate to people living with MS.

ARE THERE ANY RISKS OR DISADVANTAGES ASSOCIATED WITH TAKING PART?

There are no significant risks associated with participation. However, there is a slight chance that considering your own well-being may bring up some of your own negative emotions. If you need to discuss your mental well-being further we would advise that you contact your GP in the first instance. It may also be helpful to contact third sector MS charities such as the MS Society for emotional support:

<https://www.mssociety.org.uk/>

For more information relating to MS, please visit the NHS Inform website:

<https://www.nhsinform.scot/illnesses-and-conditions/brain-nerves-and-spinalcord/multiple-sclerosis-ms>

WILL MY TAKING PART BE KEPT CONFIDENTIAL?

All the information we collect during the course of the research will be kept confidential and there are strict laws which safeguard your privacy at every stage.

HOW WILL WE USE INFORMATION ABOUT YOU?

No personally identifiable information will be stored during the duration of this study. Once we have finished the study, we will keep some of the data so we can check the results. We will write our reports in a way that no-one can work out that you took part in the study.

What are your choices about how your information is used?

- You can stop being part of the study at any time, without giving a reason, but we will keep information about you that we already have.
- We need to manage your records in specific ways for the research to be reliable. This means that we won't be able to let you see or change the data we hold about you.

Where can you find out more about how your information is used?

You can find out more about how we use your information at

<https://www.ed.ac.uk/records-management/privacy-notice-research>

- our leaflet available from our website
- by asking one of the research team
- by sending an email to

The University of Edinburgh is the sponsor for this study based in the United Kingdom/ Scotland. We will be using information from you in order to undertake this

study and will act as the data controller for this study. This means that we are responsible for looking after your information and using it properly. The University of Edinburgh will keep anonymised data for a maximum of 10 years. The anonymised data may be used in future ethically approved research.

WHAT WILL HAPPEN WITH THE RESULTS OF THIS STUDY?

The results of this study may be summarised in published articles, reports and presentations. You will not be identifiable from any published results. Quotes or key findings will always be made anonymous in any formal outputs unless we have your prior and explicit written permission to attribute them to you by name. A summary of the findings from the study will be made available to participants who indicate they would like to receive this.

WHO IS ORGANISING THE RESEARCH?

This study has been organised by Craig Mackay-Trainee Clinical Psychologist and sponsored by the University of Edinburgh. This research is contributing towards the researchers' PHD.

WHO HAS REVIEWED THE STUDY?

All research in the NHS is looked at by an independent group of people called a Research Ethics Committee. A favourable ethical opinion has been obtained from the London Westminster Research Ethics Committee. NHS Management Approval has also been given.

WHO CAN I CONTACT?

If you have any further questions about the study, please contact the lead researcher, Craig Mackay- Trainee Clinical Psychologist:

If you would like to discuss this study with someone independent of the study please contact:

Dr Ewan Culling (Clinical Psychologist):

If you wish to make a complaint about the study, please contact:

Dr Angus MacBeth (Senior Lecturer in Clinical Psychology/ Research Lead
University of Edinburgh Doctorate Programme)

Appendix F: Participant Consent Form



THE UNIVERSITY of EDINBURGH



PARTICIPANT CONSENT FORM

Study Title: Exploring the relationship between stigma, psychological flexibility and health related quality of life in individuals with Multiple Sclerosis.

Researcher's name and contact details:
Craig Mackay, Trainee Clinical Psychologist: email:

Please tick box

1. I confirm that I have read and understood the Participant Information Sheet (Version 1 dated (05 09 2021) for the above study.
2. I have been given the opportunity to consider the information provided, ask questions and have had these questions answered to my satisfaction.
3. I understand that my participation is voluntary and that I can withdraw from the study at any time without giving a reason and without my medical care or legal rights being affected.
4. I understand that my anonymised data will be stored for a maximum of 10 years and may be used in future ethically approved research.
5. By ticking this box I agree to take part in the above study.

Appendix G: Online Study Questionnaire

Tools ▾ Saved at 9:05 PM Draft



Preview

Publish

Stigma, Psychological Flexibility & Well-being in individuals living with

iQ Score: Fair

▼ Participant Information Sheet

PIS

iQ ...

PARTICIPANT INFORMATION SHEET

This project involves completing a series of questionnaires, hosted via the online platform Qualtrics. The questionnaires relate to information regarding your general well-being, experiences of living with Multiple Sclerosis and potential previous experiences of stigma. You will also be asked some demographic information relating to your age/years since MS diagnosis for example. The questions should take you around 25 minutes to complete.

Stigma has been implicated in the relationship between well-being and other chronic health conditions such as HIV & epilepsy. However, few studies have explored the impact of stigma in relation to the well-being of people living with MS. Recent studies have suggested that the way that we think can affect the way that we engage with the positive and negative experiences that have happened to us in life, which in turn can influence our well-being.

'Psychological flexibility' is a term which relates to taking action to manage the interference of previous experiences on our wellbeing and to take action in the pursuit of personal goals. In other words, higher levels of psychological flexibility may lead to increased well-being and a higher quality of life. This research aims to see whether this is true when we consider the potential role of stigma in people living with MS.

The results of this study will help to inform educational/treatment packages for staff and people living with MS. Psychological flexibility is the core component of a third wave therapy called 'Acceptance and Commitment Therapy' which is a variant of 'Cognitive Behavioural Therapy', used in the UK to treat people with psychological issues. This study will utilise a newly created questionnaire called 'ComPACT' which aims to determine an individual's level of psychological flexibility more generally than measures used in previous studies.

Your participation in this project is entirely voluntary and should you wish to withdraw your responses at any time, you are free to do so by contacting the primary researcher.

No personal identifiable information will be obtained. All data will be held securely & in line with NHS Lanarkshire/University of Edinburgh information governance policies. Thank you in advance for your time and interest in this project.

Page Break

Consent Form



Consent Form

* I confirm that I have read and understood the Participant Information Sheet for the above study.

* I understand that my participation in this study is entirely voluntary & I can withdraw my responses at any time.

* I understand that my anonymised data will be held securely for a maximum period of 10 years.

* I understand that the anonymised data collected may be used to inform future research studies relating to MS but I will not be identifiable from the data.

* I agree to take part in this study.

* Please click/tick 'I agree' to confirm you have read the information and that you consent to take part in this study.

I agree

I do not consent

-----Page-Break-----

DA



How old are you?

-----Page-Break-----

DG



Which of the following best describes your gender?

Male

Female

Non-binary / third gender

Prefer not to say

-----Page-Break-----

DO



What is your current occupation?. If you do not work, please write 'n/a'

-----Page-Break-----

DMSS



Which of the following best describes your MS?

- Relapsing- remitting
- Primary progressive
- Secondary progressive
- Progressive relapsing

----- Page Break -----

DE



Which of the following best describes your ethnicity?

- White
- Black or African American
- American Indian or Alaska Native
- Asian
- Native Hawaiian or Pacific Islander
- Other

----- Page Break -----

DYSD



How many years & months has it been since you were diagnosed with MS?



Import from library

Add new question

Add Block

▼ ACT

compACT 1

I can identify the things that really matter to me in life and pursue them

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree

compACT 2

One of my big goals is to be free from painful emotions

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree

-----Page-Break-----

compACT 3

I rush through meaningful activities without being really attentive to them

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree

-----Page-Break-----

compACT 4

I try to keep busy to keep thoughts or feelings from coming

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree

-----Page Break-----

compACT 5

I act in ways which are consistent with how I wish to live my life

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree

-----Page Break-----

compACT 6

I get so caught up in my thoughts that I am unable to do the things that I most want to do

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree

compACT 7

I make choices based on what is important to me, even if it is stressful

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree

-----Page Break-----

compACT 8

I tell myself that I shouldn't have certain thoughts

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree

-----Page Break-----

compACT 9

I find it difficult to stay focused on what's happening in the present

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree

compACT 10

I behave in line with my personal values

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree

compACT 11

I go out of my way to avoid situations that might bring difficult thoughts, feelings, or sensations

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree

compACT 12

Even when doing the things that matter to me, I find myself doing them without paying attention

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree

compACT 13

I am willing to fully experience whatever thoughts, feelings and sensations come up for me, without trying to change or defend against them

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree

-----Page Break-----

compACT 14

I undertake things that are meaningful to me, even when I find it hard to do so

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree

-----Page Break-----

compACT 15

I work hard to keep out upsetting feelings

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree

-----Page Break-----

compACT 16

I do jobs or tasks automatically, without being aware of what I'm doing

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree

-----Page Break-----

compACT 17

I am able to follow my long-term plans including times when progress is slow

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree

-----Page Break-----

compACT 18

Even when something is important to me, I'll rarely do it if there is a chance it will upset me

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree

-----Page Break-----

compACT 19

It seems I am "running on automatic" without much awareness of what I'm doing

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree

-----Page Break-----

compACT 20

Thoughts are just thoughts – they don't control what I do

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree

-----Page Break-----

compACT 21

My values are really reflected in my behaviour

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree

-----Page Break-----

compACT 22

I can take thoughts and feelings as they come, without attempting to control or avoid them

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree

-----Page Break-----

compACT 23

I can keep going with something when it's important to me

- Strongly disagree
- Moderately disagree
- Slightly disagree
- Neither agree nor disagree
- Slightly agree
- Moderately agree
- Strongly agree



Import from library

Add new question

Add Block

▼ HADS

HADS 1 (A)

I feel tense or 'wound up'

- Most of the time
- A lot of the time
- From time to time, occasionally
- Not at all

HADS 1 (D)

I still enjoy the things I used to enjoy:

- Definitely as much
- Not quite so much
- Only a little
- Hardly at all

HADS 2 (A)

I get a sort of frightened feeling as if something awful is about to happen:

- Very definitely & quite badly
- Yes, but not too badly
- A little, but it doesn't worry me
- Not at all

HADS 2 (D)

I can laugh & see the funny side of things:

- As much as I always could
- Not quite so much now
- Definitely not so much now
- Not at all

HADS 3 (A)

Worrying thoughts go through my mind:

- A great deal of the time
- A lot of the time
- From time to time, but not too often
- Only occasionally

-----Page Break-----

HADS 3 (D)

I feel cheerful:

- Not at all
- Not often
- Sometimes
- Most of the time

HADS 4 (A)

I can sit at ease & feel relaxed:

- Definitely
- Usually
- Not Often
- Not at all

-----Page Break-----

HADS 4 (D)

I feel as if I am slowed down:

- Nearly all the time
- Very often
- Sometimes
- Not at all

HADS 5 (A)

I get a sort of frightened feeling, like 'butterflies' in the stomach:

- Not at all
- Occasionally
- Quite Often
- Very Often

HADS 5 (D)

I have lost interest in my appearance:

- Definitely
- I don't take as much care as I should
- I may not take quite as much care
- I take just as much care as ever

HADS 6 (A)

I feel restless as if I have to be on the move:

- Very much indeed
- Quite a lot
- Not very much
- Not at all

HADS 6 (D)

I look forward with enjoyment to things:

- As much as I ever did
- Rather less than I used to
- Definitely less than I used to
- Hardly at all

HADS 7 (A)

I get sudden feelings of panic:

- Very often indeed
- Quite often
- Not very often
- Not at all

HADS 7 (D)

I can enjoy a good book, radio or TV programme:

- Often
- Sometimes
- Not often
- Very seldom



Import from library

Add new question

Add Block

▼ SSCI

SSCI-1

Because of my illness, some people seemed uncomfortable with me

- Never
- Rarely
- Sometimes
- Often
- Always

SSC1-2

Because of my illness, some people avoided me

- Never
- Rarely
- Sometimes
- Often
- Always

SSC1- 3

Because of my illness, I felt left out of things

- Never
- Rarely
- Sometimes
- Often
- Always

SSCI-4

Because of my illness, people were unkind to me

- Never
- Rarely
- Sometimes
- Often
- Always

SSCI-5

Because of my illness, people avoided looking at me

- Never
- Rarely
- Sometimes
- Often
- Always

SSCI-6

I felt embarrassed about my illness

- Never
- Rarely
- Sometimes
- Often
- Always

SSCI-7

I felt embarrassed because of my physical limitations

- Never
- Rarely
- Sometimes
- Often
- Always

SSCI-8

Some people acted as though it was my fault I have this illness

- Never
- Rarely
- Sometimes
- Often
- Always

▼ LMSQOL

LMSQOL- 1

★ x→

My health has affected my relationships with my family

- most of the time
- quite often
- sometimes
- Not at all

LMSQOL-2

★ x→

I have felt lonely

- most of the time
- Quite often
- sometimes
- Not at all

LMSQOL- 3

★ x→

I have felt good about my appearance

- most of the time
- Quite often
- sometimes
- Not at all

LMSQOL- 4

★ x→

I have worried about my health

- most of the time
- Quite often
- sometimes
- Not at all

LMSQOL-5

★ x→

I have worried about other people's attitudes towards me

- most of the time
- Quite often
- sometimes
- Not at all

LMSQOL-6

★ x→

I have felt tired

- most of the time
- quite often
- sometimes
- Not at all

LMSQOL-7

★ x→

I have had as much energy as usual

- most of the time
- quite often
- sometimes
- Not at all

----- Page Break -----


LMSQOL-8

★ x→

I have felt happy about the future

- most of the time
- quite often
- sometimes
- Not at all



 Import from library

Add new question

Add Block

Q64



Debrief Form

Thank you for completing the questionnaire and providing your responses. Your time and engagement is greatly appreciated. Should you require any other information, or wish to discuss your responses further, please email the primary researcher below.

There are over 130,000 people living with Multiple Sclerosis (MS) in the UK. Nearly 7000 people are diagnosed each year. It is important to continue to investigate this neurological condition and how it affects individuals and their well-being. Your responses will help to advance on the existing literature. The responses will be collated as part of a doctorate thesis submission by the primary researcher at the University of Edinburgh in 2023. A poster presentation and accessible summary will be made available via social media groups and NHS neurology clinic(s).

If you are worried about any of your current symptoms relating to MS or wellbeing in general, please contact your GP. For more information relating to MS, visit <https://www.nhsinform.scot/illnesses-and-conditions/brain-nerve-and-spinal-cord/multiple-sclerosis-ms>

To find out about MS related research, please visit:
<https://www.mssociety.org.uk/research/take-part-in-ms-research>

Many thanks for your time!

Craig Mackay (Primary Researcher/ Trainee Clinical Psychologist)

Craig.Mackay@lanarkshire.scot.nhs.uk



 Import from library

Add new question

[Add Block](#)

End of Survey

We thank you for your time spent taking this survey.

Your response has been recorded.