Does a lack of social support and perceived stigma influence the relationship between motor neurone disease-related stress and psychological distress?

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Objectives. This study aimed to investigate the mechanisms through which social support and felt stigma influence the relationship between motor neurone disease (MND)-related stress and psychological distress for people with MND. Although a lack of social support has been identified as a significant predictor of psychological distress for individuals with MND, the mechanisms through which this relationship exists have not been assessed, nor have the predictive nature of stigma. Furthermore, the theoretical model specifying the effects of enacted stigma on self-stigma has not been tested in individuals with MND.

Design. A cross-sectional design utilizing an online survey method was used. It was hypothesized that social support would moderate the relationship between MND-related stress (operationalized as enacted stigma or physical functioning) and psychological distress (operationalized as depression, anxiety, and stress). Furthermore, felt stigma would significantly mediate the relationship between MND-related stress (enacted stigma) and psychological distress.

Methods. Individuals with a diagnosis of MND were recruited internationally through social media and through various organizations and support services. Seventy-seven participants completed the online survey.

Results. Significant correlations were identified between social support, felt, and enacted stigma and psychological distress. Moderation analysis was not significant. However, the mediation analyses identified felt stigma as a significant mediator of the relationship between enacted stigma and psychological distress. A direct relationship between enacted stigma and stress (but not depression and anxiety) was also evident.

Conclusions. A comprehensive approach to tackling stigma is important in ameliorating psychological distress for people with MND. Limitations of the current study are discussed, along with implications for clinical practice.

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Statement of contribution

What is already known on this subject?
- Individuals with MND often experience psychological distress but research on why this is the case is more sparse.
- In other conditions with perceivable physical effects, different types of stigma are known to predict poorer psychological well-being but this has not been measured or theoretically modelled in MND.
- The role of social support in predicting psychological distress has been more researched, but its theoretical role is also underspecified.

What does this study adds?
- The theoretical relationship from enacted stigma to psychological distress via self-stigmatization was confirmed.
- No role for the buffering effect of social support was found although study limitations were relevant.
- Stigma should be a target for more socially engaged interventions building on psychological understandings.

Background

Motor neurone disease (MND), also referred to as amyotrophic lateral sclerosis (ALS) and Lou Gehrig disease in the United States, is a life-limiting neurodegenerative condition. The condition progressively destroys the motor neurones in the brain and spinal cord and alters an individual’s ability to control voluntarily their muscle movements, leading to paralysis, swallowing difficulties, respiratory failure, and, ultimately, death (King, Mulligan & Stansfield, 2014). The effects of MND are not limited to motor functions; behaviour difficulties, cognitive impairment (McCluskey et al., 2009; Strong et al., 1999), emotional difficulties (depression, anxiety, and anger) and involuntary changes in mood (Orrell, 2016) are also common. With a prevalence rate for North America and Europe of around two per 100,000 of the population (Worms, 2001), median survival rates following symptom onset are generally only two to four years, with only 10–20% of individuals surviving past 10 years (Chio et al., 2009).

Current interventions for individuals with MND mainly focus upon the physical aspects of the condition, to maintain physical and biological functioning and quality of life for as long as possible (Andersen et al., 2012). However, a recent mixed-method review on the supportive needs of this group (Oh & Kim, 2017) reported that of the 37 studies included, only around half discussed psychosocial needs, concluding a significant need for more psychological, social, and emotional support, alongside physical and practical assistance. The authors also proposed that the psychological impact of receiving, adjusting, and coping with this diagnosis should be considered equally with the physical impact of the condition. Individuals with a diagnosis of MND are reported to experience heightened psychological distress and decreased well-being (Hogg, Goldstein & Leigh, 1994; Lou, Reeves, Benice & Sexton, 2003; Montgomery & Erickson, 1987; Tedman, Young & Williams, 1997; Vignola et al., 2008). Although several factors undoubtedly contribute to this (e.g. biological, social, psychological, and ‘spiritual’ suffering: Ganzini, Johnston & Hoffman, 1999; social withdrawal: Rigby et al., 1999; and physical impairment: Hunter, Robinson & Neilson, 1993; Hogg et al., 1994), recent research in other physical health conditions has suggested that stigma may be an important variable to consider. This has been identified as important in health conditions with perceivable physical effects such as epilepsy (Baker, Eccles &
The term stigma was originally defined by Goffman (1963) as 'an undesired differentness' (p. 5), and this has since been developed to include two concepts of stigma: felt and enacted (Scambler & Hopkins, 1986). ‘Felt stigma’ refers to a feeling of shame about being different and feeling that discrimination for this difference will occur, whereas ‘enacted stigma’ refers to actual experience of this discrimination. Link and Phelan (2001) further developed the concept and described stigma as a set of components which include labelling, stereotyping, separation, status loss, and discrimination. Therefore, the term stigma can encompass a range of negative actions and associations that can be attributed to an individual based on their perceived differentness. In relation to MND, only one study has quantitatively assessed stigma (van der Beek, Bos, Middel & Wynia, 2013). Although the authors found that stigma was a major predictor of poorer quality of life, with ‘felt stigma’ a stronger predictor than ‘enacted stigma’, the study’s outcome measure was quality of life, rather than psychological distress in particular, and individuals diagnosed with MND only made up 9% of the total number of participants. Furthermore, no research has been conducted to assess the role that felt stigma plays in mediating the relationship between enacted stigma and psychological distress, as theorized in the self-stigma model proposed by Corrigan and Watson (2002) and Corrigan, Watson and Barr (2006).

Another variable which has been less studied in motor neurone disease as opposed to other neurodegenerative conditions is social support. Research has identified social support as a significant predictor of psychological distress (conceptualized as depression and quality of life; Matuz, Birbaumer, Hautzinger & Kübler, 2010). Furthermore, longitudinal research has also identified social support as a significant predictor of depression and quality of life (Matuz, Birbaumer, Hautzinger, & Kübler, 2015) and depression and anxiety (Goldstein, Atkins, Landau, Brown, & Leigh, 2006). Despite research identifying social support as a significant predictor of psychological distress for individuals with MND, no research has been conducted, to the best of the authors' knowledge, to assess whether social support acts as a moderator between MND-related stress and psychological distress. This moderating effect of social support would suggest that social support could act as a ‘buffer’ on the relationship between MND-related stress and psychological distress, as proposed in the stress buffering model (Barrera, 1986; Cohen & Wills, 1985; Cutrona & Russell, 1990; Thoits, 1986).

Consequently, this study aims to investigate the mechanisms through which perceived stigma and social support influence the relationship between MND-related stress and psychological distress for people with MND. The research aim was to investigate whether increased levels of perceived stigma and lower levels of social support influenced the relationship between MND-related stress and psychological distress. It was hypothesized that both enacted and felt stigma would be significant predictors of psychological distress in individuals with MND and that felt stigma would significantly mediate the relationship between enacted stigma and psychological distress (depression, anxiety, and stress). Furthermore, it was hypothesized that social support would significantly moderate the relationship between MND-related stress and psychological distress.
Method

Design
A quantitative cross-sectional survey design was used to investigate the following: (1) whether felt stigma mediated the relationship between enacted stigma and psychological distress, and (2) whether social support had a moderating effect on the relationship between MND-related stress (conceptualized as scores on a measure of both physical functioning and enacted stigma) and psychological distress (depression, anxiety, and stress) in individuals with a diagnosis of MND. Both the mediation and moderation analyses were conducted using Hayes’ PROCESS Tool (Hayes, 2018).

Participants
Individuals aged 18 years or over who had a diagnosis of MND/ALS/Lou Gehrig disease and who could complete an online survey written in English (either alone or with assistance from another person) were eligible to take part. An opportunistic sampling method was employed as participants volunteered to participate following advertisement of the study details online, using social media and through international organizations. If a participant wished to take part in the study, then they accessed the study link provided in the study advert and were given the opportunity to read and download the participant information sheet. Eligibility for the study was based on self-report and was recorded through a demographic questionnaire at the beginning of the survey.

The literature was consulted to determine the required sample size to detect the mediated effect and based on using an indirect effect method of mediation with bias-corrected bootstrapping and medium effect size in both arms (\(a = .39\) and \(b = .39\)), and 71 participants were needed for 80% power (Fritz & MacKinnon, 2007). For moderation analyses, effect sizes are typically small (Aguinis, Beaty, Boik, & Pierce, 2005), with suggested sizes for \(f^2\) of 0.005, 0.01, and 0.025 for small, medium, and large effects, respectively (Kenny, 2018). A large effect size would need 316 participants to detect an effect (\(G^*\)power; Faul, Erdfelder, Buchner, & Lang, 2009). A total of 77 participants were recruited, 34 women and 43 men (\(M_{\text{age}} = 59.14\)). Of the 94 participants who opened the survey, 84 completed the demographic data, with only 80 of these continuing to complete the outcome measures. Of these, only 78 completed all the measures (two participants did not complete the physical functioning measure and their data were withdrawn). A further participant’s data were withdrawn due to not meeting the inclusion criteria of being completed by an individual with a diagnosis of MND. See Table 1 for participants’ self-reported demographic characteristics.

Procedure
Ethical approval was gained from the first author’s host academic institution after input from service users. Recruitment took place online from a variety of sources, mainly through international organizations supporting people with MND. This was achieved by sharing the study advert and link on social media (Facebook and Twitter) pages by the Motor Neurone Disease Association (MNDA) Australia, MNDA UK, MNDA New Zealand, MNDA Scotland, ALS Association Canada, ALS Society Quebec, Minds and Movement, and the first author’s own contacts. An online survey was constructed using Qualtrics software which was used to collect the data. Prior to completing the survey, each participant
accessed the participant information sheet and then consented to take part. Following completion of the questionnaires, a debrief sheet was available which also provided the contact details of organizations who could provide support if participation caused any distress.

**Measures**

To control for potential confounders, measures were included to assess demographic variables (age and gender), along with the three variables of interest (physical functioning, social support and stigma). One measure was used to assess the three outcomes of psychological distress (measuring depression, anxiety, and stress). To situate the sample, additional variables were collected including nationality, relationship status, time since symptom onset and time since diagnosis.

| Characteristic                          | N   | %    |
|----------------------------------------|-----|------|
| Male                                   | 43  | 55.8 |
| Female                                 | 34  | 44.2 |
| Nationality                            |     |      |
| Australian                             | 8   | 5.8  |
| British/English/Welsh/Northern Irish   | 18  | 23.4 |
| Canadian                               | 14  | 18.2 |
| Dutch                                  | 1   | 0.7  |
| German                                 | 1   | 0.7  |
| Indian                                 | 1   | 0.7  |
| Irish                                  | 3   | 2.2  |
| New Zealander                          | 18  | 23.4 |
| NZ European                            | 3   | 2.2  |
| South African                          | 4   | 2.9  |
| Swedish                                | 1   | 0.7  |
| USA                                    | 2   | 1.4  |
| USA Canadian                           | 1   | 0.7  |
| Unknown                                | 2   | 1.4  |
| Relationship status                    |     |      |
| Single                                 | 3   | 2.2  |
| Cohabiting/married/civil partnership   | 58  | 71.4 |
| Divorced                               | 9   | 6.5  |
| Widowed                                | 4   | 2.9  |
| Other                                  | 3   | 2.2  |

**Carer assistance required to complete survey**

|                        |     |      |
|------------------------|-----|------|
| Yes                    | 7   | 9.1  |
| No                     | 70  | 90.9 |

*Note.* Participants were on average 59.1 years old (SD = 10.6; range = 36–83). Participants had an average time since symptom onset of 4.8 years (SD = 4.4; median = 3.5; range = 0.6–22). Participants had an average time since diagnosis of 3.5 years (SD = 3.9; range = 0.1–21).
Physical functioning

The Self-Administered Amyotrophic Lateral Sclerosis Functional Rating Scale – Revised (SA-ALSFRS-R; Cedarbaum et al., 1999; Montes et al., 2006) includes 12 questions which assess the domains of motor function, bulbar symptoms, and breathing ability in individuals with MND. Individual items are rated on a scale of 0–4 with a total score range of 0–48; higher scores indicate higher levels of physical functioning. Although there is no current evidence regarding the reliability and validity of the SA-ALSFRS-R, there is evidence regarding the clinician-administered ALSFRS-R. This has been shown to be a reliable and valid measure by the authors during development (Cedarbaum et al., 1999). Montes et al. (2006) compared the use of the SA-ALSFRS-R to the clinician-administered ALSFRS-R and reported an intraclass correlation coefficient score of \( r = 0.93 \), implying that the self-administered version is as reliable as the clinician-administered version. This measure was chosen as it is aimed specifically at individuals with a diagnosis of MND to assess their physical functioning and symptom severity. The use of the ALSFRS-R instrument online compared to on-site face-to-face assessment was assessed by Maier et al. (2012), and their results supported the use of the measure online, due to a highly significant correlation between on-site evaluation and online testing (\( r = 0.96 \)).

Social support

The Multidimensional Scale of Perceived Social Support (MSPSS; Zimet, Dahlem, Zimet & Farley, 1988) is a 12-item measure scored on a scale from 1 (very strongly disagree) to 7 (very strongly agree), with a total score range of 12–84; the scale contains three subscales (four items in each): significant other, family, and friends (scores range from 4 to 28 for each subscale). Both the total scale and the individual subscales can be used, with higher scores indicating higher levels of social support. The authors report highly acceptable Cronbach’s coefficient alpha coefficients and good test–retest reliability. The scale is relatively short, easy to complete, and measures individuals’ levels of social support from different sources.

Stigma

The Stigma Scale for Chronic Illness (SSCI, Rao et al., 2009) comprises 24 questions with two subscales, one scale for felt stigma (labelled self-stigma; 13 questions) and one scale for enacted stigma (11 questions). Each item is scored on a scale of 0 (never) to 4 (always) with a total score range of 0–96 for the full scale, 0–52 for the felt scale and 0–44 for the enacted scale. Higher scores indicate a higher level of stigmatization. Internal reliability for this scale was assessed by the authors and was found to be highly acceptable. The measure was specifically developed for people with chronic neurological illnesses, including individuals with MND. The total score was used alongside the two subscales (felt and enacted stigma) for the correlational analyses for this study, and the two subscales were used in the regression analyses of this study. As part of the validation process, it was administered online (Rao et al., 2009).

Psychological distress

The Depression, Anxiety and Stress Scale (DASS-21; Lovibond & Lovibond, 1995) is a 21-question scale with three subscales, depression, anxiety, and stress (seven questions for each subscale). These are scored on a range of 0 (never) to 3 (almost always) with a total
range of 0–21 for each of the subscales; higher scores indicate a higher level of distress, and cut-offs have been created to categorize scores as: ‘normal’, ‘mild’, ‘moderate’, ‘severe’, or ‘extremely severe’. The scale has excellent internal reliability both in its full form and in the three subscales (Antony, Bieling, Cox, Enns, & Swinson, 1998; Henry & Crawford, 2005). The measure has been used online in previous research with individuals with MND (Caga, Ramsey, Hogden, Mioshi & Kiernan, 2015; Lillo, Mioshi, Zoing, Kiernan & Hodges, 2011).

**Data analysis**

Data analysis was completed using SPSS (Version 26) and Hayes PROCESS macro Version 3.4.1. The sample was checked for missing data prior to any analysis being conducted; seven participants had not provided a response to the same question on the SSCI: ‘people with my illness lost their jobs when their employers found out’. This missing datum was replaced with the mean value of this specific subscale of the SSCI (enacted stigma subscale) for each person. No other missing data were identified in the sample. Outliers were identified using boxplots, and scores were checked for errors. Given the relatively small sample size, a less conservative method (dividing the skewness or kurtosis value by its standard error) was used to assess for skewness and kurtosis (Field, 2005). Using parameters of $-3$ to $+3$, the only scale not normally distributed was the MSPSS total score.

As the MSPSS total score was not normally distributed, non-parametric correlations were calculated using Spearman’s correlation coefficients to identify relationships. Mediation analyses were then conducted; in each analysis the predictor variable was enacted stigma, the mediator variable was felt stigma and the outcome variable was either depression, anxiety, or stress. Each analysis was based on 5,000 bootstrap samples to estimate the confidence intervals. Moderation analyses were then conducted to assess the moderating effects of social support on the relationship between MND-related stress and psychological distress. In each analysis, the predictor variable to represent MND-related

| Variable                  | Mean  | SD    | Cronbach’s alpha |
|---------------------------|-------|-------|------------------|
| DASS-21 stress            | 7.03  | 4.59  | .86              |
| DASS-21 anxiety           | 5.32  | 3.85  | .75              |
| DASS-21 depression        | 7.19  | 5.22  | .91              |
| SA-ALSFRS-R               | 29.00 | 8.62  | .82              |
| MSPSS total               | 66.47 | 15.66 | .94              |
| MSPSS significant other   | 24.10 | 5.68  | .94              |
| MSPSS family              | 22.16 | 6.16  | .90              |
| MSPSS friends             | 20.21 | 6.27  | .91              |
| SSCI total                | 32.83 | 16.12 | .92              |
| SSCI self-subscale        | 22.48 | 10.36 | .90              |
| SSCI enacted subscale     | 10.09 | 7.09  | .87              |

*Note. DASS-21 = Depression, Anxiety and Stress Scale (measuring psychological distress); MSPSS = Multidimensional Scale of Perceived Social Support (measuring social support); SA-ALSFRS-R = Self-Administered Amyotrophic Lateral Sclerosis Functional Rating Scale Revised (measuring physical functioning); SSCI = Stigma Scale for Chronic Illness (measuring stigma); SSCI Self-subscale measures ‘felt stigma’ and SSCI Enacted Subscale measures ‘enacted stigma’.*
stress was either enacted stigma or physical functioning and the outcome variable was either depression, anxiety, or stress, with social support as the moderating variable.

All regressions analyses which contributed to the mediations, and moderations were checked to ensure that they met assumptions of independent errors, homoscedasticity, and normalized residuals and that no multicollinearity was present.

**Results**

The mean scores, standard deviations, and Cronbach’s alpha scores for each measure are reported in Table 2.

The mean (SD) score for the SA-ALSFRS-R was 29 (8.62), indicating that levels of independent functioning were within the mid-point range. The majority of scores fell within the ‘mild to moderate’ category (43%, \( n = 33 \)), with ‘moderate to severe’ (30%, \( n = 23 \)), ‘advanced disease’ (16%, \( n = 12 \)), and the ‘minimal to mild’ category (12%, \( n = 9 \)).

The mean score for the MSPSS total (\( M = 66.47; SD = 15.66 \)) indicated that levels of social support were towards the higher end of the scale for this sample (possible range: 12–84). Moreover, the mean score for the SSCI total score (\( M = 32.83; SD = 16.12 \)) indicated that levels of stigma were quite low in this sample (possible range: 0–96, with higher scores indicating higher levels of stigma).

Regarding the DASS scores, for the depression subscale, the majority of scores were in the ‘normal’ category (70%, \( n = 54 \)), followed by the ‘mild’ (18%, \( n = 14 \)), and the ‘moderate’ category (12%, \( n = 9 \)). For the anxiety subscale, all the scores fell within the ‘normal’ category (100%, \( n = 77 \)). For the stress subscale, the majority of scores were in the ‘normal’ category (62%, \( n = 48 \)), followed by ‘moderate’ (22%, \( n = 17 \)), ‘mild’ (9%, \( n = 7 \)), ‘severe’ (5%, \( n = 4 \)), and finally the ‘extremely severe’ category (1% \( n = 1 \)). This indicates that the majority of the sample had non-clinical levels of stress, anxiety, and depression.

**Correlations**

Spearman’s \( r_s \) correlation coefficients were calculated to determine the relationships between the variables; see Table 3.

**Table 3. Correlation matrix of Spearman’s correlation coefficients**

| Variable   | 1   | 2   | 3   | 4   | 5   | 6   | 7   | 8   | 9   |
|------------|-----|-----|-----|-----|-----|-----|-----|-----|-----|
| 1. Age     | 1.00| .090| -.031| .198| -.134| -.140| -.179| -.175| -.026| -.052|
| 2. Gender  | 1.00| -.165| -.097| .028| .014| -.016| .114| -.067| -.070|
| 3. SA-ALSFRS-R | 1.00| .255*| -.402**| -.395**| -.232*| -.153| -.209| -.180|
| 4. MSPSS total | 1.00| -.483**| -.483**| -.433**| -.385**| -.399**| -.437**|
| 5. SSCI total   | 1.00| .929**| .801**| .538**| .447**| .660**|
| 6. SSCI self    | 1.00| .586**| .525**| .526**| .689**|
| 7. SSCI enacted | 1.00| .440**| .244*| .465**|
| 8. DASS-21 stress | 1.00| .627**| .787**|
| 9. DASS-21 anxiety | 1.00| .588**|
| 10. DASS-21 depression | 1.00|   |

Note. *\( p < .05 \); **\( p < .01 \).
As can be seen in Table 3, none of the demographic or clinical variables correlated significantly with the three outcome variables (depression, anxiety, or stress). However, statistically significant relationships were found between the two stigma subscales and social support variables and all three outcome variables. Associations were in the predicted direction with social support (total score) negatively correlated with depression, anxiety, and stress and stigma (total score and the two subscales) positively correlated with the three outcome measures.

**Mediation analyses**

As physical functioning significantly correlated with both stigma variables, this was initially included in the mediation models as a covariate. Upon further inspection, this variable did not make any significant contribution to the model or impact the findings greatly; therefore, this was not included in the final models. In each analysis, the predictor variable was enacted stigma, the mediator variable was felt stigma, and the outcome variable was either depression, anxiety, or stress. Mediation analyses were conducted to assess the mediating effects of felt stigma; see Table 4 and Figures 1–3 for the main findings of these analyses.

**Mediation model for depression**
The total effect of the model was significant \( (c = .401, 95\% \text{ CI } [0.259, 0.543], p < .001) \). Higher enacted stigma predicted higher felt stigma \( (a = .941, 95\% \text{ CI } [0.683, 1.198], p < .001) \), and higher felt stigma predicted higher depression scores \( (b = .316, 95\% \text{ CI } [0.211, 0.420], p < .001) \). A significant indirect effect was found for enacted stigma through felt stigma on depression scores \( (ab = .297, 95\% \text{ CI } [0.191, 0.429]) \). The effect size of the completely standardized indirect effect was .404. The direct effect of the model did not remain significant when controlling for the effect of the mediational variable of felt stigma \( (c' = .104, 95\% \text{ CI } [-0.049, 0.257], p > .05) \).

**Mediation model for anxiety**
The total effect of the model was significant \( (c = .196, 95\% \text{ CI } [0.080, 0.312], p < .01) \). Higher enacted stigma predicted higher felt stigma \( (a = .941, 95\% \text{ CI } [0.683, 1.198], p < .001) \), and higher felt stigma predicted higher anxiety scores \( (b = .206, 95\% \text{ CI } [0.113, 0.300], p < .001) \). A significant indirect effect was found for enacted stigma through felt stigma on anxiety scores \( (ab = .1941, 95\% \text{ CI } [0.092, 0.313]) \). The effect size of the completely standardized indirect effect was .358. The direct effect of the model did not remain significant when controlling for the effect of the mediational variable of felt stigma \( (c' = .002, 95\% \text{ CI } [-0.134, 0.138], p > .05) \).

**Mediation model for stress**
The total effect of the model was significant \( (c = .365, 95\% \text{ CI } [0.242, 0.488], p < .001) \). Higher enacted stigma predicted higher felt stigma \( (a = .941, 95\% \text{ CI } [0.683, 1.198], p < .001) \), and higher felt stigma predicted higher stress scores \( (b = .193, 95\% \text{ CI } [0.092, 0.294], p < .001) \). A significant indirect effect was found for enacted stigma through felt stigma on stress scores \( (ab = .182, 95\% \text{ CI } [0.069, 0.278]) \). The effect size of the completely standardized indirect effect was .281. The direct effect of the model remained significant...
### Table 4. Mediation model of the indirect effect of MND-related stress (enacted stigma) through felt stigma on psychological distress (N = 77)

| Analyses                                      | Point estimate | SE  | 95% CI          | Completely standardized effect |
|-----------------------------------------------|----------------|-----|-----------------|-------------------------------|
| **Depression model**                          |                |     |                 |                               |
| Enacted Stigma → Felt Stigma (IV to mediator, path a) | .941***        | .129| [0.683, 1.198]  |                               |
| Felt Stigma → Depression (mediator to DV, path b)    | .316***        | .053| [0.211, 0.420]  |                               |
| Enacted Stigma → Depression (total effect, path c)  | .401***        | .071| [0.259, 0.543]  |                               |
| Enacted Stigma → Depression (direct effect, path c') | .104           | .077| [−0.049, 0.257] |                               |
| Enacted Stigma → Depression (indirect effect, path a × b) | .297\(^a\)      | .061| [0.191, 0.429]  | 0.404                         |
| **Anxiety model**                              |                |     |                 |                               |
| Enacted Stigma → Felt Stigma (IV to mediator, path a) | .941***        | .129| [0.683, 1.198]  |                               |
| Felt Stigma → Anxiety (mediator to DV, path b)      | .206***        | .047| [0.113, 0.300]  |                               |
| Enacted Stigma → Anxiety (total effect, path c)     | .196**         | .058| [0.080, 0.312]  |                               |
| Enacted Stigma → Anxiety (direct effect, path c')   | .002           | .068| [−0.134, 0.138] |                               |
| Enacted Stigma → Anxiety (indirect effect, path a × b) | .194\(^a\)      | .056| [0.092, 0.313]  | 0.358                         |
| **Stress model**                                |                |     |                 |                               |
| Enacted Stigma → Felt Stigma (IV to mediator, path a) | .941***        | .129| [0.683, 1.198]  |                               |
| Felt Stigma → Stress (mediator to DV, path b)       | .193***        | .051| [0.092, 0.294]  |                               |
| Enacted Stigma → Stress (total effect, path c)      | .365***        | .062| [0.242, 0.488]  |                               |
| Enacted Stigma → Stress (direct effect, path c')    | .184*          | .074| [0.036, 0.332]  |                               |
| Enacted Stigma → Stress (indirect effect, path a × b) | .182\(^a\)      | .052| [0.069, 0.278]  | 0.281                         |

Note. \(^a\)Due to the way that the indirect effect is calculated, only a confidence interval, but not a p-value, is available.; *p < .05; **p < .01; ***p < .001.
when controlling for the effect of the mediational variable of felt stigma ($c' = .184$, 95% CI [0.036, 0.332], $p < .05$).
Moderation analysis

To assess the moderating effects of social support on the relationship between MND-related stress and psychological distress, moderation analyses were conducted. In each analysis, the predictor variable to represent MND-related stress was either enacted stigma or physical functioning and the outcome variable was either depression, anxiety, or stress with social support as the moderating variable. There was no significant moderation effect of social support found in any of the models. For MND-related stress, conceptualized as either enacted stigma or physical functioning, neither of the models including stress were significant (\( p = .179 \) for enacted stigma; \( p = .237 \) for physical functioning), neither of the models including anxiety were significant (\( p = .789 \) and \( p = .816 \), respectively) and nor were those models including depression (\( p = .164 \) and \( p = .379 \), respectively).

Discussion

The present study investigated the impact of social support and perceived stigma on the relationship between MND-related stress and psychological distress in individuals with MND. Statistically significant correlations were found between social support, stigma (total score, self-(felt) subscale and enacted subscale) and all the outcome variables in the predicted directions. The correlational findings suggest that individuals with higher levels of social support had lower levels of psychological distress (depression, anxiety, and stress) and that individuals with higher levels of stigma (both felt and enacted) also experienced higher levels of psychological distress.

To explore these relationships further, mediation and moderation analyses were conducted. In relation to the role of felt stigma in mediating the relationship between MND-related stress (enacted stigma) and psychological distress (depression, anxiety, and stress), analyses yielded significant results. The effect of felt stigma as a mediator of the relationship between enacted stigma and psychological distress was significant. Each of the mediation models found a significant indirect effect via this mediator. This suggests that the relationship between enacted stigma and psychological distress is mediated by the individual’s levels of felt stigma. These findings are consistent with the theoretical model of self-stigma proposed by Corrigan and Watson (2002) and Corrigan, Watson and Barr (2006), whereby public attitudes (enacted stigma) produce personal responses and self-stigmatization which then leads to negative consequences for the individual.

This process begins when an individual with a condition that induces stigmatization experiences discrimination (enacted stigma) which makes them aware of the negative stereotypes attached to their condition. This awareness of the negative stereotype is conceptualized as felt (perceived) stigma and the actual experience of discrimination (e.g. social exclusion) are conceptualized as enacted stigma. The process can lead to the individual agreeing with the negative stereotypes and then internalizing the stereotype. This internalization is conceptualized as self-stigma, which then results in consequences for the self (such as psychological distress). The results of the present study lend support to this model as it identified a significant indirect effect of felt stigma on the relationship between enacted stigma and depression, anxiety, and stress. However, the direct path between enacted stigma and stress also remained significant after mediation analysis, suggesting that the actual experience of discrimination also relates to stress levels for individuals with MND independent of the process of internalization of the negative stereotype. It is interesting to note that theoretical accounts of the effects of disablism from other disciplines, such as the social model of disability (Thomas, 2007) and the later
concept of psycho-emotional disablism (see Simpson & Thomas, 2014), would also predict a direct route from hostile disablist attitudes to well-being and an indirect one via the internalization of these attitudes. Moderation analyses revealed no significant effect for the role of social support as a moderator on the relationship between MND-related stress (enacted stigma or physical functioning) and psychological distress (depression, anxiety, and stress).

**Limitations**

While this study has used established procedures to measure mediation, it is still acknowledged that this approach is limited in confirming causal inferences (Levin, 2006). Sample size is a further limitation in this study, as only a relatively small sample size was achieved. The suggested sample size to achieve a medium effect for the moderation analysis was 472 participants, and this study only recruited 77 participants, falling well below the suggested requirement. Due to the relative rarity of the condition, with prevalence rates for North America and Europe of around two per 100,000 of the population (Worms, 2001), and its rapid degenerative effect, recruiting enough participants to achieve a sample size to test moderation was challenging. While an online survey method was used to extend recruitment internationally, this did rely on the cooperation of organizations and support services. For example, only one local organization in the United States shared this information, resulting in only a small proportion (3%) of participants from this country.

Levels of stigma were quite low in this sample and most of the sample had ‘normal’ levels of stress, anxiety, and depression. A review of psychosocial aspects of MND (McLeod & Clarke, 2007) identified varying levels of depression and anxiety within MND populations; these ranged from 0% to 50% for depression (defined as moderate to severe) and 11% to 26% for anxiety. Therefore, given the relatively high variability, it is unclear whether the current sample may be considered representative although the present findings may not be applicable to samples with higher levels of stigma and psychological distress. Furthermore, participants in this study were mainly longer surviving individuals, which again might limit generalizability, with the very low levels of depression, anxiety, and stress not similar in more recently diagnosed individuals.

Finally, this study had a very highly selected sample as, despite at least 16 counties included, only a small number from each country answered the questionnaire. Given the online format, this also restricted or privileged those individuals interested in this topic and who had access to an online platform.

**Clinical implications**

The findings from this study provide implications for clinical practice. In particular, results suggest that if psychological distress is identified in an individual with MND, then it may be beneficial to use both social and psychological factors to inform formulations and be aware of the potential of both routes to impact when considering a comprehensive intervention plan.

Interventions aimed at targeting stigma often operate on several levels: intrapersonal, interpersonal, organizational/ institutional, community, and governmental/ structural (Heijnders & Van Der Meij, 2006). Systematic reviews have identified that the most effective interventions are aimed at the intrapersonal, interpersonal, and community levels (Heijnders & Van Der Meij, 2006; Rao et al., 2019). Effective intervention strategies
for reducing stigma for conditions such as HIV, mental health diagnoses, and leprosy include education (Ngoc, Weiss & Trung, 2016), counselling (Lusli et al., 2016), cognitive behavioural therapy (Corrigan & Calabrese, 2005), social marketing (Henderson et al., 2012), drama therapy (Orkibi, Bar & Eliakim, 2014), and social support groups (Thurman, Jarabi & Rice, 2012), and combinations of these (Uys et al., 2009). Moreover, research suggests that mindfulness is positively associated with stigma resistance in individuals with a psychiatric diagnosis (Chan, Lee & Mak, 2018) suggesting that mindfulness-based interventions may be beneficial in bolstering the self against the effects of enacted stigma. Mindfulness-based interventions have also been identified as effective in reducing psychological distress for individuals with a diagnosis of MND (Pagnini et al., 2017; Pagnini et al., 2015) which could make them useful for a number of adaptive purposes.

However, individually focused interventions are clearly not sufficient on their own to tackle stigma and, for example using the concept of psycho-emotional disablism (Thomas, 2007), it is also important to look at addressing the limitations and barriers society impose on individuals with impairments. Socially engaged activism, while not the usual intervention route for health and clinical psychologists (Simpson & Thomas, 2014), is important in effecting societal change to highlight and break down these structural barriers.

**Conclusion**
Mediation analyses identified significant findings for the indirect effect of felt stigma on the relationship between MND-related enacted stigma and psychological distress (depression, anxiety, and stress), while a direct route from enacted stigma to psychological distress also remained in the case of stress (one of the three relationships examined). Moderation analysis revealed no significant role for social support as a moderator of the relationship between MND-related stress and psychological distress. These findings should be used to improve interventions for individuals with a diagnosis of MND as they highlight the importance of considering both social and psychological factors when psychological distress has been identified. For individuals with MND, the roots and causes of psychological distress are complex – and our approach to intervention needs to acknowledge this.

**Conflicts of interest**
All authors declare no conflict of interest.

**Author contributions**
Natalie Leigh (Conceptualization; Formal analysis; Investigation; Methodology; Project administration; Writing – original draft) Jane Simpson (Conceptualization; Methodology; Supervision; Validation; Writing – review & editing) Fiona J.R. Eccles (Formal analysis; Methodology; Supervision; Writing – original draft; Writing – review & editing).
Data availability statement

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

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