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Development and evaluation of a digital mental health intervention for people with Motor Neurone Disease and their caregivers

by

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Thesis for the degree of Doctor of Philosophy

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Abstract

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Motor neurone disease (MND) is a neurodegenerative disease which results in declining physical function and has a very poor prognosis. It causes progressive muscle weakness in the limbs, difficulties communicating, difficulties with swallowing and eating, breathlessness as well as other sensory and cognitive symptoms. People with MND and caregivers can experience high levels of psychological distress and burden. Psychological support can help improve quality of life, however research evidence for psychological interventions to support people with MND and caregivers is sparse and more research is urgently needed. The overall aim of this thesis was to develop and explore the use of a digital intervention for psychological support in MND.

The intervention was developed using the person-based approach. A qualitative interview study was conducted to explore the emotions experienced by people with MND and their caregivers and to identify what impacts emotional distress and wellbeing. Findings presented the different triggers of emotional distress and suggested that any intervention needs to focus on hope, control, and compassion as these factors are important for wellbeing. A systematic review and qualitative meta-synthesis highlighted the experiences, benefits, and challenges with psychological interventions for neurodegenerative diseases. The review concluded that people with neurodegenerative diseases and caregivers experience wide-ranging benefits from psychological interventions including changes in insight, perspective, self-efficacy, emotions, and relationships. However, burden-reducing adaptations and sensitive tailoring to the specific disease context are required to improve intervention acceptability and engagement. Findings from these two studies were used to develop a digital mental health intervention for MND titled ‘Coping And Living well with MND’ (CALM). Two studies were conducted to explore experiences of using the intervention. Findings suggested that people with MND and caregivers found the intervention useful for developing a positive outlook and regaining a sense of control. The importance of intervention accessibility and flexibility for developing acceptable and engaging interventions for MND was emphasized.

Keywords: Motor Neurone Disease, psychological intervention, digital intervention, qualitative, person-based approach

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Research Thesis: Declaration of Authorship

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I declare that this thesis and the work presented in it are my own and has been generated by me as the result of my own original research.

I confirm that:

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Definitions and Abbreviations

ACT	Acceptance and commitment therapy
ALS	Amyotrophic lateral sclerosis
ALSAQ	Amyotrophic lateral sclerosis assessment questionnaire
ALSFRS	Amyotrophic lateral sclerosis functional rating scale
CALM	Coping and living well with MND
CASP	Critical appraisal skills program
CBT	Cognitive behavioural therapy
CERqual	Confidence in the Evidence from Reviews of Qualitative research
COREQ	Consolidated criteria for reporting qualitative research
CSM	Common sense model
DMHI	Digital mental health intervention
ECAS	Edinburgh cognitive and behaviour ALS screen
ENTREQ	Enhanced transparency in reporting the synthesis of qualitative research
MND	Motor neurone disease
MNDA	Motor neurone disease association
MOSCOW	Must have, Should have, Could have, Would like, system for prioritising changes in the table of changes
MS	Multiple sclerosis
NHS	National health service
PBA	Person-based approach
PBP	Progressive bulbar palsy
PD	Parkinson's disease
PLS	Primary lateral sclerosis
PMA	Progressive muscular atrophy
PPI	Patient and public involvement
PRISMA	Preferred Reporting Items for Systematic Reviews and Meta-analyses

pwMND	people with Motor neurone disease
RCT	Randomised controlled trial
ZBI	Zarit burden interview

Chapter 1 An introduction to Motor Neurone Disease, psychological interventions, and methods used in this thesis

1.1 Background: Motor Neurone Disease (MND)

1.1.1 Clinical picture

Motor neurone disease (MND) is a neurological disease that involves the progressive degeneration of the motor neurones that leads to progressive muscle weakness (Leigh & Ray-Chaudhuri, 1994). In the United Kingdom (UK), the term MND is used to include the complete spectrum of the disease including amyotrophic lateral sclerosis (ALS), progressive bulbar palsy (PBP), primary lateral sclerosis (PLS), and progressive muscular atrophy (PMA) (Leigh & Ray-Chaudhuri, 1994; Thompson & Swash, 2001). The cause of MND is still unknown, some risk factors have been proposed but not confirmed (Moore, McDermott, & Shaw, 2008; Thompson & Swash, 2001). There is no specific test for diagnosing someone with MND and people often go through a lengthy process to obtain diagnosis, from the time they notice the first symptoms to getting a diagnosis from a neurologist (Househam & Swash, 2000; O'Brien, Whitehead, Jack, & Mitchell, 2011). There is no known cure for MND. The drug Riluzole is the only disease-specific treatment available, and treatment largely focuses on symptom management and improving quality of life (Miller, Mitchell, & Moore, 2012; Thompson & Swash, 2001).

1.1.2 Epidemiology

MND is considered a relatively rare disease because of its low prevalence. In 2016, 330, 918 individuals had MND globally and the worldwide all-age prevalence was 4.5 per 1000, 000 people (Logroscino et al., 2018). In the UK in 2016, 13, 249 individuals had MND (Logroscino et al., 2018). MND has a prevalence rate of 7 per 100,000 (Talbot, 2009). There are approximately 5000 cases of MND in the UK at any one time (Moore et al., 2008).

The average age of onset is 65 years; however, younger (10% under 45 years) and older (20% over 70 years) onsets are also observed (Talbot, 2009). MND affects slightly more males than females, ratio 1.5:1 (Goldstein & Leigh, 1999). It usually occurs sporadically, although there is a familial vulnerability in 5-10% of cases (Leigh & Ray-Chaudhuri, 1994). Although the overall prevalence of MND is low, it is a fatal disease which causes severe disability.

1.1.3 Physical and cognitive symptoms

The progressive degeneration of motor neurones with MND can lead to muscle weakness in several areas. People experience difficulties with movement in the limbs, difficulties communicating, difficulties with swallowing and eating, breathlessness, as well as other sensory symptoms, cognitive impairment and emotional lability (Moore et al., 2008). The onset of muscle weakness can affect limb, bulbar, and respiratory muscle (Hobson, Harwood, McDermott, & Shaw, 2016). Limb onset is the most common; bulbar onset occurs in about 20% of those affected (McDermott & Shaw, 2008; Thompson & Swash, 2001). Bulbar symptoms are often associated with emotional lability (i.e. inappropriate and uncontrollable laughing or crying, or exaggerated changes in mood) (McDermott & Shaw, 2008). The severity and distribution of these symptoms differs for people. Although disease progression is unpredictable, symptoms typically increase in severity and distribution (Moore et al., 2008).

Cognitive and behavioural changes occur in some cases, specifically alterations in language, social cognition and executive functioning difficulties (Bora, 2017; Goldstein & Abrahams, 2013).Around 35% of people with MND experience mild cognitive impairment and up to 15% develop frontotemporal dementia (Goldstein & Abrahams, 2013; Strong et al., 2017). Cognitive impairment is associated with a more rapid disease progression and poorer prognosis (Benbrika, Desgranges, Eustache, & Viader, 2019; Garcia-Willingham, Roach, Kasarskis, & Segerstrom, 2018; Gordon et al., 2011).

1.1.4 Prognosis, treatment, and care

Prognosis can range from a few months to over 10 years, typically people die within 2-3 years from diagnosis (Moore et al., 2008). Indicators of poorer prognosis are older age, and bulbar or respiratory onset (Moore et al., 2008). However, longevity is determined by many factors, and it is difficult to predict the prognosis for individual people (Talbot, 2009).

Effective disease-modifying treatments are limited. Riluzole is the only drug available that can increase survival time by 2-3 months (Miller et al., 2012). However, interventions that focus on symptom management are available (Moore et al., 2008). Treatment and care are centred around optimising quality of life for patients and their families. A multidisciplinary approach to care is recommended with professionals such as neurologists, physiotherapists, occupational therapists, specialist nurses, social workers, dietitians, speech and language therapists, respiratory nurses, psychologists, and palliative care specialists (McDermott & Shaw, 2008). Providing holistic care has led to significant improvements in the management of physical, social, and psychological symptoms (Moore et al., 2008).

1.2 Impact on emotions and wellbeing

1.2.1 Impact for people with MND

1.2.1.1 Emotions, distress, and difficulties with adjustment

With few effective therapies, limited prognosis, and declining physical function, MND can be a distressing condition. People with MND often experience anxiety and depression, although there is still some uncertainty about the true prevalence rates for depression and anxiety based on differences in the assessment methods (Pagnini, 2013). Identifying and reporting anxiety and depression may also prove difficult because some of the physical symptoms may be conflated with MND symptoms (Averill, Kasarskis, & Segerstrom, 2007; Felgoise et al., 2009). Some studies suggest that prevalence rates can be as high as 44% for depression and 30% for anxiety (Ferentinos et al., 2011; Kurt, Nijboer, Matuz, & Kübler, 2007; Taylor, Wicks, Leigh, & Goldstein, 2010). There has been more research on depression compared to anxiety in MND (Pagnini, 2013). Some research shows that anxiety may be prevalent particularly around the time of diagnosis (O'Brien et al., 2011; Pavey, Allen-Collinson, & Pavey, 2013) and during the final stages of MND (Whitehead, O’Brien, Jack, & Mitchell, 2012).

In addition to anxiety and depression, people with MND often experience other forms of psychological or emotional distress. The non-curative nature of the disease and limited prognosis can lead to feelings of hopelessness and demoralization, which are qualitatively different from depression (Clarke, McLeod, Smith, Trauer, & Kissane, 2005; Pagnini, 2013; Plahuta et al., 2002). Feelings such as anger and frustration are experienced because of the loss of function and ability, and because of increased reliance/dependence on others for help (Brott, Hocking, & Paddy, 2007; Brown, 2003; Cox, 1992). People may also experience feelings of sadness because of what has been lost and feelings of hopelessness for the future (Foley, Timonen, & Hardiman, 2014; Hugel, Grundy, Rigby, & Young, 2006; Hughes, Sinha, Higginson, Down, & Leigh, 2005; Pavey et al., 2013). Locock and colleagues used the term ‘biological abruption’ to describe the commonly reported feeling among people with MND and caregivers of a sudden ending of an anticipated future (Locock, Mazanderani, & Powell, 2012). People may also experience low self-esteem (Goldstein, Atkins, Landau, Brown, & Leigh, 2006; Hogg, Goldstein, & Leigh, 1994) and struggle to deal with changes to their identity, social roles, and relationships (Brott et al., 2007; Foley, O'Mahony, & Hardiman, 2007; Goldstein et al., 2006; Sakellariou, Boniface, & Brown, 2013).

Emotional distress is particularly experienced around the time of diagnosis. People with MND often describe the impact of the diagnosis as a ‘shock’, ‘death sentence’ or ‘a bombshell’, and the sense of devastation at how their lives and future had suddenly changed (Brown, 2003;

Locock et al., 2012; Mistry & Simpson, 2013). Some people with MND have described experiencing frustration, worry, and anger at how the diagnosis was communicated or due to the delay in the diagnostic process (O'Brien et al., 2011; Pavey et al., 2013). Some feel a sense of relief at knowing the diagnosis (Hugel et al., 2006; O'Brien et al., 2011), whereas others may go through a process of denial or question the diagnosis and why it happened to them (Mistry & Simpson, 2013; O'Brien et al., 2011).

As physical function declines, people with MND have to make changes to their work, hobbies, social activities, or use equipment or assistance from others to enable them to continue doing these activities. Sometimes people accept these changes and make adjustments, and sometimes people struggle with dealing with how their lives and identity have changed (Brott et al., 2007; Hugel et al., 2006; Hughes et al., 2005; King, Duke, & O'Connor, 2009; Mistry & Simpson, 2013). For some people, these losses can threaten their sense of connection and meaning, and some find they engage less with life (Brott et al., 2007; Locock et al., 2012; Mistry & Simpson, 2013). Others struggle with being dependent on others for even simple self-care activities such as bathing or eating (Brott et al., 2007; Brown, 2003; Hugel et al., 2006). Towards the end stages of the disease, people can experience anxiety about the future and dying, about losing more control over their bodily functions, and some express a wish for euthanasia (Whitehead et al., 2012).

1.2.1.2 Coping and Adjustment

Psychological adjustment is possible for people with MND despite physical impairment and deterioration (Bremer, Simone, Walsh, Simmons, & Felgoise, 2004; Lulé et al., 2012; McLeod & Clarke, 2007). People with MND make use of a number of coping strategies, including both problem-focused and emotion focused coping strategies (Larsson, Nordin, & Nygren, 2016; Montel, Albertini, Desnuelle, & Spitz, 2012). Active coping strategies and maintaining independence were commonly used by people particularly at the early stages of the disease when physical disability is less (Jakobsson Larsson, Nordin, Askmark, & Nygren, 2014; Montel et al., 2012). Strategies like avoidance or denial may be helpful in the short term for coping with particular stressors, but generally unhelpful as people need to find ways to adapt to ongoing symptom progression (McLeod & Clarke, 2007). Searching for information was important at different points in the disease (Hecht et al., 2002). People often coped with unrelenting losses by exerting control over how they engage with healthcare services (Foley et al., 2014). People with MND constantly make decisions about the extent to which they have control or the disease has control over them and this influences their choice of either more active or passive coping strategies respectively (King et al., 2009).

People also describe using strategies to change their thinking and be more optimistic. Strategies like having a positive attitude and reframing, thought control, reminiscing and living each day at a time were often used (McLeod & Clarke, 2007; Montel et al., 2012; Young & McNicoll, 1998). One study showed that hope was manifested through hoping for a cure, social support, searching for information, having spiritual beliefs, limiting the impact of MND, adapting to ongoing changes, living in the moment and self-transcendence (Fanos, Gelinas, Foster, Postone, & Miller, 2008). In the face of adversity, people also described the importance of having an appreciation of life, fighting the disease and maintaining their identity (Foley et al., 2007). Having faith and spiritual beliefs are also important for coping with MND (Fanos et al., 2008; Foley et al., 2007). Family and social support were also consistently reported as important for coping (Foley, Timonen, & Hardiman, 2014; Hecht et al., 2002; Jakobsson Larsson et al., 2014; Lulé et al., 2012). The literature sheds some light on the different coping strategies people use. We also need more of an understanding of what influences the use of these strategies and how they impact wellbeing.

1.2.1.3 The importance of psychosocial factors for wellbeing

Wellbeing and quality of life appear to be linked to a number of factors. The evidence for the relationship between physical functioning and wellbeing and quality of life is mixed. Some studies show that physical functioning may be an important variable that influences quality of life, particularly symptoms like fatigue and breathlessness (Gibbons et al., 2013; Young et al., 2019) and disease progression rate or pattern (Cipolletta, Gammino, & Palmieri, 2017; Prell et al., 2019). However, other studies have shown that there is no relationship between physical impairment and quality of life (Chiò et al., 2004; Goldstein, Atkins, & Leigh, 2002; Matuz, Birbaumer, Hautzinger, & Kübler, 2010; Tramonti, Bongioanni, Di Bernardo, Davitti, & Rossi, 2012).

On the other hand, psychosocial factors have been consistently reported to be important for wellbeing. Psychological symptoms like anxiety and depression have been found to be important variables influencing individual’s quality of life (Gibbons et al., 2013; van Groenestijn, Kruitwagen-van Reenen, Visser-Meily, van den Berg, & Schröder, 2016; Young et al., 2019). Coping strategies have also been found to influence quality of life. Coping strategies such as independence, autonomy through using assistive devices, and positive thinking were linked to emotional wellbeing (Jakobsson Larsson et al., 2014; Larsson et al., 2016; Matuz, Birbaumer, Hautzinger, & Kübler, 2015; Tramonti, Bongioanni, Fanciullacci, & Rossi, 2012). Rumination and trait mindfulness has also been found to be linked to emotional wellbeing (Hecht et al., 2002; Pagnini, Phillips, Bosma, Reece, & Langer, 2015). The appraisal of coping potential has been identified as important for wellbeing (Matuz et al., 2010, 2015). Social support and the perceived quality of support has been found to be linked to individual’s quality of life and this has found to be a consistent predictor over time from diagnosis to disease progression and across different studies (Chiò et al., 2004; Fisher, Dodd, Barrow, Makin, & Cherry, 2019; Goldstein et al., 2006;

Goldstein et al., 2002; Jakobsson Larsson, Ozanne, Nordin, & Nygren, 2017; Matuz et al., 2015; Tramonti, Bongioanni, Di Bernardo, et al., 2012). Some active coping strategies such as planning, acceptance, and positive reframing were even found to be linked to longevity (Montel et al., 2012).

Psychological factors are clearly important for quality of life and wellbeing. However, a large proportion of this research is correlational, which means that it is not clear if wellbeing outcomes influence the coping strategies or vice versa (Jakobsson Larsson et al., 2014; Larsson et al., 2016; Montel et al., 2012). Further research is needed to understand more about psychosocial factors and how they influence wellbeing in MND, so that they can be modified and used to develop interventions (van Groenestijn et al., 2016).

1.2.2 Impact for caregivers

1.2.2.1 Emotions, distress, and burden

Family members or caregivers of people with MND also often struggle with the emotional impact and burden of the disease and have high rates of psychological morbidity (Goldstein & Leigh, 1999; Pagnini, 2013; Rabkin, Albert, Rowland, & Mitsumoto, 2009). The high burden and strain experienced by MND caregivers is largely due to the rapid and progressive nature of MND (Aoun et al., 2013). Caring for a person with MND can be very demanding with people requiring assistance with many activities of daily living from eating to transport and medical care, and the time spent caregiving increases as the disease progresses (de Wit et al., 2018; Galvin et al., 2016). MND caregivers are also confronted with difficulties accepting the disease, increased levels of dependency, and the shifting nature of their relationship with the person they are caring for (Galvin et al., 2016). Depression, anxiety, and psychological distress have been most commonly reported among MND caregivers (Aoun et al., 2013; Lillo, Mioshi, & Hodges, 2012). Depression appears to be connected with the experience of multiple losses for the person with MND and an increase in caregiving intensity over time (Aoun et al., 2013). Fewer studies have examined anxiety in caregivers. Caregiver anxiety may be associated with uncertainty about the disease trajectory and experiencing a loss of control (Aoun et al., 2013; Holkham & Soundy, 2018). Psychological distress may also occur from watching the patient’s decline and from changes in relationship with the patient (Conroy et al., 2021; Galvin et al., 2016).

Caregivers also experience feelings of anger, worry, stress, fear, frustration associated with providing care for the person with MND and with the uncertainty and inability to plan for the future (Galvin et al., 2016). Particularly around the time of diagnosis and the period prior to getting a diagnosis, caregivers report feeling fear, worry, anger and frustration at the process of obtaining a diagnosis (Galvin et al., 2018). Caregivers also report experiencing guilt in taking time out to look after themselves (Weisser, Bristowe, & Jackson, 2015) and also feeling guilt post-bereavement in relation to the care they provided for their family member (Whitehead et al., 2012). Anxiety, distress and burden can be heightened for caregivers during the final stages of the disease (Whitehead et al., 2012). Caregivers expressed worries about how they would cope as the disease advanced and also how they would cope with being bereaved (Whitehead et al., 2012). Distress and burden seem to increase over time (De Wit, Bakker, et al., 2019; Galvin, Gavin, Mays, Heverin, & Hardiman, 2020; Goldstein, Atkins, Landau, Brown, & Leigh, 2006). Caregivers may also struggle with complex emotions and distress following bereavement (Whitehead et al., 2012).

MND caregivers can also experience burden or strain (Aoun et al., 2013; Lillo et al., 2012). Caregivers report not having enough personal time and support to be able to do activities to look after their own wellbeing (De Wit, Bakker, et al., 2019). They also have the strain of tasks such as learning to care for the person with MND, making changes to their own lives and not having time for themselves (Brown, 2003; Cox, 1992; Ozanne O, Anneli G, Strang, & Persson, 2011). The practicalities of providing care can be restrictive and relentless, and can affect family dynamics and caregivers’ self-identity (Galvin et al., 2016). Caregivers also describe challenges dealing with agencies and services, managing competing family and personal responsibilities and worries about the future (Weisser et al., 2015). Caring for someone with MND can also limit access to social support which can result in poorer outcomes for caregivers (Gluyas, Mathers, Anderson, & Ugalde, 2017). Despite experiencing burden and needing support, some caregivers see their needs as secondary to that of the person with MND (De Wit, Bakker, et al., 2019).

1.2.2.2 **Coping and wellbeing**

There have been fewer studies that have described how caregivers cope with and adjust to caring for someone with MND. In a study by Weisser and colleagues (2015), caregivers describe how social support, actively seeking information and being proactive are beneficial when coping with MND (Weisser et al., 2015). Focusing on simple pleasures, living in the moment, and seeing things from different perspectives also helped caregivers cope with the daily challenges. Another study by Caga et al (2021) found that caregivers mostly used problem-focused coping strategies such as active coping and planning (Caga et al., 2021). Family, social activities and hobbies were also described as important and contributed to quality of life (Galvin et al., 2020). Not all caregivers expressed difficulties with coping and some reported having adequate support (Galvin et al., 2018). Caregivers can also experience rewards or benefits from caregiving (Kennedy et al., 2022; Weisser et al., 2015). Some of the positive aspects of caregiving include strengthening relationships and increasing trust in themselves (Weisser et al., 2015).

Coping and adaptation is a dynamic process and focusing on particular coping strategies may be too simplistic (De Wit, Bakker, et al., 2019). Caregivers may require different strategies for different situations or for stressors at different stages of the disease (Caga et al., 2021).

1.2.2.3 Factors that influence wellbeing and quality of life

Factors related to the person with MND can have an impact on wellbeing outcomes for caregivers. Poorer psychosocial outcomes for caregivers are associated with looking after patients with a more severe clinical profile that require more care, and caregiver burden increases with an increase in patient's loss of function (Chiò et al., 2004; Creemers et al., 2016; Gauthier et al., 2007). However, there are some contradictions in the literature. Aoun and colleagues (2013) found that caregiver quality of life appears to be unrelated to the severity of MND, patient's quality of life, or level of care (Aoun et al., 2013). Another study showed that caregiver burden was linked to behavioural issues in patients such as disinhibition or impulsivity and less by the level of disability of the patient (Lillo et al., 2012). This was contradictory in part by a study by de Wit and colleagues (2018) where they demonstrated that caregiver burden was associated with greater behavioural and physical impairment of the patient (de Wit et al., 2018). Some research also demonstrates that there is a relationship between psychosocial outcomes in the person with MND and caregiver distress and strain (Creemers et al., 2016; Goldstein et al., 2006). It was also difficult for caregivers if patients refused to accept or were resistant to formal care services (Galvin et al., 2018). Despite some contradictory findings, there is sufficient evidence that points to a link between patient and caregiver experience (Gluyas et al., 2017).

There is some research that suggests that psychosocial factors are important and influence wellbeing and quality of life in MND caregivers. Psychological distress and burden were found to be linked to quality of life in MND caregivers (Galvin et al., 2016; Galvin et al., 2020). There also appears to be a link between coping strategies and wellbeing outcomes. Adaptive problem-solving coping skills and spiritual beliefs/faith have a positive influence on psychosocial outcomes (Gluyas et al., 2017; Weisser et al., 2015). Dysfunctional coping strategies such as self-blame, denial, self-preoccupation and passivity were found to be associated with increased caregiver distress especially during the early stages of the disease (Caga et al., 2021). Caregiver strain was also associated with a passive coping style and feeling less supported by the care team (Creemers et al., 2016). In terms of social factors, lower levels of social support, dissatisfaction with social relationships, and having more dependents predicted caregiver distress (Goldstein et al., 2006). Similar to the body of research with people with MND, studies are largely correlational and our knowledge of what influences wellbeing in caregivers remains limited to factors that have been pre-identified and measured through these studies.

1.2.3 Defining emotional distress and wellbeing

The literature around the emotional impact of MND suggests that distress for people with MND and caregivers is not limited to anxiety and depression, but can include a broad range of emotions, burden, and difficulties with adjustment. In this thesis, the term 'emotional distress' refers to these broad range of emotional concerns. Similarly, the term 'emotional wellbeing' covers improvements in psychosocial outcomes (e.g. a reduction in distress, satisfaction with social support) and in quality of life. The term 'psychological distress or wellbeing' was not used because it could imply cognitive and behavioural issues in MND which were not covered in this thesis. The focus of this thesis and the intervention developed through this project is on emotions and managing emotions in MND.

1.3 Psychological interventions for MND

1.3.1 Need for psychological interventions

Despite the significant impact on wellbeing for both people with MND and caregivers, there is very little evidence on psychological interventions and more intervention research is urgently needed. A review by Gould and colleagues (2015) found that the available evidence was insufficient to recommend a specific therapeutic approach for people with MND (Gould et al., 2015). Some interventions had promising results but there was a lack of evidence for long-term effectiveness. A few more intervention studies have been added to the evidence base since then, however a more recent review still found that the evidence for psychological interventions was limited (Zarotti, Mayberry, Ovaska-Stafford, Eccles, & Simpson, 2021). Similarly, research on psychological interventions for caregivers is limited. Many psychological interventions include both people with MND and caregivers as participants, and there are very few interventions specifically for caregivers' wellbeing. There are also questions about the methodological quality of existing evidence for caregiver interventions (Cafarella, Effing, & Chur-Hansen, 2022; Harris et al., 2018).

1.3.2 Current evidence on different psychological interventions

The psychological interventions for MND researched so far include expressive disclosure, hypnosis, dignity therapy, cognitive behavioural therapy (CBT), and mindfulness. Some of the studies had promising results, however there was often a lack of post-intervention follow up data to draw conclusions of long-term intervention effectiveness.

Averill et al (2013) conducted a randomised controlled trail (RCT) on self-help expressive disclosure where participants were asked to write or talk about their thoughts and feelings related to MND over a week. They found that those participants in the disclosure condition reported significantly higher psychological wellbeing than the non-disclosure condition, but this effect was not maintained at 6 months post-intervention. Additionally, ambivalence over emotional expression moderated the effect on wellbeing, indicating that this kind of intervention may be helpful to those who have difficulty expressing their emotions (Averill, Kasarskis, & Segerstrom, 2013). Two studies evaluated hypnosis-based interventions (Kleinbub et al., 2015; Palmieri et al., 2012). Palmeiri and colleagues found that in a small sample of 8 people with MND, there were significant improvements in anxiety and depression post-intervention, but there was no follow up data to provide evidence of the long-term effects. Kleinbub et al (2015) tested the same intervention with both people with MND and caregivers and found that the intervention significantly improved psychological outcomes post-intervention. Effects on patients' and caregivers' anxiety and caregivers' depression were also found at 3 and 6 month follow ups.

Two studies evaluated dignity therapy, which involves going through a life review and developing a document to reflect this in collaboration with the therapist (Aoun, Chochinov, & Kristjanson, 2015). The dignity therapy intervention was well-received by both people with MND and caregivers, including by people who used assistive devices to communicate. However, there were no significant improvements in psychosocial outcomes post-intervention. CBT approaches and techniques have also been used to develop interventions for MND. Diaz and colleagues (2014) combined CBT with some counselling techniques in a four-session intervention and found that there were significant improvements in anxiety and depression post-intervention (Díaz et al., 2016). No follow up assessments were conducted so longer-term effects are unknown. Van Groenestijn and colleagues conducted an RCT to test the effects of a CBT intervention that was designed for MND-specific problems and for both people with MND and caregivers (Van Groenestijn et al., 2015; van Groenestijn et al., 2011). The trial had to stop prematurely because of problems with recruitment. However, preliminary data showed that psychological outcomes were better in the intervention group compared to the control group (Van Groenestijn et al., 2015). Recruitment was a major problem in the trial and the authors concluded that their eligibility criteria was overly restrictive. Additionally, uptake was low and drop-out was high as a result of participants perceiving the intervention (up to 10 face-to-face sessions) as burdensome. Therefore, CBT might be beneficial to people with MND and caregivers, however there may be issues with engaging with time-intensive face-to-face therapy.

There is also encouraging evidence for the effectiveness of mindfulness-based interventions with MND. Mindfulness involves bringing one's attention to the present moment and accepting feelings and sensations without judgement (Kabat-Zinn, 1990). One way of doing this is through the practice of meditation that encourages an awareness of what is happening in the body at the present moment. This may be helpful for people with MND because it can promote greater acceptance of feelings and physical limitations, and can help people focus on and make best use of the resources and abilities that are currently available (Pagnini et al., 2017). Pagnini and colleagues evaluated an MND-specific mindfulness-based stress reduction intervention for people with MND and caregivers (Pagnini et al., 2017). They found significant improvements in anxiety, depression, and quality of life immediately post-intervention, as well as 3 and 6 months later. A qualitative study examining the experiences of participants confirmed the positive impact as participants reported an increase in acceptance and non-judgemental attitude (Marconi et al., 2016). However, barriers to participation in the intervention were also identified including things like difficulties with transportation to the sessions, worrying about adding to caregiver burden, and completing homework tasks. More recently, Thomas and colleagues (2022) developed and tested a psychosocial intervention for people with MND and family members. This was a supportive and psychoeducational intervention that involved assessing and teaching coping skills, providing information about the disease and management, providing emotional support, and providing or strengthening support links with the multidisciplinary care team and community organisations. Participants found this kind of intervention beneficial, and authors concluded that it was feasible. More research is still needed to determine efficacy (Thomas et al., 2022).

With psychological interventions for caregivers, the results have not always shown significant effects in improving psychological wellbeing post-intervention. In 2018, Ugalde and colleagues developed a group intervention for MND caregivers that aimed to promote self-care, problem-solving, and mindfulness. Overall, participants found the intervention acceptable and highly valued the group format. However, there were no significant changes in psychological outcomes post-intervention (Ugalde et al., 2018). The group support format seems to be important and other research has also showed that caregivers valued peer support, as it helped overcome isolation and helped them care for their relatives with MND better (Cipolletta, Gammino, Francescon, & Palmieri, 2018). More recently, an RCT of a blended support program (face-to-face and online support) based on acceptance and commitment therapy was conducted in the Netherlands. The authors found that the intervention had no significant effect on primary or secondary outcome measures. However, it had a significant effect on the intervening variable self-efficacy (De Wit et al., 2020). A qualitative study on experiences of this same intervention found that participants found the intervention beneficial, as it helped caregivers feel in control over the caregiving situation, accept negative thoughts and emotions, and feel acknowledged (de Wit, Vervoort, et al., 2019). These benefits were not seen in the quantitative outcome measures for the RCT. Participants also reported that lack of time was a barrier to completing the intervention (De Wit et al., 2020).

Overall, the research evidence on psychological interventions for MND show some promising results for certain therapeutic approaches. Although there is not sufficient evidence for recommending a particular therapy or approach, it may be that multiple therapeutic approaches or models can be used to improve wellbeing (Zarotti et al., 2021). Moreover, there might be some issues with engagement and adherence to psychological interventions that need to be considered when developing interventions for this group.

1.3.3 Relevant theories, promising interventions, and therapeutic approaches

As there is limited consensus and evidence on specific psychological interventions and therapeutic approaches that would be effective for improving psychological outcomes in MND, this section presents the wider literature including relevant theories and psychological interventions that have been used with other neurodegenerative and life-limiting illnesses.

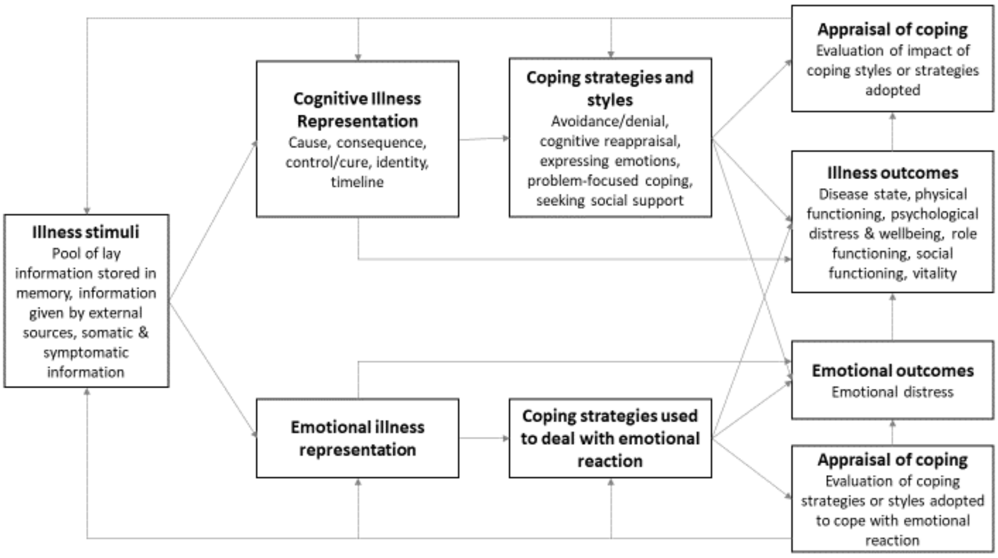
1.3.3.1 The common sense model (CSM)

The CSM describes the process by which an individual constructs representations or beliefs about their illness, which then influences the selection and use of coping strategies in response to a perceived health threat, and this in turn contributes to health outcomes (e.g. psychological distress or adjustment) (Leventhal, Meyer, & Nerenz, 1980). Individuals' representations of the illness are dynamic and developed through interactions with their social and cultural context, previously held beliefs, and through direct and vicarious experiences of their illness and treatment (Hagger & Orbell, 2003; Leventhal et al., 1980). Illness representations are constructed around five main dimensions – identity (the label for the illness and associated symptoms), cause (beliefs about the cause of the illness), consequence (the social, psychological, physical consequences of the illness), timeline (how long the disease or symptoms will last), and cure/control (beliefs about the extent to which the illness can be cured or controlled).

There has been some research on illness representations in neurological illnesses that has shown they are relevant and can influence psychological outcomes. For example, in Huntington's disease, Parkinson's disease, and multiple sclerosis, illness representations or perceptions were associated with variance in psychological distress and wellbeing outcomes (Arran, Craufurd, & Simpson, 2014; Evans & Norman, 2009; Jopson & Moss-Morris, 2003; Kaptein et al., 2006; Simpson, Lekwuwa, & Crawford, 2013). However, this research is largely correlational and causal relations between illness perceptions and psychological outcomes cannot be presumed. Relationships between illness perceptions and psychological outcomes are more likely to be dynamic than linear (Simpson et al., 2013) and may not be mediated by coping strategies (Kaptein et al., 2006). Clinical variables or disease stage can also influence illness perceptions (Miglioretti, Mazzini, Oggioni, Testa, & Monaco, 2008). Therefore, several factors play a role in explaining distress and wellbeing, and illness representations alone only partially explain wellbeing in neurological diseases. Psychological interventions can target and change illness perceptions but other factors that contribute to wellbeing may also need to be considered.

Figure 1

The common sense model of illness representations. Adapted from Hagger & Orbell (2003)



1.3.3.2 Stress and coping framework for serious illness

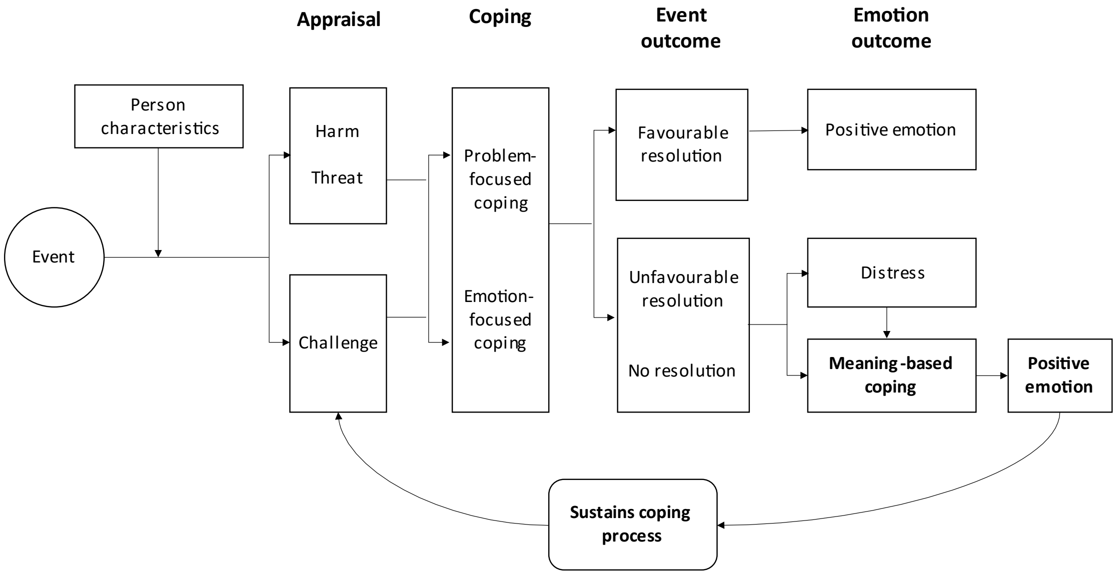
Lazarus and Folkman's theory of stress and coping is a useful framework to understand and explain stress and coping in chronic illness (Lazarus & Folkman, 1984). This model was adapted in 2000 to incorporate aspects that were relevant to coping with serious and progressive illnesses (Folkman & Greer, 2000). Based on this model, wellbeing depends on two processes – appraisal and coping. Appraisal involves evaluating the significance of an event and the individual's coping resources. There are different types of coping – problem-focused coping (actions to manage the problem causing distress), emotion-focused coping (managing thoughts and behaviours to regulate distress), and meaning-based coping (making sense of and seeing benefit in negative circumstances). Coping can influence the outcome of the event and the individual's appraisal of the success of coping strategies used, which can then influence subsequent coping (see Figure 2). Typically, greater control is associated with more problem-focused coping, whereas less control is associated with higher levels of emotion-focused coping. In serious illness, there is often no control over symptoms or disease progression and variable amounts of control over how the disease can be managed. Meaning-based coping strategies are useful as they help the person relinquish control over unattainable goals, and formulate new ones which generates positive affect and can motivate further coping (Folkman & Greer, 2000).

According to this theoretical model, there are three categories of variables that contribute to wellbeing in serious illnesses. These are dispositional variables related to optimism and personal control, situational beliefs about efficacy and control, and coping processes that create positive meaning. The latter two categories are amenable to change through interventions and form the basis of the following recommendations. Interventions should first seek to create conditions for a challenge, an eagerness or opportunity for meaningful mastery or gain. This involves finding out what is meaningful to the person, helping them establish realistic goals, and emphasizing opportunities for personal control. Interventions should also help encourage behaviour to help achieve these goals, and simultaneously help maintain background positive mood.

Research on the application of this model with advanced cancer showed that it was a useful model to explain stress and coping (Roberts et al., 2018). The authors found that events that were appraised were not just the diagnosis, but also test results or changes in disease stage and symptoms. Consequently, interventions need to have the flexibility to continually re-appraise and adjust to new events and people need to be supported to re-evaluate their goals and re-attain positive emotion as things fluctuate and change with the disease.

Figure 2

Theoretical model of appraisal and coping process in serious illness. Adapted from Folkman and Greer (2000)

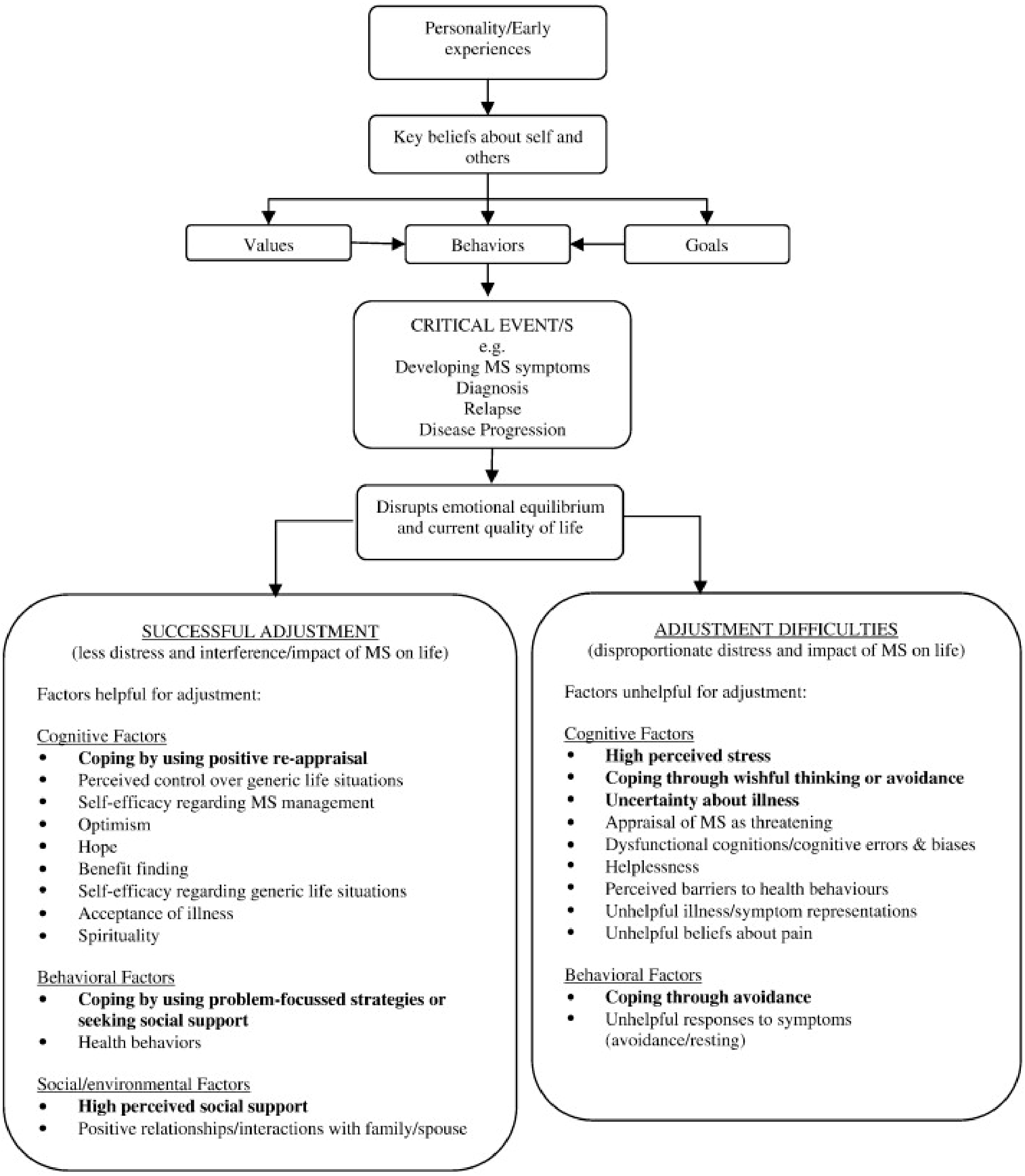


1.3.3.3 Disease-specific models of adjustment and wellbeing

There have been some attempts to develop models that explain adjustment and wellbeing in neurodegenerative diseases that take into account illness-specific factors. Dennison and colleagues developed a working model of adjustment for multiple sclerosis (MS) (Dennison, Moss-Morris, & Chalder, 2009; Moss-Morris, 2013). According to this model, critical events (e.g. getting a diagnosis or experiencing a change in symptoms) challenge existing values, goals and behaviours and disrupt the individual's equilibrium. Equilibrium includes psychological, social, and physical adjustment and involves factors such as good illness management, positive affect, and reduced illness interference on roles and relationships (Moss-Morris, 2013). Background factors such as an individual's personality or early beliefs can influence adjustment outcomes. Additionally, illness-specific factors (i.e. cognitive, behavioural, social factors) can be linked to either successful adjustment or difficulties with adjustment (Dennison et al., 2009). The utility of this model is that adjustment is seen as a dynamic process and factors that may be successful at one stage may not be useful as the disease gets progressively worse. This also has relevance for MND. Moreover, the model draws attention to both individual and disease-specific factors that can have an impact on adjustment outcomes, and the need for healthcare professionals and intervention developers to consider both these sets of factors.

Figure 3

A working model of adjustment to MS. Adapted from Dennison et al (2009)



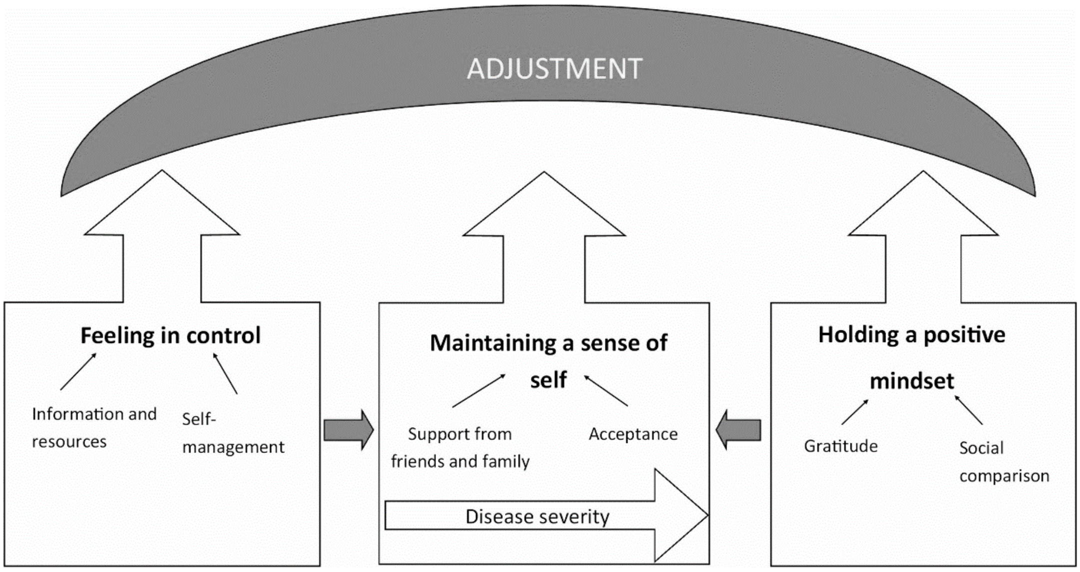
Bold text= Strong evidence. Many studies conducted and literature consistently supports the role of this factor

Normal text= Modest evidence. Less research conducted, or some conflicting evidence from studies about the importance of the factor

Drawing on a similar conceptualization of adjustment, Wieringa and colleagues developed a model of adjustment in Parkinson's disease (PD) through a synthesis of evidence on individuals' experiences of adjusting to living with PD (Wieringa, Dale, & Eccles, 2021). According to this model, three main themes were identified as important for adjustment – maintaining a coherent sense of self, feeling in control, and holding a positive mindset. Individuals' experience having PD as a threat to their self-identity and therefore, maintaining a coherent sense of self is a central component of adjusting to PD. As with the previous model with MS, the goal of adjustment is to maintain equilibrium and reduce the impact of PD on their life. This model goes further to say that equilibrium does not involve restoring to a former sense of self, but also involves altering or constructing a new self-identity that incorporates their experience of having PD. Therefore, what 'equilibrium' is for the individual will constantly change as the illness progresses. This process of re-constructing their identity can help individuals make sense of and find meaning in an event that has disrupted their life and can help them adjust to living with PD. Gaining control was another important factor for adjustment. This involves both control over aspects of the disease through self-management and control over the impact of the disease on their lives. Having a positive mindset was also important for adjustment. For individuals with PD this was achieved through social comparisons and experiencing gratitude. Having a positive mindset was important even in the face of deterioration or disease progression, and could co-exist with experiences of negative affect. These three themes and their relationship with each other are illustrated in Figure 4.

Figure 4

Sense of self adjustment model for Parkinson's disease. Adapted from Wieringa et al (2021)



Two models have been proposed that attempt to explain adaptation to loss and psychological wellbeing in MND (King et al., 2009; Soundy & Condon, 2015). Both models emphasize the importance of having a sense of control and describe successful adjustment as re-establishing periods where the individual is more in control as MND progresses. Soundy and Condon's model proposes two types of responses to loss in MND – a more self-controlled or dominant response where the individual responds actively, or a more disease-dominated response where the individual takes a more passive role (Soundy & Condon, 2015). The individual undertakes a more self-controlled response by using cognitive adaptation strategies (e.g. focusing on the present, focusing on what they can do, using faith or religion, planning activities) or pragmatic adaptation strategies where individuals make the most of the time they have left and do whatever they can to live positively. A more disease-controlled response includes things like denying the effects of the disease or an inability to accept the diagnosis, worry and fear about the uncertain future, resignation to what will happen in the future, being overcome when considering the implications of the disease. According to this model, periods of coping are possible where the person chooses responses that enable hope and control, and this can have a positive effect on wellbeing (see Figure 5).

King and colleagues describe the cognitive process that an individual goes through when adapting to loss and changes in MND (King et al., 2009). This involves perceiving and reacting to the change, appraising the change (either of the individual being in control or MND being in control), and adopting coping strategies based on this appraisal. After the initial shock of diagnosis, people with MND are constantly making decisions about how to live with the disease progression and deteriorating abilities. Unlike other diseases, change is ongoing with MND. Windows of normality may appear but they are short-lived and people are faced with continuous change or decline and the decision-making process is repeated (see Figure 6). Decisions to adapt or not are based on maintaining self-esteem and control (King et al., 2009).

Figure 5

Model of hope enablement for MND. Adapted from Soundy and Condon (2015)

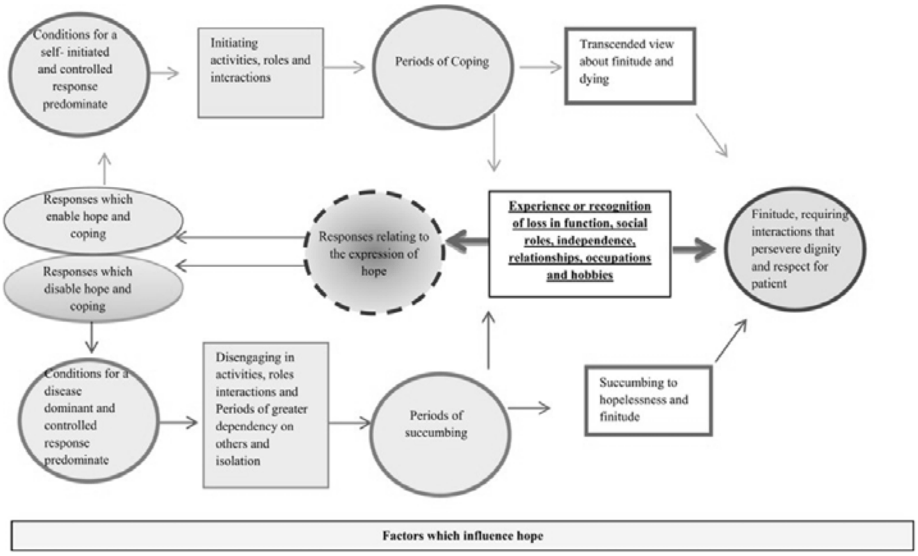
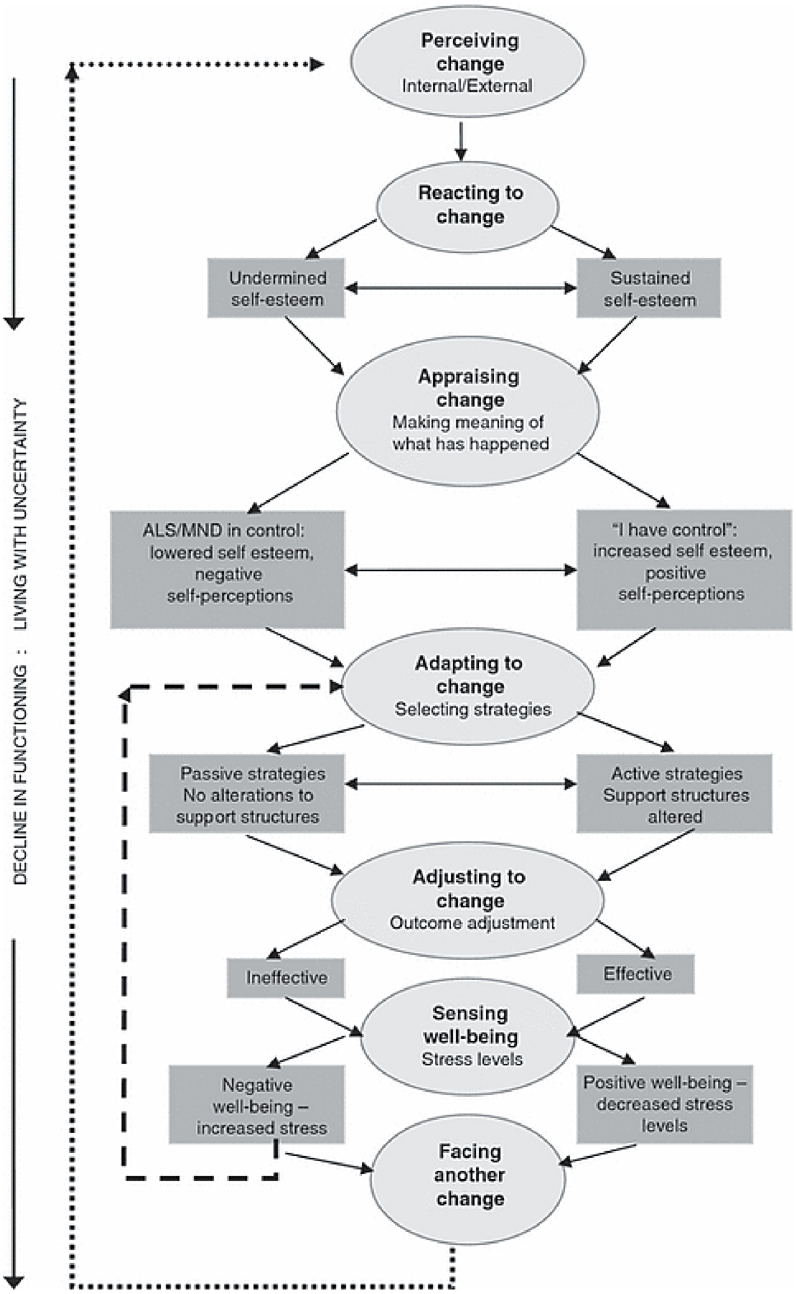


Figure 6

Model of adapting to changes in MND. Adapted from King et al (2009)



A selection of theories and models are presented because they all have some relevance to understanding wellbeing in MND. The theories apply more generally to chronic illnesses and there is currently limited evidence to support the disease-specific models of adjustment and wellbeing. The Folkman and Greer (2000) model was drawn on for intervention development and their suggestions for developing interventions were used. In line with the person-based approach, a combination of theory, previous evidence, and target users' feedback was used to develop and optimise the intervention. These details have been presented in the intervention development section (Pg 52).

1.3.3.4 Evidence for psychological interventions with other diseases and other promising therapeutic approaches

Due to the limited evidence on psychological interventions for MND, it is useful to draw from intervention research with other diseases and similar populations. There is some evidence to suggest that meaning-centred psychotherapy is effective in improving psychosocial outcomes for people with advanced cancer (Breitbart et al., 2015). Meaning-centred therapy is a form of psychotherapy that has its foundations in existential psychology. It uses a mixture of education, discussion and exercises designed to sustain and enhance a sense of meaning, peace, and purpose (Breitbart, Gibson, Poppito, & Berg, 2004). Meaning-centred psychotherapy has also been used with people receiving palliative care. The intervention was abbreviated in terms of shortening the duration of therapy and tailored to the most essential needs first, to make it more suitable for people receiving palliative care (Rosenfeld et al., 2017). There is also some evidence to suggest that positive psychological interventions like mindfulness, positive savouring, life-summary, expressive-based, hope-based, and character strengths interventions improve psychological wellbeing in people with a range of neurological diseases (Lai, Lim, Low, & Tang, 2019). A brief supportive expressive psychotherapeutic intervention called ‘Managing Cancer and Living Meaningfully’ was found to be effective for alleviating depressive symptoms in people with advanced cancer (Rodin et al., 2017). The intervention covered domains such as symptom management and communication with healthcare providers, changes with self and relationships, sense of meaning or purpose, and future-oriented concerns. This has been adapted to the needs of people with MND to include two more domains – communication skills, and emotional expression and control and is in the process of being evaluated (Oberstadt, Esser, Classen, & Mehnert, 2018). Overall, meaning-focused and positive psychology interventions appear to be suitable for people with advanced or progressive disease. There is also a pattern of adapting or keeping interventions brief to ensure that they are appropriate for people with progressive or terminal illnesses.

Acceptance and Commitment Therapy (ACT) has been seen as a promising therapy that is suitable for MND and currently a trial is underway in the UK to evaluate an ACT intervention for MND (COMMEND trial - ISRCTN12655391 https://doi.org/10.1186/ISRCTN12655391). A study that explored patients’ preferences for psychological interventions also concluded that ACT may be an appropriate approach as it helps people focus on meaningful activity and to focus on what can be realistically achieved, which is appropriate for MND where people have to deal with progressive losses (Weeks et al., 2019). The ACT framework highlights the importance of psychological flexibility for wellbeing, and includes pursuing valued behaviours whilst accepting thoughts and feelings without changing or avoiding them (Hayes, Luoma, Bond, Masuda, & Lillis, 2006). There is some evidence that points to the benefits of using ACT in other chronic illnesses (Hayes et al., 2006; Thompson, Moghaddam, Evangelou, Baufeldt, & das Nair, 2022). An intervention for caregivers is also currently being evaluated in Ireland. This is a group intervention that consists of a combination of CBT techniques, mindfulness, and peer support (Burke et al., 2019). Another potential way forward is to adapt and deliver therapy and interventions in a digital format.

1.4 Digital mental health interventions (DMHIs) for MND

Digital mental health interventions are either self-guided or therapist-assisted psychological interventions delivered through smartphone apps, internet websites, virtual reality, wearable devices or video games (Borghouts et al., 2021). The number of DMHIs to support people with chronic conditions has been increasing, and there is evidence that these interventions can have a positive impact on wellbeing outcomes for people living with chronic conditions and also for caregivers (Maisto et al., 2021; Petrovic & Gaggioli, 2020).

1.4.1 Digital interventions for MND: opportunities

DMHIs can be a useful way to provide access to psychological support for MND. Problems with mobility and speech make it difficult for people with MND to access face-to-face psychological support. Some studies have shown that there may be difficulties engaging with intensive face-to-face therapy because of issues with mobility or transportation (Marconi et al., 2016; Van Groenestijn et al., 2015). For caregivers as well, lack to time can act as a barrier to engaging with therapy (de Wit, Vervoort, et al., 2019). The remote and digital delivery of psychological support can enable participation from people with a range of symptoms or levels of ability and facilitate access to therapy at their own pace and around existing schedules.

People with MND and caregivers may not always have access to psychological support, as the provision of psychological support services across the UK are variable. Some people may have to wait long periods of time in order to be able to access psychological support, particularly from professionals who have experience working with people with MND. Informational and self-guided resources can be particularly helpful during this time. Providing this support in a digital format also means these resources can reach people regardless of location and local availability of support.

1.4.2 Digital interventions for MND: challenges

Despite the many benefits of providing psychological support through a digital format, there are also some important considerations when developing this support for MND. Physical and cognitive symptoms may make it difficult for people to either use the particular technology, or the structure and format of the intervention may need to be modified so that people with different symptoms can use the DMHI. Simblett and colleagues (2011) were optimistic about the potential for using digital interventions to managing distress in patients with neurological conditions but highlighted the need to investigate how to accommodate physical and cognitive impairments and address pertinent and meaningful issues for these patients (Simblett et al., 2011). Similarly, research on multiple sclerosis found that generic DMHIs were not suitable for people with multiple sclerosis and that interventions needed to be designed to cater to the specific needs and contexts of people with multiple sclerosis (Hind et al., 2010). It is therefore important to consider these different physical and cognitive symptoms when designing DMHIs for MND, and examine people's experiences of using digital interventions to ensure that they are designed in an appropriate and acceptable way for people with MND and caregivers.

1.4.3 Current evidence on use of digital interventions with MND

There is some evidence to suggest that people with MND do engage positively with technology and that the internet is widely used for information about the disease and treatment (Abdulla et al., 2014). Studies also report that online forums and patient communities are viewed positively, and help foster empathy, connection and social support (Hargreaves, Bath, Duffin, & Ellis, 2018; Loane & D'Alessandro, 2013). Digital legacies have also been used by people with MND to create memories, and share their personality and life experiences with their family members (Clabburn, Knighting, Jack, & O'Brien, 2019).

More recently, an online non-meditative mindfulness intervention has been developed and evaluated for people with MND and caregivers (Pagnini et al., 2022). The study provided promising results on the uptake and effectiveness of an online program to improve wellbeing (Pagnini et al., 2022). Similarly, a dignity therapy intervention for terminal illnesses, including MND, has been adapted to an online format to reduce costs and improve efficiency of intervention delivery. Again, this was a small pilot study, but results showed that feasibility and acceptability was comparable to delivering the therapy in a face-to-face format. Participants also reported that it was convenient to use especially via email, but that technological issues could be challenging (Bentley, O'Connor, Williams, & Breen, 2020). In a blended intervention of both face-to-face and online delivery of therapy, MND caregivers reported that it was easy to use the intervention online and the online format provided flexibility so caregivers could pace themselves and complete modules or exercises at their own convenience (de Wit, Vervoort, et al., 2019). Therefore, digital interventions could offer a promising alternative to providing mental health support for both people with MND and caregivers and could help overcome some of the engagement and adherence difficulties encountered with face-to-face psychological interventions.

1.5 Methodological approach used in the thesis

1.5.1 Ontology, Epistemology, and Methodology

My approach to this thesis was influenced by a critical realist paradigm. In terms of ontology and epistemology, critical realism combines ontological realism with epistemological constructivism (Denzin & Giardina, 2016). Critical realism assumes that objective reality does exist, but that our knowledge of reality is dependent on context and historical situation, and this reality is continually evolving (Bhaskar, 2014). Phenomena exist and operate within open systems, where different contexts and mechanisms affect outcomes (Clark, MacIntyre, & Cruickshank, 2007). For critical realists, both mental and physical entities are equally real, and mechanisms and processes are seen as real phenomena (Maxwell, 2012). Reality is also multi-layered, and there is a distinction between the empirical (what we perceive or experience), actual (events that occur but may be perceived or observed), and real (mechanisms or structures that generate or explain events) events (Cassell, Cunliffe, & Grandy, 2017). Actual events can be caused by real mechanisms that are often invisible to the researcher.

In line with this paradigm, understanding the views of people with MND and caregivers has been central to my thesis, and the methods and approaches I have used have enabled me to view the world from their lens. At the same time, I also believe that an objective reality exists and can be explored and have used methods to identify patterns of experience or identify factors or mechanisms that contribute to people’s experience of distress, wellbeing, and of using psychological interventions.

Qualitative research methodology was mainly used in this thesis to investigate participants’ experiences of emotions and wellbeing, and experiences using psychological interventions. It is a valuable tool for exploring topics that have not been researched and to gain access to the subjective experiences of participants (Camic, Rhodes, & Yardley, 2003). In this thesis, qualitative methods were used to understand participants’ experiences of emotional distress and wellbeing, to synthesize the experiences of using psychological interventions for neurodegenerative diseases, and to understand the experiences of people with MND and caregivers who used the specific intervention that was developed. As there were very few digital mental health interventions for MND, qualitative research methods were ideal for exploring their acceptability and appropriateness for MND.

The paradigm of critical realism is compatible with the use of qualitative methodology. Qualitative research can be useful to understand the causal mechanisms that underpin reality. In each of the papers in this thesis, specific research questions and methods were used to explore and identify mechanisms that influenced participants' experiences of distress, wellbeing, and their experiences of psychological interventions. This move towards explaining how phenomena come to be, can also help qualitative research have real-world impact (Wiltshire, 2018).

Critical realism holds that there are many different valid perspectives of reality. These perspectives, as held by the people under study and by the researchers, are part of the reality or world that is being understood or studied (Maxwell, 2012). I have used qualitative methodology to understand the views and perspectives of participants, which are central to each of the papers and important in terms of intervention development. My own views were also seen as important in the interpretation of knowledge, and I followed procedures to stay reflexive and critical in this process of acquiring knowledge. Critical realism acknowledges the importance of context rather than trying to draw general conclusions (Denzin & Giardina, 2016). For the purpose of intervention development, I focused on the experiences and meaning of participants receiving care for MND in the UK and the research is concerned with the application of the findings to this particular context. Although findings may be applied more broadly to people with MND and caregivers, this thesis is concerned with the pragmatic utility and validity of application to the particular context and population under study.

Critical realism endorses a pragmatic approach towards selecting research methods (Maxwell, 2012). I was pragmatic in how I approached each of the studies in this thesis in terms of design, data collection and analysis methods, and how I approached intervention development. Having a pragmatic approach involves choosing methods and theories that are useful in particular contexts to solve the problem under study (Giacobbi, Poczwardowski, & Hager, 2005). Using a pragmatic approach also involves creative and imaginative thought to develop solutions, and reflective inquiry and trial and error activity to solve problems (Polkinghoime, 2000). Developing the CALM website involved drawing from existing interventions and research evidence, and presenting information and strategies in novel and sensitive ways for people with MND. Through using think-aloud interview methods and gaining user feedback, the information and strategies were tried and tested with users and the intervention was refined accordingly. This enabled me to use the findings in a practical way to guide intervention development and make recommendations for developing psychological interventions for MND and other neurodegenerative diseases.

1.5.2 Person-based approach

The person-based approach (PBA) was used in this thesis to develop and optimise the intervention. The aim of this approach is to ground the development of the intervention in an understanding of the context of the target user, gained through iterative in-depth qualitative research (Yardley, Morrison, Bradbury, & Muller, 2015). It provides a systematic framework that can guide intervention developers to identify key features that can make the intervention meaningful, useful, and engaging (Yardley, Morrison, et al., 2015). Although not intended to be used solely for digital interventions, it is well-suited for the development and optimisation of digital interventions because these interventions are typically used independently, so understanding the users' context and behaviour is key to facilitating engagement. The PBA can also complement theory- and evidence-based approaches to intervention development, whilst keeping users' perspectives central to the development process (Yardley, Morrison, et al., 2015). The PBA has been widely and successfully used to develop psychological and behavioural interventions. Whilst it has most commonly been used for interventions that support self-management of physical symptoms or health behaviour change e.g. (Band et al., 2017; Bradbury, Dennison, Little, & Yardley, 2015), it has also been used successfully to develop mental health interventions e.g. (Bradbury et al., 2019; Geraghty et al., 2016). This approach was appropriate for developing a DMHI for MND as the evidence for DMHIs and psychological interventions for MND was relatively sparse. Therefore, adopting an exploratory approach to understanding the context of people with MND and caregivers was necessary, as well as exploring their responses to and views of self-guided DMHIs. This approach was also in line with my epistemology and ontology, as it offered a systematic way to guide intervention development whilst being grounded in the perspectives of users through in-depth qualitative research.

Using the PBA at the intervention planning stage involves drawing on theory, evidence, and perspectives of the target users. Qualitative research is central to the PBA at all stages of intervention development and evaluation. Qualitative research can provide rich, contextual information about target users' views and experiences that can influence how they engage with and use the intervention (Morrison, Muller, Yardley, & Bradbury, 2018). The first two papers in this thesis capture relevant contextual factors that influenced the design of the intervention. The first paper involves primary research with people with MND and caregivers about experiences of emotional distress and wellbeing, and the second paper analyses existing qualitative research evidence about people's experiences using psychological interventions in neurodegenerative diseases. Findings from these two studies, along with evidence for effective psychological interventions for MND were used to select intervention content and develop guiding principles that informed intervention development (details are provided in Chapter 4).

Identifying and developing guiding principles is a key element of the PBA. Guiding principles specify the core design objectives and key intervention features that will be used to achieve those objectives (Morrison et al., 2018). Once the intervention is developed, the PBA can be used to understand people's views and experiences of using the intervention. Interventions can then be modified based on users' feedback to make sure they are acceptable, persuasive, and easy to use. In the PBA, this is typically done through think-aloud interviews that enable researchers to observe participants using the intervention whilst saying what they think aloud which can give useful insights into their views of using the intervention (Morrison et al., 2018). After the prototype intervention has been refined based on the think aloud interviews, it is useful to ask users to try out the intervention in real life and then seek feedback about their experiences (Yardley, Ainsworth, Arden-Close, & Muller, 2015; Yardley, Morrison, et al., 2015). In this thesis, Paper 3 reports on the findings from the intervention optimisation process. It includes findings from initial think-aloud interviews, as well as interviews conducted after people with MND and caregivers had used the intervention for a specific period of time. Further details about how the PBA was used can be found in the thesis outline.

1.5.3 Patient and Public Involvement

Patient and public involvement (PPI) is widely recognised as important for healthcare research and developing interventions. PPI is defined as research carried out with or by members of the public, rather than to, about, or for them (NIHR, 2021). In this thesis, PPI contributors or members were both people with MND and caregivers Initially, a call was put out via the MNDA research network for people who were interested in contributing to the project. Three people joined the PPI team through this method. After the first qualitative interview study was conducted, two interview participants wanted to continue to be involved with the project and they were invited to join the PPI team. Recruiting new PPI members at different stages of the project was important, as participation varied when some members' symptoms got worse. Similarly, engagement and participation at PPI meetings varied, and it was decided that it was more efficient and suitable to seek feedback from individuals separately rather than hold PPI meetings.

Including PPI can offer a valuable alternative to the opinions and views of researchers and can improve the design and conduct of research and intervention development (Muller et al., 2019). In this thesis, PPI contributors assisted with various tasks including piloting interview topic guides and open-ended questionnaires, reviewing initial drafts of the intervention, providing feedback on the recruitment and data collection procedures, and insights on the preliminary findings. It can be useful to combine PPI with approaches like the PBA to get a diversity of feedback and develop more engaging interventions (Muller et al., 2019).

1.6 Research context and quality

1.6.1 Researcher background, experience, and training

Reflecting on the researcher's background, training, and interpretations is important for qualitative research. The researcher needs to be aware of how they can influence the research and acknowledge and deal with this in a reflexive way throughout the process (Boyland, 2019). The next few paragraphs outline relevant aspects of the researcher's background, training, and experience, and include descriptions of the steps taken to be aware of or understand how it might influence the research.

In terms of background, I had previously conducted research with people receiving palliative care, people with cancer-treatment related lymphoedema, and people with Parkinson's disease. I also had some clinical experience conducting support groups for people with Parkinson's disease and caregivers. I had some understanding of the problems that people with MND faced, as well as some understanding of the care provided for people with serious illness. As MND is a relatively rare disease, I did not meet many people with MND prior to this project and needed to better understand their needs and context. At the start of the project, I did this by shadowing clinicians at a neurology clinic and attending local MND support groups. My understanding improved during the project through activities such as interviewing people with MND and caregivers in their homes or in hospice settings, attending international and more local MND conferences. I also attended training events organised by the MNDA and British Psychological Society to further understand aspects of psychological support provision for people with MND and caregivers.

With respect to knowledge and training in qualitative research methods, I had some previous experience of using qualitative methods for my studies and work. I needed to further develop some of my skills at qualitative research, particularly the skills needed to develop interventions using the PBA. I undertook further training in qualitative methods at the university, and also contributed to teaching sessions on qualitative methods. I attended training on the PBA and engaged with other researchers using this approach and other qualitative methods through journal clubs, seminars, and conferences. Supervision was extremely important for developing my qualitative research skills particularly during data collection and analysis phases. I sought feedback from my supervisors regarding my interview technique, and regularly discussed any challenges faced during data collection. All my supervisors had extensive experience in using both qualitative research methods and the PBA, so this was a very useful learning experience. I also wrote field notes after every interview and reflected on the methods and questions used.

To familiarise myself with various interventions and therapeutic approaches, I read through the literature and intervention manuals developed by my supervisors, mainly using CBT and mindfulness techniques for other populations. I undertook additional training in assessment, formulation, and interventions for health psychology and attended a session on using acceptance and commitment therapy organised by the MNDA professional support network. I also had a brief placement with Dr. Francesco Pagnini at Università Cattolica del Sacro Cuore to learn about how to adapt and use mindfulness-based interventions with people with MND.

My interest and motivation for doing this project was based on my previous research and clinical experiences. I wanted to continue to work with people with neurodegenerative or serious illnesses and develop interventions to support this group. I was also passionate about research that had application value and would have an impact on improving the lives of people with MND, especially since there is a lack of psychological support options. In order to conduct research that could be applied in the 'real-world', I also needed to engage with and understand the wider care system. I networked with healthcare professionals and the MNDA at various points in this project, especially to advise on more clinical aspects of the disease and available support options. I also liaised with association visitors at the MNDA through local groups and annual general meetings. These interactions led to the development of a public engagement activity at the end of the project where I disseminated my findings and sought feedback about implementing the CALM website in clinical practice.

1.6.2 Quality and trustworthiness

To demonstrate rigour in qualitative research, it is important to assess the quality and trustworthiness of the research. Criteria for judging quality in quantitative research (e.g. representativeness of the sample, reliability of measures) are often inappropriate and incompatible with the ethos of using qualitative methods (Yardley, 2000). Yet, it is important to be able to judge the quality and trustworthiness of qualitative research. Quality can be demonstrated and evaluated in terms of four key dimensions – sensitivity to context, commitment and rigour, transparency and coherence, and impact and importance (Yardley, 2000). These criteria have not been used as rigid prescriptions but have been applied flexibly to the research conducted in this thesis. The next few paragraphs elaborate on these dimensions and provide details about how they were addressed.

Sensitivity to context in qualitative research includes awareness of the context of theory, previous evidence, the socio-cultural setting of the study, and the relationship between the investigators and the participants (Yardley, 2000). This thesis provides a review of existing literature in the area, relevant theory, and previous psychological interventions, and an outline of how the studies fit in with this. In the research context section, the researcher's training and background and reflections on the researcher's knowledge and assumptions has been presented. After the interviews conducted for Paper 1 and 3, field notes were written to reflect on the interview context and the relationship dynamics between interviewer and interviewee. With face-to-face interviews, the researcher could get a sense of the participants' context. With phone and videocall interviews, the researcher asked participants at the beginning to provide a few details about their background and context. At the start of the interviews, the researcher spent some time establishing rapport with participants, explaining that there were no right or wrong answers, and that it was okay to give either positive or negative feedback about the intervention. Input from PPI members also helped ensure that the research methods and questions were sensitive to the context of participants.

The next dimension is commitment and rigour. Commitment refers to prolonged engagement with the topic, development of competence and skill in the methods used, and immersion in the relevant data (Yardley, 2000). The researcher had training and previous experience using qualitative methods, and also developed further skills through more training and supervision (explained in previous section about researcher background). Immersion in relevant data involved conducting interviews and reflecting on them, familiarising through reading interview transcripts and data from the individual studies included in the systematic review. Throughout the data analysis, the researcher wrote memos to reflect on and capture evolving codes and themes. Rigour refers to the completeness of the data collection and analysis, which includes adequacy of the sample and completeness of interpretation of data (Yardley, 2000). The sample in each of the studies was carefully selected to include participants with different characteristics that might influence their experience. In terms of data analysis, the researcher constantly compared themes with codes that reflected the data. Both convergent and divergent cases within each theme were searched for and presented.

The dimension of 'transparency and coherence' relates to the clarity and cogency of the description and argumentation (Yardley, 2000). In Papers 1 and 3, detailed aspects of the data collection and analysis procedure were presented. In Paper 2, the search process, study selection, data extraction and analysis were presented in detail. Checklists were used to enhance the transparency of reporting (e.g. COREQ and ENTREQ) (Tong, Flemming, McInnes, Oliver, & Craig, 2012; Tong, Sainsbury, & Craig, 2007). Checklists were not used in a prescriptive way to indicate rigour or quality, but were used as a guide for transparent reporting and were embedded in a broader understanding of quality in qualitative research (Barbour, 2001). Wherever possible, appendices provided additional study information to enhance transparency of the methods. Themes were presented along with appropriate data extracts or quotes so that readers can discern the patterns identified in the analysis. Transparency also includes adding information about the researcher's assumptions and how they may have affected the product of the research investigation. This was briefly presented in the papers but reflected on in more detail in the section on the researcher's background and context. The story and quality of the narrative is also important in qualitative research. The researcher presented findings to PPI collaborators, supervisors, at academic conferences and to clinical audiences. Engaging with these different stakeholders helped clarify and refine the narrative of research findings.

The dimension 'impact and importance' refers to the theoretical, practical, and socio-cultural impact and utility of the study (Yardley, 2000). The overall aim of the thesis was to develop a DMHI for MND and increase access to psychological support. All 3 studies in this thesis contributed in some way to this overall aim. Additionally, in each paper an explanation of the practical and clinical implications of the findings were presented. Another indirect impact of this piece of work was that it encouraged open discussion about mental health issues and psychological support. This happened not just through the interviews conducted, but also through dissemination activities (e.g. presenting findings at local support groups or at study days for healthcare professionals).

These key dimensions for assessing good qualitative research are intended to be extremely flexible. Their aim is not to prescribe a particular approach to qualitative research but to help researchers to reflect on the methods they use (Yardley, 2015). The quality dimensions and reporting checklists used in this thesis have been used to guide the researcher to reflect on the methods chosen, and have been addressed and adapted differently in the 3 papers.

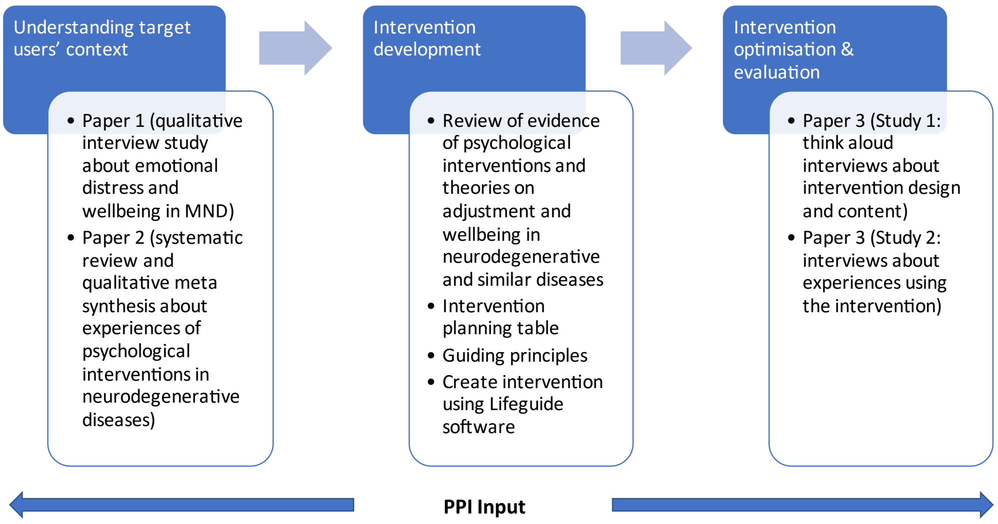
1.7 Thesis outline and three papers

1.7.1 Overview of the thesis and three papers

The overall aim of the thesis was to develop a DMHI for people with MND and caregivers and to develop a broader understanding of how people with MND and caregivers might use or engage with self-guided digital psychological support. This was structured as 2 studies that contributed to the intervention development and 2 studies that contributed to the intervention optimisation and evaluation. Paper 1 was a qualitative interview study with people with MND and caregivers and the aim was to get an in-depth understanding of emotional distress and wellbeing, including the factors that impact distress and wellbeing. Paper 2 was a systematic review that aimed to understand and synthesize the experiences of using psychological interventions among people with neurodegenerative diseases and caregivers. The findings from both these papers were used to guide intervention development, particularly for selecting intervention content and identifying guiding principles. The intervention was created drawing from the website design (e.g. pictures, layout, colour schemes), CBT and mindfulness techniques from an existing intervention for managing distress in primary care called 'Healthy paths through distress' (Geraghty et al., 2016), as well as adding new components (e.g. activities from acceptance and commitment therapy and compassion-focused therapy), and making the overall structure simpler and more accessible for people with MND and caregivers. Paper 3 included findings from two intervention refinement and optimisation studies. In the first study, user feedback was gained using think-aloud interviews with people with MND and caregivers and the intervention was refined accordingly. The second study involved conducting interviews with people with MND and caregivers about their experience using the refined intervention. Figure 7 outlines the different studies within the thesis and how they relate to each other.

Figure 7

Outline of and relationship between different studies in the thesis



In the following sections the aims and the methods of Paper 1, 2 and 3 are described. These sections summarise and extend the information provided within the papers themselves. Due to limited journal word counts it was not always possible to include extensive descriptions and justifications relating to study methods.

1.7.2 Paper 1: Emotional distress and wellbeing among people with Motor Neurone Disease (MND) and their family caregivers: a qualitative interview study

The aim of this paper was to get an in-depth understanding of the emotions experienced by people with MND and caregivers and to explore what impacts emotional distress and wellbeing. The main reason for this focus was to inform the selection of content and relevant activities for the DMHI. As described in the introduction to Paper 1, existing MND literature shed some light on the emotional impact of having MND. However, previous qualitative literature described people's experiences living with MND and experiences of care more generally. Few studies looked specifically and in depth at emotional experiences. For the thesis and the intervention development, it was important to understand the experience of emotional distress and wellbeing from the perspective of people with MND and caregivers, in their own words, as well as understand what may impact distress and wellbeing. It was also important to understand how distress and wellbeing was experienced by participants with diverse symptoms and characteristics, especially if the plan was to develop an intervention that was accessible and appropriate for them. Certain groups of people with MND have been under-represented in existing research (e.g. people with speech difficulties and difficulties with cognition). The next few paragraphs explain the methods used to include the perspectives from these groups in this study.

Purposive sampling was used in this study to select a diverse sample in terms of demographic and clinical characteristics that might influence people's experience of emotional distress and wellbeing. In particular, efforts were made to select and include participants with speech and cognitive difficulties. Disease stage and severity of symptoms could influence people's experiences of distress and wellbeing, and participants with different symptom severities and varying lengths of time since diagnosis were selected.

Interviews were chosen as an appropriate method to collect data because they allowed the interviewer to explore participants' experiences in-depth and they allowed the interviewer to delve into the underlying meaning of what was being said. This was particularly important when talking about emotional distress and wellbeing as participants may not be as open discussing their emotions. Semi-structured interviews were ideal because interview questions could be structured around emotional distress and wellbeing, but also let participants talk about their experiences more generally and enabled unanticipated responses to emerge. Follow up questions can also be formulated based on the interviewee's responses (Flick, 2017). Interviews were initially conducted locally and face-to-face. This enabled the researcher to get an insight into how people with MND and caregivers lived and to see first-hand the problems that people encountered in their daily life. These initial participants were recruited from local MND support groups and represented a certain group of people, i.e. people who had higher levels of disability and had a certain level of acceptance because they were comfortable attending a support group and seeing other people with MND. Telephone interviews were then conducted to recruit people from a larger geographical location. This also meant that people who were earlier on in the disease and those who did not typically access peer support could take part in the interviews. Telephone interviews can be useful when discussing sensitive topics as respondents may prefer the distance and anonymity that a telephone interview offers (Sturges & Hanrahan, 2004). Equally, topics that are emotionally painful may benefit from face-to-face interviews (Sturges & Hanrahan, 2004). A combination of face-to-face and telephone interviews allowed people to choose a method they were more comfortable with, which was important considering the sensitive nature of the interview topic. Written interviews were also conducted to enable people who had difficulties with speech to share their experiences. The length of these interviews varied and follow up questions were asked if more detail or clarification was required. In general, written interviews did not provide very rich accounts, however it was important to capture the experience of this group. Including face-to-face, telephone, and written interviews enabled people with different levels of disability to take part and enabled participants' to express themselves in a format they felt comfortable.

A reflexive thematic analysis was selected to analyse data from the interviews. This method enabled an identification of common patterns across participants' experiences and look for underlying meaning behind experiences of distress and wellbeing (Braun & Clarke, 2006, 2019). This method was selected because it allowed the researcher to build on existing knowledge about the emotional impact of MND, and obtain a more detailed and nuanced account of emotional distress and wellbeing. The patterns of shared meaning are typically united by a central, multi-faceted concept in reflexive thematic analysis (Braun & Clarke, 2021). In Paper 1, these central concepts were identified as things that impacted distress and wellbeing for people with MND and caregivers. The data from interviews with people with MND and caregivers were coded separately and brought together at the theme development stage as the factors that were identified were similar for both people with MND and caregivers. Some differences were present, but these were explained in the paper in relation to the respective theme.

1.7.3 Paper 2: Experiences of psychological interventions in neurodegenerative diseases: a systematic review and thematic synthesis

The aim of this paper was to synthesise the experiences of engaging with and using psychological interventions in neurodegenerative diseases and identify relevant barriers and facilitators. The original focus of this research study was to explore experiences of digital interventions in MND and other neurodegenerative diseases. However, there was a lack of digital interventions developed for this group. The scope of the search was broadened to include psychological interventions in neurodegenerative diseases. Previous reviews have looked at the psychological interventions with MND and concluded that the evidence in this area was limited (Gould et al., 2015; Zarotti et al., 2021). Qualitative studies examining experiences of using psychological interventions in MND is even more scarce. Therefore, the scope was broadened to include other neurodegenerative diseases. The focus of the systematic review was determined through a series of scoping searches.

The decision was made to focus on qualitative and mixed method studies that included a qualitative component. Qualitative syntheses are increasingly used in health research in order to develop an understanding of patient experiences and to explain relationships or unexpected quantitative findings (Atkins et al., 2008). Qualitative syntheses can also be a useful source of evidence to take into account in health policy decisions and forming clinical guidelines to add to the understanding gleaned from quantitative reviews (Higgins & Green, 2011). Qualitative research findings would be able to shed light and give a more meaningful account of barriers and facilitators with respect to acceptability and engagement with psychological interventions in MND. Furthermore, drawing on qualitative research about users' experiences of interventions in order to inform intervention development is consistent with the PBA.

The scope of the systematic review was broadened to include experiences of people with different neurodegenerative diseases and both patient and caregiver experiences. At the same time, the scope was narrowed to include only psychological interventions (educational or therapeutic). Multi-disciplinary and self-management interventions were only included if they had a substantial psychological component and the findings about experience related to this psychological support component. Similarly, if studies used mixed methods but did not have a substantial qualitative component, they were not included in the review.

Findings were analysed using a thematic synthesis. Thematic synthesis is an appropriate method for generating practical recommendations, and can be used to identify barriers and facilitators to using interventions (Thomas & Harden, 2008). This method was also selected because it allowed the researcher to stay close to the original context of the primary studies, and draw conclusions about participants' experiences across a heterogeneous group of neurodegenerative diseases and intervention types through developing analytic themes which go beyond the interpretations presented in the primary studies. In order to assess confidence in the analytic themes developed from this thematic synthesis, the GRADE CERQual approach was used (Lewin et al., 2018). This was a systematic and transparent way to evaluate confidence in our principal review findings that considers 4 components: methodological limitations, coherence, adequacy of data, and relevance. This approach is useful to support the use of findings from qualitative syntheses in intervention or guideline development and policy formulation as it assesses the extent to which the findings are a reasonable representation of the phenomena under study.

1.7.4 Intervention development

The activities undertaken for intervention development were outside of the research activities conducted for the three papers, but they were necessary steps to creating an intervention. The aim of the intervention was to reduce psychological distress and improve psychological wellbeing for people with MND and caregivers.

The intervention was originally planned to be modelled around an existing DMHI called ‘healthy paths to managing stress’ that was used to manage depression in primary care settings (Geraghty et al., 2016). However, the findings from papers 1 and 2 indicated that the website structure needed to be simplified further and be less guided. Some activities from ‘healthy paths to managing stress’ might need to be adapted and new activities added to suit the particular needs of people with MND and caregivers. Therefore, the overall look and design and some activities in the CALM website were replicated from the intervention ‘healthy paths to managing stress.’ However, the CALM website also included some new activities and information.

Intervention development drew on Folkman and Greer’s model of coping with serious illness (Folkman & Greer, 2000). This theoretical model’s focus on meaning-based coping was considered relevant for MND and the theory covered how people used different coping strategies as symptoms changed and got progressively worse. This theoretical model and its recommendations for interventions helped provide a deeper understanding of how to improve wellbeing for people with MND and caregivers and helped guide the selection and structure specific intervention components (e.g. the selection of meaning-based coping strategies and maintaining background positive mood through activities such as the values and goals activity, pleasant activities, and finding positives).

One of the steps for intervention development involved reviewing the literature around evidence for particular psychological interventions or therapeutic approaches, both with MND and other similar life-limiting illnesses. The literature that contributed to specific intervention components has been presented in the intervention planning table in Appendix G. From this review, strategies and suggestions from acceptance and commitment therapy, cognitive behaviour therapy, mindfulness, and compassion-focused therapy were selected.

The findings from the review of people’s experiences using psychological interventions in different neurodegenerative diseases (paper 2) helped shape the guiding principles that were used for intervention development. In particular, findings around the variability of coping preferences, levels of readiness, and potential for interventions to add to burden were used to ensure that the activities in the CALM website were short and easy to carry out and that the overall intervention structure was flexible and not prescriptive.

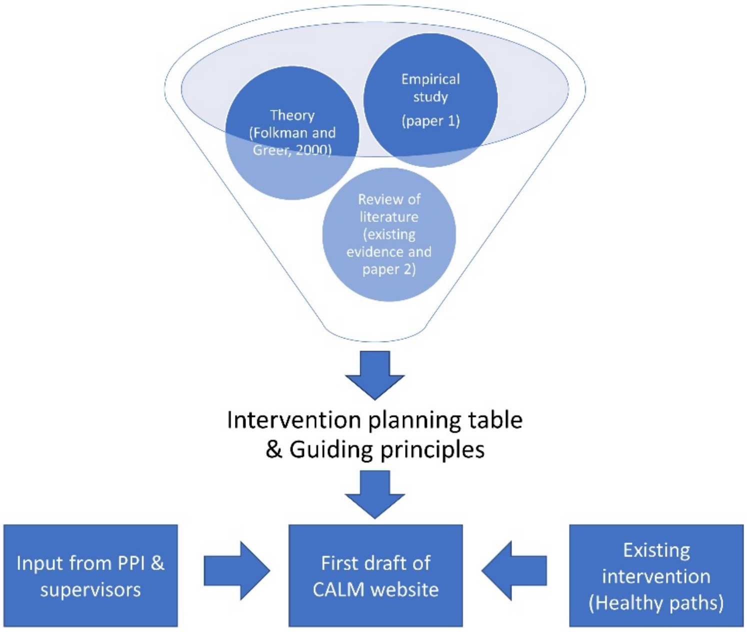
Findings from the empirical study in Paper 1 also contributed to the development of the intervention structure and components through the intervention planning table and guiding principles. In particular, findings around the need for hope and positivity and self-kindness influenced the selection of strategies in the CALM website (e.g. finding positives, compassion break). The importance of a sense of control and experiencing support or empathy from others shaped the structure and features of the CALM website (e.g. adding quotes and examples from peers, having a less guided structure that allowed people to choose activities that suited them). See Appendix G.1 and G.2 for specific details on how these sources of information influenced intervention planning and guiding principles.

The CALM website was created using a software called Lifeguide. The particular software was chosen because it was easy for the researcher to learn and make iterative changes after receiving user feedback. Initial drafts of the website pages were presented to the supervisory team and PPI members and refined based on their feedback. This website was further refined and optimised in the next two studies that are part of Paper 3.

Figure 8 shows the intervention development process for the CALM website that was influenced by these different sources of information and data.

Figure 8

Intervention development process



After integrating information from these different sources, the core structure and content of the CALM website was developed and is outlined below:

1. All users went through a few introduction pages when they first logged in that explained the intervention, its purpose, its creators, the main intervention sections, and how the intervention could be used flexibly to suit their own needs and preferences.

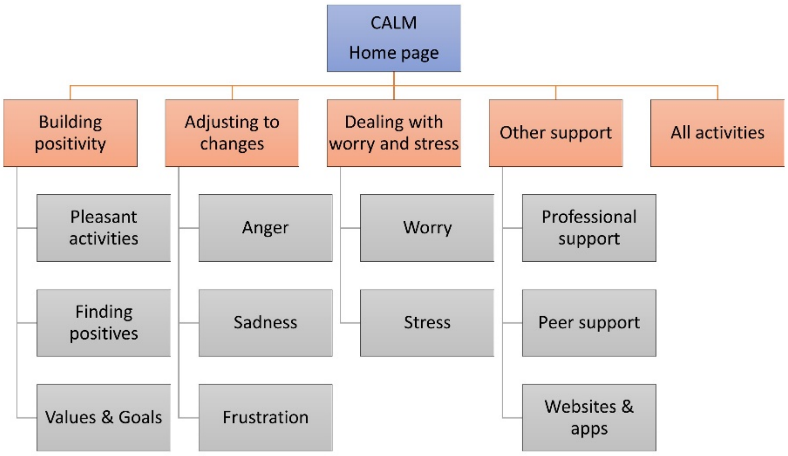
2. Subsequently, whenever users logged in they were taken to the main home page which contained 5 sections – building positivity, adjusting to changes, dealing with worry and stress, other support, and all activities.
3. Building positivity: This section contained 3 activities – pleasant activities, finding positives, and values and goals. Folkman and Greer's theory (2000), findings from the empirical study (paper 1), and existing literature influenced the development of this section. Pleasant activities were taken and adapted from the 'healthy paths' intervention and the other two activities were new additions.
4. Adjusting to changes: This section contained information, suggested activities, and practical tips to deal with anger, sadness, and frustration. The findings from the empirical study (paper 1) influenced the selection and information presented in relation to these 3 emotions. The suggested activity for dealing with anger was a 3-minute breathing space (adapted from healthy paths), for sadness thought distancing was suggested (new activity), and a compassion break for dealing with frustration (taken from healthy paths).
5. Dealing with worry and stress: This section included information, suggested activities, and practical tips for dealing with worry and stress. Similarly, this section was influenced by the findings from paper 1 with respect to worrying about the future and feeling stressed and overwhelmed because of the rate and number of symptom changes with MND. A thought distancing activity was suggested for dealing with worry (new activity) and a safe place meditation was suggested for dealing with stress (new activity).
6. Other support: This section had information that signposted users to further support and resources including professional support, peer support, and other websites or apps. This section was added in response to some of the feedback from the think-aloud interviews about some users needing additional support beyond the CALM website and the information in the section was refined through subsequent feedback from more think-aloud interviews.
7. All activities: This section housed all the different activities that were presented in the CALM website. These included self-kindness activities (compassion break, befriending yourself, self-kindness letter), mindfulness activities (3-minute breathing space, safe place meditation, body scan), activities to help stay positive (pleasant activities, finding positives, values and goals), and a thought distancing activity to help deal with difficult thoughts.
8. Other important features: Quotes were added to foster empathy with the information presented in the website and the value of using some of the suggested

strategies; these were taken from participants in paper 1. Activities were presented along with practical tips as people may have different preferences for coping. There were some suggested activities if users wanted some guidance, but largely the website allowed users to choose sections and activities that were most relevant to them. All information and activities were tailored so that they were relevant to the needs of people with MND and caregivers and attempts were made to word the information sensitively so that they could be used by people with different symptoms and levels of ability.

The structure of the CALM website and the different sections have been presented in Figure 9. A detailed description of the intervention based on the TIDieR checklist has been presented in Appendix H (Hoffmann et al., 2014). For examples of the webpages, layout, and different activities, see Figure 15, Figure 16, Figure 17, and Figure 18.

Figure 9

Main structure and components of the CALM website



1.7.5 *Paper 3: How do people with MND and caregivers experience a digital mental health intervention? A qualitative study*

The aim of this paper was to explore users' experiences of the intervention. The paper focused on evaluating the acceptability, engagement, and usability of the intervention for people with MND and caregivers. Two studies were conducted as part of the intervention optimisation and evaluation process. In the first study, think-aloud interviews were used to understand users' first impressions of the intervention design and content, and feedback was used iteratively to make changes. In the second study, participants' experiences were explored via in-depth interviews after they had used the website for a specific period of time. This was in line with the PBA, with the aim of integrating the perspectives of the target users to develop a more effective and engaging intervention. By using these methods at the early intervention development stage, barriers to acceptability and feasibility can be anticipated, and it is likely that the resulting intervention will be engaging, feasible to implement and effective (Yardley, Ainsworth, et al., 2015).

Purposive sampling was used to select participants for both studies. The aim was to select a diverse range of users who vary in certain characteristics that are important so that the intervention could be adapted to suit these diverse needs (Yardley, Morrison, et al., 2015). Age, gender, type of symptoms (e.g. problems with speech, using hands and legs, cognition), severity of symptoms, and length of time since diagnosis were considered important variables that might influence how people used and engaged with the intervention.

In order to understand the background and characteristics of the target users, demographic and clinical details were collected. Two standardized questionnaires were included – the Amyotrophic Lateral Sclerosis Assessment Questionnaire (ALSAQ-40), a self-report of health status capturing physical mobility, activities of daily living, eating and drinking, communication, and emotional reactions (Jenkinson, 2000). The Amyotrophic Lateral Sclerosis functional rating scale (ALSFRS) was considered for inclusion as it is a validated instrument commonly used across trial with MND (Cedarbaum & Stambler, 1997; Cedarbaum et al., 1999). However, the ALSAQ 40 was thought to be easier for people with MND to complete without support from a researcher or clinician, and it also included a more detailed picture of emotional functioning which was relevant to the particular intervention being researched. This scale also gave a comprehensive picture of the different symptoms people with MND experience. This data also provided context about the different characteristics of people who used the CALM website.

Caregivers completed the Zarit burden interview (ZBI-12). This scale provided a useful indication of distress and burden and was short and easy to complete. The ZBI-12 has also been validated and used with caregivers of people with long-term serious illnesses (Higginson, Gao, Jackson, Murray, & Harding, 2010). The two studies excluded people with dementia or those that did not have the mental capacity to give informed consent. However, people with MND can still experience mild cognitive difficulties and it was important to include people with cognitive difficulties as cognitive impairment can impact quality of life (Mitchell, Kemp, Benito-León, & Reuber, 2010) and it might also impact acceptability and engagement with the intervention. The Edinburgh Cognitive and behaviour ALS Screen (ECAS) is a validated multi-domain screening tool commonly used to detect cognitive deficits in people with motor disorders (Niven et al., 2015).

Although the ECAS is quick and easy to use and has high specificity and sensitivity to impairment characteristics of MND, it does require administration by a healthcare professional (Niven et al., 2015). This was difficult to undertake remotely via videocall and it would involve additional time and burden for participants. Furthermore, the purpose of the two evaluation studies did not require an in-depth assessment of cognitive deficits, but simply an indication of whether this intervention might be applicable to people experiencing cognitive difficulties for inclusion in the sample. Therefore, a trade-off was made and the best measure of cognitive impairment was not included, but brief questions that would provide some indication of cognitive impairment. Three questions were developed with input from supervisors and one PPI member and used so that people with MND could self-report any cognitive difficulties or caregivers could report if their family member was experiencing any cognitive difficulties (see Appendix I). These questions had been previously used in a study with people with Parkinson's disease and were judged by a consultant geriatrician to have face validity and be a useful proxy (Owen, 2020).

Think-aloud interviews were used to gain insight into whether the intervention prototype was acceptable, easy to use, and engaging. Think-aloud interviews are useful as they ask people to give their immediate reactions to different parts of the intervention and allows the researcher to observe how people use the intervention (Yardley, Morrison, et al., 2015). Think-aloud interviews were conducted via videocall, as data collection had to be done remotely due to the COVID-19 pandemic (initially due to lockdown restrictions and later to protect vulnerable participants from infection). Participants viewed the intervention on their own devices and simultaneously answered interview questions. Participants were asked to share their screen so that the researcher could see which part of the intervention the participant was on and how they navigated through the intervention. After every 2-3 interviews, the researcher collated participants' feedback and entered them into the table of changes. The table of changes was used as part of intervention development, hence details of all the changes made were not presented in Paper 3. Participant feedback was discussed within the research team and decisions about changes were made based on whether the suggested change was in line with the guiding principles, easy to implement, and repeated by multiple participants. The PBA provides a systematic approach for deciding how to modify the changes based on the user feedback and also draws on the MOSCOW (Must have, Should have, Could have, Would like) approach for prioritising changes (Bradbury, Watts, Arden-Close, Yardley, & Lewith, 2014). This process was an iterative cycle where the next few interviews checked if the changes made were suitable, and this continued until fewer suggestions for change were made and participants found the intervention suitable and easy to use.

After the intervention has been refined, it is useful to ask users to try out the intervention on their own, without the researcher present, and interview them about their experiences using the intervention (Yardley, Morrison, et al., 2015). Participants were given 6 weeks to try out the intervention. Based on the size and amount of information and activities presented in the intervention, 6 weeks was considered an appropriate amount of time for participants to go through the whole intervention, and put into practice any advice or techniques they found helpful. At the end of this 6-week period, semi-structured interviews were used to gain feedback from participants. To enable people who had difficulties with speech to give feedback on the intervention, written interview responses were also collected. This involved sending participants a link to a survey with open-ended questions outlining questions about the different sections and activities in the intervention. The researcher then followed up with an email with specific follow-up questions drafted based on participants' responses. Interviews via email are a convenient and practical alternative and can offer interviewees time and space to think about and answer interview questions (Hawkins, 2018). Although this does not always mean that interviewees will answer questions in a spontaneous manner as with face-to-face interviews, they helped give participants with speech difficulties time and space to type out their responses without the time burden. Written responses also helped capture the views and experiences of participants who would be excluded from some research and therefore it was important to have this data collection option. A disadvantage of written or email interviews is that it can often result in short, concise answers and the lack of non-verbal and contextual cues may make interpreting the interviewees' responses difficult (Hamilton & Bowers, 2006; Hawkins, 2018). This was also found in the evaluation studies where responses were often concise and sometimes difficult to fully understand how participants used certain components of the intervention or how it benefitted them. The researcher followed up with further questions via email, asking participants to elaborate on how they used particular techniques that they mentioned in their responses, or asked them to clarify the meaning of responses that were not clear. Some participants would have likely used communication aids or relied on other people to type their responses, and this may have also contributed to the limited amount of data provided from the written interview responses.

Data from the think-aloud and post-intervention interviews were combined and analysed in order to understand participants' experiences of using the intervention. Interview data was analysed drawing on methods for conducting an inductive thematic analysis (Braun & Clarke, 2006). Some of the analysis included qualitative description, which was less interpretive and provided more of a summary of experiences in relation to the particular theme (Sandelowski, 2000). These methods were chosen because the researcher wanted to identify and report patterns that were grounded in participants' experiences of using the intervention. Having an inductive approach is in line with the PBA where the goal is to ascertain the most important issues and challenges for the participant (Morrison et al., 2018). The aim was to provide a rich thematic description of the entire dataset as participants' views on this topic are under-researched or not known (Braun & Clarke, 2006).

The next 3 chapters provide details and findings from these 3 papers. Two of the papers have been published in peer-reviewed journals and Paper 3 has been submitted to a journal for consideration. The final chapter brings the findings from the 3 papers together and discusses implications for research and clinical practice.

Chapter 2 Emotional distress and wellbeing among people with Motor Neurone Disease (MND) and their family caregivers: a qualitative interview study

Pinto, C., Geraghty, A. W., Yardley, L., & Dennison, L. (2021). Emotional distress and well-being among people with motor neurone disease (MND) and their family caregivers: a qualitative interview study. *BMJ open*, *11*(8), e044724.

2.1 Abstract

Objective: We aimed to get an in-depth understanding of the emotions experienced by people with Motor Neurone Disease (MND) and their caregivers and to explore what impacts emotional distress and wellbeing. **Design:** Qualitative study using semi-structured interviews with people with MND and caregivers. **Setting:** Participants were recruited from across the UK and took part in interviews conducted either face-to-face, by telephone or email to accommodate for varying levels of disability. **Participants:** 25 people with MND and 10 caregivers took part.

Participants were purposively sampled based on their MND diagnosis, symptoms and time since diagnosis. **Data analysis:** Data were analysed using inductive reflexive thematic analysis. **Results:** Eight broad themes were generated (20 subthemes). Participants described the emotional distress of losing physical function and having a threatened future because of poor prognosis. Keeping up with constant changes in symptoms and feeling unsupported by the healthcare system added to emotional distress. Finding hope and positivity, exerting some control, being kinder to oneself, and experiencing support from others were helpful strategies for emotional wellbeing. **Conclusion:** The study provides a broad understanding of what impacts emotional distress and wellbeing and discusses implications for psychological interventions for people with MND and caregivers. Any communication and support provided for people with MND and their caregivers, needs to pay attention to concepts of hope, control, and compassion.

2.2 Introduction

Motor neurone disease (MND) is a neurodegenerative disease, which results in declining physical function and has a very poor prognosis (Moore et al., 2008). In terms of psychological impact, many people with MND (pwMND) experience depression (Clarke et al., 2005; Goldstein et al., 1998; Hogg et al., 1994). Anxiety is prevalent around the time of diagnosis (O'Brien et al., 2011; Pavey et al., 2013) and during the final stages (O’Brien, Whitehead, Jack, & Mitchell, 2012). PwMND also experience feelings of hopelessness (Averill et al., 2007; McLeod & Clarke, 2007; Soundy & Condon, 2015) and demoralization (Clarke et al., 2005).

Research has described the impact of the significant losses that come with MND on personal, social and occupational relationships (Soundy & Condon, 2015). Family members or caregivers of pwMND often struggle with the emotional impact and burden of the disease, and have high rates of psychological morbidity (Goldstein & Leigh, 1999; Pagnini, 2013; Rabkin et al., 2009). Studies with caregivers have described the strain from caring tasks, from having to make changes to their own lives and not having time for themselves (Brown, 2003; Cox, 1992; Olsson Ozanne, Strang, & Persson, 2011). Distress in MND has largely been described as anxiety and depression (Averill et al., 2007; Galvin et al., 2016; Pagnini, 2013; Rabkin et al., 2009), or caregiver burden and strain (de Wit et al., 2018; Goldstein et al., 2006). Wellbeing has largely been described as reduced anxiety and depression or improvements in quality of life (Larsson et al., 2016; Matuz et al., 2010, 2015). In this paper, we use the terms ‘emotional distress’ to refer to the broad range of negative or difficult thoughts and emotions, and ‘emotional wellbeing’ for the broad range of positive or helpful thoughts and emotions experienced by pwMND and caregivers.

Studies have looked at demographic and clinical factors that might explain emotional distress or protect against it (Matuz et al., 2015). Results from these studies show that psychosocial factors like coping strategies and social support, are more strongly related to wellbeing and quality of life than demographic and clinical factors (Goldstein et al., 2006; Goldstein et al., 2002; Matuz et al., 2015). Emotions are also affected by low self-esteem (Goldstein et al., 2006; Hogg et al., 1994), end of life concerns (Averill et al., 2007), faith/existential concerns (Bolmsjö, 2001; Foley et al., 2007), sense of loss (Foley et al., 2007; Mistry & Simpson, 2013), and changes in identity, roles and relationships (Foley et al., 2007; Goldstein et al., 2006; Sakellariou et al., 2013). Psychosocial factors are clearly important for distress and wellbeing in MND, and we need a better understanding of what influences emotional distress and the use of different coping strategies (Fisher et al., 2019; Larsson et al., 2016).

Qualitative research can be useful for understanding emotional distress and wellbeing because it allows participants to express their own understandings and experiences without being limited to concepts pre-determined by the researcher. Qualitative studies have largely explored the experience and impact of living with MND (Cox, 1992; Foley et al., 2007; King et al., 2009; Soundy & Condon, 2015), which have added to understanding of distress in MND. Few studies have focused specifically on the emotions experienced. One qualitative study examined the use of metaphors to express emotions (Locock et al., 2012), and a small number have focused on emotions at specific time periods (e.g. during diagnosis or the final stages) (Mistry & Simpson, 2013; Pavey et al., 2013; Whitehead et al., 2012). Emotional distress at other time points and things that influence distress and wellbeing need further exploration.

This study aims to build on our understanding of emotional distress and wellbeing in MND. More importantly, we aim to hear from people whose experiences are not well represented in research, including people with speech difficulties, cognitive impairment, and people at different stages of the disease. It is particularly timely to do this research because recent articles have highlighted that there are few effective psychological interventions to improve wellbeing (Gould et al., 2015) and more research and interventions are needed (Pagnini, 2013; Zarotti, Mayberry, Ovaska-Stafford, Eccles, & Simpson, 2021). This study is part of a larger project to develop an intervention to improve emotional wellbeing among pwMND and caregivers.

In this current study, we aim to understand the emotional impact of living with MND and explore what impacts experiences of emotional distress and wellbeing among pwMND and their family caregivers.

2.3 Methods

2.3.1 Design

Qualitative study using in-depth semi-structured interviews and reflexive thematic analysis, in line with an interpretivist approach. The paper is reported in accordance with the Consolidated criteria for reporting qualitative research (COREQ) (see Appendix A) (Tong et al., 2007).

2.3.2 Participants

We aimed to recruit 20-30 pwMND and used purposive sampling to represent people with difficulties with movement, speech and cognition, and different lengths of time since diagnosis. Caregiver participants had fewer sampling criteria (age, gender), therefore we aimed to recruit 10-15 caregivers.

Eligibility: Participants were above 18 years of age, had an MND diagnosis, and had mental capacity to consider participation in the study (assessed by the researcher through correspondence about the study). We included participants who self-reported difficulties with cognition, but had mental capacity to give informed consent, as the views of pwMND who have cognitive impairment have been underrepresented in previous research. We included caregivers above 18 years of age, both current and recently bereaved (bereavement within 1 year from the time of the interview).

2.3.3 Data collection

Participants were recruited through a UK charity that supports pwMND and their families (Motor Neurone Disease Association). Study information was circulated via the charity's website, newsletters, social media outlets and local support groups. People willing to take part contacted CP, who screened for eligibility and provided further details about the interview.

Before each interview, participants gave written informed consent and filled a demographic/clinical details form. This process was completed either in-person, by post or email, based on the interview mode. CP, who has training and experience in conducting interviews for qualitative research, conducted all interviews. Interview mode was flexible (face-to-face, email or phone) to accommodate for various levels of disability. Face-to-face interviews were conducted in a place convenient to participants, usually at home or a hospice. Where two members of a couple had both consented to participate participants were interviewed separately where possible, but jointly where this was requested for reasons of comfort or to facilitate easier communication. The interview topic guide was developed iteratively by CP, LD, AG and patient and public involvement members. In line with an interpretivist approach, questions were broad and open-ended to allow participants to give rich, in-depth accounts of their emotional experiences in relation to MND, and follow-up questions were led by participants' responses. The final interview topic guide (see Appendix B) covered questions about people's experiences living with MND, with a focus on their thoughts and feelings and coping with emotional concerns.

2.3.4 Data analysis

Face-to-face and phone interviews were audio recorded and transcribed, and all interviews were anonymized. Field notes were completed after each interview, reflecting on participants' responses, and interview method and procedure. Data were analysed using reflexive thematic analysis (Braun & Clarke, 2006, 2019) as this was a flexible method that suited the research question, helped us identify common patterns across participants' experiences and allowed us to look for underlying meaning behind experiences of emotional distress and wellbeing. In line with an interpretivist approach, we used an inductive approach to data analysis, and included convergent and divergent cases in theme development. The analysis was mainly conducted by CP, a PhD student with previous experience as a qualitative researcher. After familiarisation with the interview transcripts, Nvivo 12 software was used to code the data inductively, focusing on semantic and latent features of the data. The codes went through several iterations as new interviews were coded. During the coding process, notes were made about interesting features of the data and how different concepts related to each other. Following this, similar codes about emotional distress and emotional wellbeing were grouped together and candidate themes were generated. Themes were then reviewed to see if they represented experiences across the dataset. Theme names were revised and findings written up; this was also an iterative process whereby descriptions were clarified and overlap between themes identified. CP had regular meetings with qualitative research experts LD and AG who helped refine codes and themes. A lay summary of the findings was sent to all participants.

2.3.5 Ethics

We obtained ethics from the University of Southampton ethics committee (ERGOII-46996). Written informed consent was obtained for all participants. Participants were debriefed and steps were taken following each interview to ensure participants were not distressed.

2.3.6 Patient and Public Involvement (PPI)

Three PPI members (one person with MND and 2 former caregivers) contributed to this study. They were involved in the recruitment stage to help identify potential participants. They also helped pilot the interview topic guide, refine interview questions and trial the data collection procedure to ensure that it was not burdensome. Some PPI members looked over early drafts of the findings and offered insight about elements to highlight or discuss in the reporting of the results.

2.4 Results

Participants were 25 pwMND and 10 family caregivers (see Table 1). 29 interviews were conducted in total; 6 joint interviews and 23 one-to-one interviews with either pwMND or caregiver. 14 interviews were conducted face-to-face, 8 via telephone, and 7 via email. Interviews lasted an average of 39.6 minutes.

Table 1

Demographic and clinical details of the sample

Characteristic	People with MND (n=25)	Caregivers (n=10)
Age (range 39-80)		
35-50 years	3	0
51-65 years	12	4
66-80 years	10	6
Gender		
Male	15	5
Female	10	5
Relationship to person with MND		
Spouse/partner		10
Diagnosis		
ALS limb	18	
ALS bulbar	1	
Primary Lateral Sclerosis	4	
Progressive Bulbar Palsy	1	
Progressive Muscular Atrophy	1	
Time since diagnosis (range 2 months-17 years)		

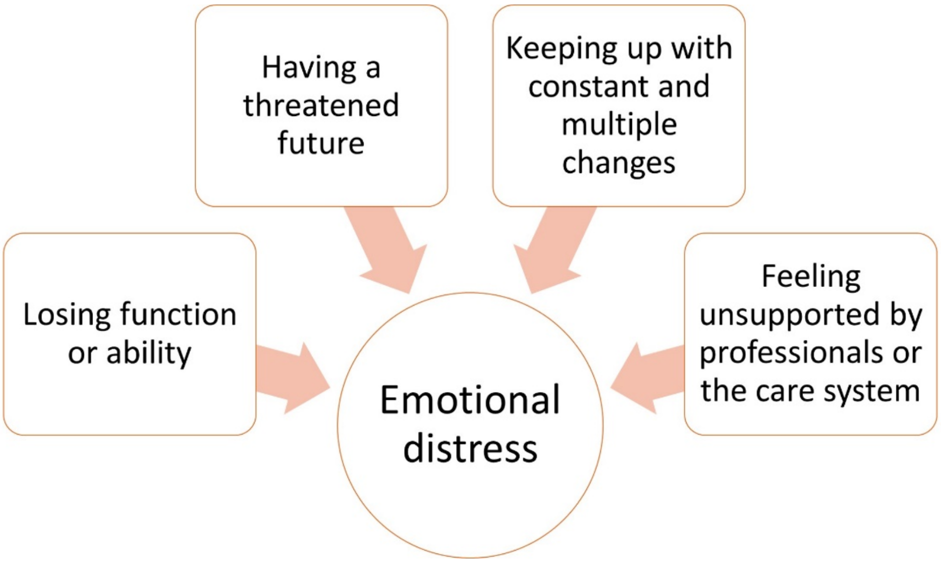
Characteristic	People with MND (n=25)	Caregivers (n=10)
Less than 1 year	8	3
1-3 years	6	4
3-5 years	7	2
More than 5 years	4	1
Difficulties reported often/always [*](ALSAQ-5 scale)		
Difficult to stand up	16	
Difficult to use arms and hands	16	
Difficulty eating solid food	4	
Speech is not easy to understand	9	
Feeling hopeless about the future	3	
Self-reported concerns about cognitive ability	2	

[*] Amyotrophic Lateral Sclerosis Assessment Questionnaire (ALSAQ-5) is a patient self-report questionnaire used to briefly measure the impact of ALS/MND on patients (Jenkinson & Fitzpatrick, 2001)

We developed 8 themes and 20 subthemes. The first four themes relate to triggers of emotional distress (see Figure 10); the remaining four themes capture strategies to improve emotional wellbeing (see Figure 11). All themes were present in some degree and form in both patient and caregiver interviews; where differences between these groups were apparent, they are described.

Figure 10

Triggers of emotional distress



2.4.1 Triggers of emotional distress

Participants described 4 main triggers of emotional distress and how they had an impact on emotions (themes are numbered, *subthemes in italics*).

2.4.1.1 Losing function or ability

Most participants described how losing function or ability was distressing because of a *reduced sense of autonomy and control*. PwMND spoke about feelings of anger, frustration, sadness or grief at not being able to do the things they wanted to, at losing their independence and relying on others for help. Caregivers expressed similar emotions at seeing their family member lose capability and independence.

> Hobbies that I had, I can't pursue any of them because I just don't have any capability enough in my arms now to do that. So, I think mentally I've seen a dip in the last six months into a much more kind of negative and angry stage really. (Person with MND, P13)

> ...and seeing him, and it saddened me, where you're sort of doing the job of hoisting and moving, the manual handling, and then you step back and you're seeing it from a distance, that's actually quite [pauses] hard as well, because he's

> got no control over his body and he's having to have people do that for him. (Caregiver, C05)

Losing function led to *changes in self-identity*, specifically those of becoming a 'patient' and 'caregiver'. These changes, especially losing one's voice which is strongly linked to identity, were experienced as distressing.

> Losing mobility gradually was bad enough… but losing my voice is much more dreadful. It's taking away part of my personality. I can no longer express myself properly. (Person with MND, P21)

Losing function or ability sometimes led to *changes in relationships and interactions*. PwMND spoke about negative feelings of becoming a burden for their partner/spouse. Both patient and caregiver participants spoke about how having MND sometimes changed or limited interactions between family members.

> It's emotionally difficult not being able to physically help my son or my parents and siblings and friends. Not being able to hold and play with my nephew. (Person with MND, P26)

Participants also had *practical concerns or worries about managing tasks* as a result of losing more ability and function.

> My main worry at the moment really is moving him from there to there because his knees and lower legs are getting weaker and I've got that [points to hoist] to move him. (Caregiver, C03)

2.4.1.2 Having a threatened future

The short prognosis that accompanies a diagnosis of MND led to feelings of anger, disappointment, sadness and feelings of being robbed/cheated of a future (*threat to life narrative and future plans*).

> But I'm having to accept I might have a lonely old age. Disappointment, more than anything else. (Caregiver, C27)

Participants also described emotional distress when *anticipating future symptoms or disease progression*. Many things triggered worry about the future including researching MND online, meeting other pwMND, discussions with healthcare professionals and going through symptom checklists. Some participants also spoke about experiencing such worries about the future even if symptoms had not progressed or were relatively stable. This was true particularly just after diagnosis, where worries about the future led to feelings of depression, low mood and withdrawal from others.

> Even though physically, say that first month afterwards, there was almost no change but my mind's thinking quite morose almost. You almost think of death at that point... It's not even anger at that stage, it's just hopelessness at that point because you're just thinking oh that is it isn't it? It's all over. (Person with MND, P10)

The *uncertainty of the disease progression* also caused emotional distress among both patients and caregivers, because of being unable to manage symptoms or plan for the future.

> I think the biggest thing about this is that it's such an unknown. Because everybody is different and, who knows how much, how long this disease is going to affect you. And the uncertainty, for me, it's very hard to cope with. (Caregiver, C15)

2.4.1.3 Keeping up with constant and multiple changes

Participants discussed how the *timing and relentless nature of changes* in symptoms was difficult physically and emotionally. As time went on, for some it became easier to fall into a care routine. However, if deterioration happened quickly, both patient and caregivers expressed that it was difficult to cope and left them feeling tired, not in control and in need of respite.

> If something's difficult, we find a way round it. And by the time we've found a way round it, things have moved on again, and it doesn't work. So that's very frustrating. And a bit… not depressing… soul destroying, because we're always playing catch up. (Person with MND, P24)

Sometimes physical symptoms necessitated *changes in many areas of life.* This included changes to their home, work, social and leisure activities. Both pwMND and caregivers spoke about the effort involved in either living with MND or looking after someone with MND. This sometimes affected thoughts and mood negatively including feelings of frustration, anxiety, tiredness or strain/burden. For example, C03, a caregiver, recalled being upset and needing someone to talk to due to exhaustion from physically moving her spouse; “I used to get so tired that sometimes when he had gone to bed in the evening, I did just feel like standing there crying.”

2.4.1.4 Feeling unsupported by professionals or the care system

Participants reported feeling distressed when healthcare professionals or the care system did not support their needs. *Infrequent or inefficient care provision* left people feeling abandoned and left on their own to deal with MND.

> I felt completely unsupported by the neurologist or the health service, we were really just left on our own, completely in the dark as to what was gonna happen... So, it was a very, very distressing time. (Person with MND, P14)

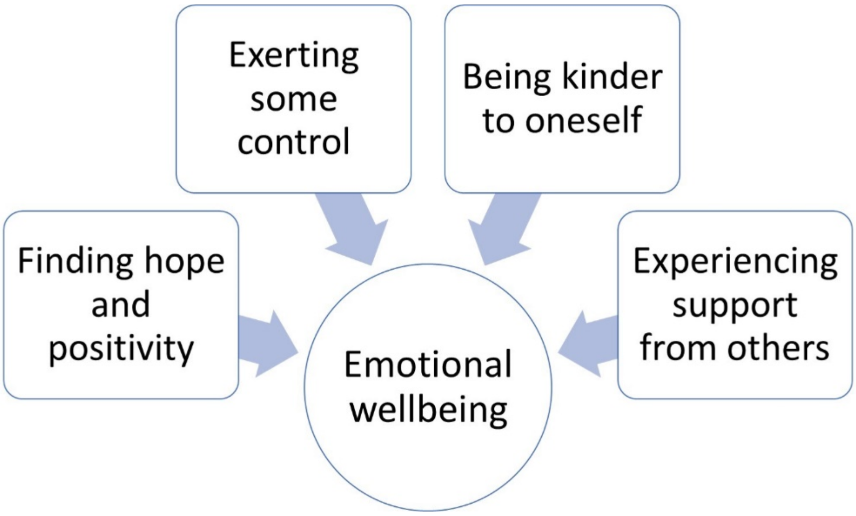
Poor coordination between different professionals/organizations, difficulty accessing equipment and financial benefits also triggered feelings of frustration, injustice and being ‘let down’ by the system.

Poor or insensitive communication from healthcare professionals added to emotional distress. In particular, participants described upset as a consequence of a lack of empathy from professionals, or insensitive or unsupportive presentation of care.

> Our life was shattered in 2 minutes. And I felt that was cruel, very very cruel [pause]. There should be a way of discussing it with you and saying “well look, you have such and such a condition, but hey, this is what we can do.” Not just you’ve got this and throw you out, and so we had to find our own way round this problem. (Caregiver, C01)

Figure 11

Strategies used to improve emotional wellbeing



2.4.2 Strategies used to improve emotional wellbeing

Participants described strategies they used to improve emotional wellbeing (themes are numbered; *subthemes in italics*).

2.4.2.1 Finding hope and positivity

Despite the many losses, participants actively found ways to introduce hope and positivity. For many people, acceptance of MND was a gradual, learned process and even once there was acceptance, an attitude of not giving up on life was important for coping (*acceptance and not giving up)*.

> I always wanna think I've got options, got somewhere to go because think at the point where you think "well I've got nowhere left to go with this," that's when you may sort of deteriorate and let it get the best of you. But I think if you if you say, "right well I'm going to, you know, there's this trial, this might happen, I'll go and see a physio…" and just see if that helps me. (Person with MND, P18)

For some, not giving up included hoping for a cure or trying alternative therapies. For others, not giving up included finding solutions to make life easier, and not letting MND define them as they continued normal and valued activities. This strategy was endorsed mainly by pwMND.

> Not giving up, not allowing it to put you away in a corner somewhere, waiting for the inevitable… not allowing that to define what you do with whatever time you have left. (Person with MND, P06)

Despite severe losses of function, many participants continued *doing activities that they enjoyed and that were meaningful*. Planning ahead and having things to look forward to in the immediate future added to feelings of hope.

> Key thing is to have lots of little goals, lots of little hopes of things to do, normalities, weekends away, holidays but not in the far future. Everything's in a couple of weeks, 2-3 weeks, something to look forward to… it keeps the mind occupied to know that there is something coming up. (Person with MND, P10)

Most participants tried to *focus on the positive aspects of life*. This strategy became difficult in the context of increasing loss, but participants still stressed that it was important.

> I'm very much sort of an optimist. You can't…you know, because it's horrible we know that. But we just make [name of pwMND] as comfortable as possible.
>
> You've got to be positive, otherwise you just sink and go under. (Caregiver, C07)

To stay positive and deal with negative emotions, participants tried to reframe difficult situations by focusing on what they still can do, using humour, feeling grateful for symptoms they did not have or feeling lucky because of their circumstances or because of the support they received.

2.4.2.2 Exerting some control

When MND took some control away, participants found other ways to exert control over their lives. Although a sense of control was important for most participants, they exerted this control in very different ways. One way was by *focusing on the present moment* and making the most of what they could currently do. This strategy was adopted mainly by pwMND; it stopped them from worrying about the future or feeling sad for what they could no longer do, and instead had a more positive impact on emotions.

> I realised there's absolutely no point in dwelling on the past because there's absolutely nothing I can do to get that back. So, it's waste of energy and there's no point in worrying about the future because there's absolutely nothing I can do

> about it. So, what I kind of taught myself to do is try to live in here and now because I do have some control over that. (Person with MND, P04)

Some participants described how it was helpful to exert *control over decisions about their care,* organizing or planning care and taking a more active problem-solving approach. This gave people a sense of empowerment when they would otherwise be struggling to cope, and also helped people hold onto their independence or identity despite the loss.

> I hate the fact that I can't talk. Although I can talk, but it's a bit drained and doesn't sound like me. But I've done voice banking and I think you've got to be a step ahead. (Person with MND, P19)

> They're [professionals] pretty good at picking up what might be the next thing that's necessary or how tired I'm looking and dropping in a suggestion here and there about what you might change, what you might need next… in terms of future proofing, so you're on top of a problem and not floundering about solving it. (Caregiver, C05)

However, some people exerted control by avoiding interactions or information that might remind them of future symptoms or deterioration and associated negative emotions.

> Everyone's different, aren't they? And their MND is different. Some people like to know the ins and out, but I don't know, I like to bury my head in the sand. Because I know that I would sit and worry about it all the time. And I don't want to. (Person with MND, P24)

2.4.2.3 Being kinder to oneself

This strategy was mentioned by both pwMND and caregivers but for different kinds of tasks. Living with MND was often described as overwhelming and participants expressed the need for *taking a break* from MND. Caregivers spoke about the need to take a physical break from MND care; while pwMND spoke about taking a break from MND by doing regular activities and not thinking about MND.

> I'll have days where I deliberately avoid looking at anything and trying just to have days or a couple of days where I don't think about MND. (Person with MND, P18)

Some participants also expressed the need to give themselves *time and space to adjust to changes* in symptoms. This was done by allowing themselves space to vent if they felt overwhelmed, pacing their activities, lowering expectations, modifying how tasks were done, or asking for help.

> Keep doing the things you enjoy, just lower your expectations and standards so you achieve and don't feel disappointed. (Person with MND, P21)

2.4.2.4 Experiencing support from others

Emotional and practical support from healthcare professionals, friends and family helped pwMND and caregivers cope with any emotional distress. Emotional support included being empathic, listening, being encouraging and positive. Participants also valued support from other people with MND because they felt truly understood and less alone.

> I had a confirmation diagnosis at [hospital] and then I had a phone call from the [hospice] asking if I wanted to attend a kind of MND first contact group... and it's been a fantastic thing and I still go now. And that was really good, really supportive. (Person with MND, P19)

There were some differing views about the timing of offering support, especially just after diagnosis. Some participants wanted access to information and support almost immediately, however others needed some time to adjust before they accessed support.

Supportive communication from healthcare professionals and reliability of care helped patients and families feel reassured and confident about their care, which led to positive experiences and emotions. This included providing information based on the patient and families' readiness, communicating information in a sensitive and empathic manner, focusing on what can be done in terms of care and timely provision of care and equipment.

> I would also mention my occupational therapist who has been brilliant at assessing my needs and getting in equipment quickly, usually just before they were needed. This has given me more confidence in the care and support I am given. (Person with MND, P22)

2.5 Discussion

This study advances our understanding of what influences emotional distress and wellbeing among people with MND and their caregivers. Our findings come from a diverse sample, thereby highlighting key triggers of distress and coping strategies used by people with different abilities, symptoms and at different stages of having MND. PwMND and caregivers use coping strategies differently but still rely on similar concepts (hope, positivity, control, self-kindness, social support) to improve emotional wellbeing.

Some of our findings are in line with previous MND research, such as the distress caused by loss of ability and threats to the future (Foley et al., 2014; Mistry & Simpson, 2013; Sakellariou et al., 2013; Soundy & Condon, 2015). Previous research has also identified distress caused through not receiving appropriate professional support (Holkham & Soundy, 2018; Hugel et al., 2006; O'Brien et al., 2012; Pavey et al., 2013; Soundy & Condon, 2015; Whitehead et al., 2012). We highlight the emotional distress triggered by multiple and constant changes brought about by disease progression and constantly 'playing catch up.' This is a new finding in terms of emotional distress for pwMND, but has been described in the literature around caregiver experience (Aoun et al., 2013; Galvin et al., 2016; Oyebode, Smith, & Morrison, 2013). This is an important finding for intervention development, and we need to ensure that interventions are perceived as manageable, not burdensome. Psychological interventions delivered via videoconference have been suggested as a way to make interventions more manageable and accessible for people whose MND symptoms make mobility difficult (Weeks et al., 2019). Intervention flexibility in terms of choice of content, session duration, number and structure of sessions may also make interventions less burdensome for people with MND and caregivers.

In terms of emotional wellbeing, hope and control are particularly important for MND (Fanos et al., 2008; Foley et al., 2014, 2014; Holkham & Soundy, 2018; Soundy & Condon, 2015). Soundy and Condon (2015) have developed a model to show how concepts of hope and control can affect mental wellbeing in MND (Soundy & Condon, 2015). Hope and control can be difficult concepts to apply in MND where hope and control are constantly being threatened. Our findings highlight these complexities, and provide examples of how pwMND and caregivers use hope and control to maintain and improve wellbeing, despite this threat. For example, we found that many people use meaning-based coping strategies by either re-appraising difficult circumstances in a positive way or doing activities that were important and meaningful. However, for some accepting their circumstances was difficult and hope meant adopting an attitude of not giving up and not letting MND define them. Psychological interventions such as acceptance and commitment therapy can be useful and can foster meaning-based coping strategies (Graham, Simmons, Stuart, & Rose, 2015; Kangas & McDonald, 2011; Weeks et al., 2019). However, there might be certain challenges in how interventions are presented to people who may find acceptance difficult with MND. Our findings also show how control is exerted differently in the face of loss, by focusing more on the present moment, or by making decisions about receiving information and care. This has implications for support services, especially in providing options for care, equipment and communication aids (Pavey, Warren, & Allen-Collinson, 2015). The importance of focusing on the present demonstrates the value of mindfulness-based approaches for psychological interventions. The differences in how people use control highlights the importance of having flexible support that is tailored to the individual's needs and coping preferences (Weeks et al., 2019). Another novel finding in relation to emotional wellbeing and MND is the importance of self-kindness and self-care. Self-compassion has been associated with adaptive coping strategies and wellbeing other chronic illnesses (Pinto-Gouveia, Duarte, Matos, & Fráguas, 2014; Sirois, Molnar, & Hirsch, 2015; Wren et al., 2012) and similar approaches may be useful for pwMND and their caregivers.

2.5.1 Strengths and Limitations

We aimed to recruit a diverse range of participants and succeeded in sampling people with different symptoms of varying severities, particularly people with difficulties speaking. We did not manage to recruit many people with mild cognitive impairment and could have benefitted from a more targeted sampling approach. Although we captured the experiences of newly diagnosed people and people who had MND for several years, we could not ascertain if we captured the experiences of people who were at the end stages. Conducting email interviews ensured we included the experiences of people with speech difficulties. The total number of email interviews were small and there were some methodological difficulties in using prompts and asking follow-up questions. Despite these limitations, including the experiences of under-represented groups gives us confidence in the application of these findings for people with different symptoms and at different stages of MND.

2.5.2 Conclusion

We provide a broad understanding of what impacts emotional distress and wellbeing among pwMND and their caregivers. Findings have important implications for psychological interventions, services, and professionals that support pwMND and their families. Any communication and support provided for pwMND and their caregivers, needs to pay attention to concepts of hope, control, and compassion, and how individuals may use these concepts differently to cope with the emotional impact of MND.

Chapter 3 Experiences of psychological interventions in neurodegenerative diseases: a systematic review and thematic synthesis

Pinto, C., Geraghty, A. W., McLoughlin, C., Pagnini, F., Yardley, L., & Dennison, L. (2022). Experiences of psychological interventions in neurodegenerative diseases: a systematic review and thematic synthesis. *Health Psychology Review*, 1-23.

3.1 Abstract

Background: Psychological interventions for managing emotional distress in neurodegenerative diseases are needed, but progressive worsening of symptoms and increasing disability might pose difficulties with engagement. We aimed to synthesise the experiences of engaging with and using psychological interventions in neurodegenerative diseases and identify relevant barriers and facilitators.

Methods: Systematic searches were conducted in 6 electronic databases and results were screened. We included qualitative and mixed methods studies reporting patient or caregivers' views or experiences of psychological interventions. Qualitative data were extracted and thematically synthesised.

Results: 34 papers were included, covering a range of diseases and interventions. Engagement was facilitated by flexible intervention formats and tailoring to the specific needs of people with neurodegenerative diseases. Interventions were sometimes inaccessible or burdensome because of physical and cognitive symptoms, and the time and effort required for the intervention. Participants' levels of acceptance and readiness often differed and influenced engagement with the intervention. Across different interventions, participants experienced wide ranging benefits including changes in insight, perspective, self-efficacy, emotions, and relationships.

Conclusion: Although people with neurodegenerative diseases and caregivers experience benefits from psychological interventions, burden-reducing adaptations and sensitive tailoring to the specific disease context is required to improve acceptability and engagement.

3.2 Introduction

Neurodegenerative disease is an umbrella term for diseases that are characterized by progressive degeneration of the structure and function of the nervous system. Common neurodegenerative diseases include Alzheimer's disease, Parkinson's disease, Motor Neurone Disease/Amyotrophic Lateral Sclerosis (MND/ALS), and Huntington's disease. Each of these diseases manifest and progress differently. However, all neurodegenerative diseases have some common features – they are progressive, disabling, and are a major threat to survival (Cummings & Pillai, 2016). The psychological impact can also be similar across the diseases because of certain common changes (e.g. decreasing functional status, changes in social roles, and experiencing multiple losses as the disease gets worse) (Ovaska-Stafford, Maltby, & Dale, 2021). People with neurodegenerative diseases experience significant psychological distress including anxiety and depression (Dale & van Duijn, 2015; Kurt et al., 2007; Reijnders, Ehrt, Weber, Aarsland, & Leentjens, 2008; Thompson et al., 2012). Caregivers also experience burden, depression, and anxiety (Aoun et al., 2013; Iavarone, Ziello, Pastore, Fasanaro, & Poderico, 2014; Martínez-Martín et al., 2007) which is sometimes related to the disease severity and neuropsychiatric symptoms experienced by the person with the neurodegenerative condition (Allegri et al., 2006; Martínez-Martín et al., 2007; Schrag, Hovris, Morley, Quinn, & Jahanshahi, 2006; Tramonti et al., 2019). Psychological distress can occur early or late in the disease trajectory and can have a negative impact on quality of life for both people with neurodegenerative diseases and caregivers (Baquero & Martín, 2015; Fischer, Ismail, & Schweizer, 2012).

Psychological interventions have the potential to help people with neurodegenerative diseases and caregivers reduce psychological distress and have a better quality of life. The evidence for the effectiveness of psychological interventions is mixed, especially across different neurodegenerative diseases. For example, there is some evidence to suggest that psychological interventions are effective for improving psychological well-being in Parkinson's disease and dementia (Orgeta, Qazi, Spector, & Orrell, 2015; Zarotti, Eccles, et al., 2020). In other neurodegenerative diseases (e.g. MND/ALS and Huntington's Disease) the literature is sparse and the need for more research on interventions has been emphasized (Gould et al., 2015; Thomas, Thomas, Hillier, Galvin, & Baker, 2006; Zarotti, Dale, Eccles, & Simpson, 2020; Zarotti et al., 2021). For caregiver interventions, there is more evidence for the effectiveness of psychological interventions for caregivers of people with dementia (Cheng, Au, Losada, Thompson, & Gallagher-Thompson, 2019; Liu, Chen, & Sun, 2017; Selwood, Johnston, Katona, Lyketsos, & Livingston, 2007). In other diseases like Parkinson's disease, Huntington's disease, and MND/ALS, there has been less research on interventions for caregivers' psychological support (Harris et al., 2018; Hempel, Norman, Golder, Aguiar-Ibáñez, & Eastwood, 2008). Psychological interventions may be of benefit for people with neurodegenerative diseases and caregivers, but currently evidence is limited or inconclusive.

Researchers and clinicians have identified specific barriers that can affect how patients and caregivers with neurodegenerative diseases might engage with psychological interventions. Approaches like Cognitive Behavioural Therapy (CBT) could be challenging because the progression of physical symptoms is generally irreversible and some negative cognitions about the situation are realistic rather than distorted (Cole & Vaughan, 2005; Hind et al., 2010). Similarly with caregivers, the seemingly unchangeable nature of their situation may make some CBT therapeutic goals difficult to achieve (Fowler et al., 2021). Approaches like acceptance and commitment therapy (ACT) have been proposed as more suitable and acceptable as they focus on accepting distress and finding new ways to carry out valued activities (Fowler et al., 2021; Graham et al., 2015).

The level of physical disability and cognitive impairment with neurodegenerative diseases may also affect engagement with psychological interventions. Physical disability can make accessing mental health interventions difficult because of issues with transportation (Dobkin et al., 2013; Marconi et al., 2016). Additionally, cognitive impairment is typical in Alzheimer's disease, but it can also present in Parkinson's disease, multiple sclerosis (MS) and MND/ALS. Cognitive changes can affect how people engage with therapy and interventions (Cole & Vaughan, 2005; Weeks et al., 2019). Caregivers typically spend several hours a day assisting the person with the neurodegenerative disease or organizing care and this can leave little time for other activities (Galvin et al., 2018; Zhang, Chang, Liu, Gao, & Porock, 2018).

Given ongoing uncertainty about the effectiveness of psychological approaches in many neurodegenerative conditions and the likelihood of significant health-related barriers to engagement, being open to understanding how people experience these interventions is vital. Qualitative research allows for the patient and caregivers' perspectives to be prioritised and explored in depth and detail. Whilst qualitative studies of patient and caregivers' experiences of various psychological interventions for a range of neurodegenerative diseases have been published, this literature has not yet been synthesised. We therefore, aimed to synthesise and review findings from qualitative research in order to obtain a detailed and comprehensive understanding of engagement and acceptability, identifying issues that are relevant to patients and caregivers with a range of neurodegenerative conditions, therapeutic approaches, and delivery modes. Bringing together these findings will offer important insights to guide those involved in planning, delivering, and evaluating interventions for patients with neurodegenerative diseases and their caregivers and help drive the development of more acceptable, engaging, and effective interventions.

3.3 Methods

We conducted a systematic review in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) statement (Sohrabi et al., 2021) and analysed data using a thematic synthesis (Thomas & Harden, 2008). The protocol was prospectively registered on PROPSPERO (CRD42021223125). We followed the ENTREQ (enhanced transparency in reporting the synthesis of qualitative research) checklist to facilitate systematic reporting (Tong et al., 2012).

3.3.1 Search strategy

A systematic search was carried out in six electronic databases (MEDLINE, PsycINFO, CINAHL Plus, Web of Science, AMED and the Cochrane library). The searches were conducted in October, 2020 and updated in October, 2021. Search terms were developed from initial literature scoping, consultation with a specialist librarian, and were piloted to ensure relevant studies were being included. The search strategy combined terms relating to psychological interventions, neurodegenerative diseases, and qualitative or mixed methods research (see Appendix C and Appendix D). Grey literature was also searched (OpenGrey and ProQuest), and references of included studies were hand-searched. No date limits were applied, results were restricted to those published in English.

3.3.2 Inclusion and Exclusion criteria

The detailed inclusion and exclusion criteria are shown in Table 2. We sought to identify qualitative and mixed methods studies that had a substantial qualitative component that investigated patient and caregivers’ experiences and views of using psychological interventions. We included neurodegenerative diseases such as Alzheimer’s disease, Huntington’s disease, Parkinson’s disease, Motor neurone disease/Amyotrophic Lateral Sclerosis (MND/ALS), Multiple system atrophy, and multiple sclerosis (MS). We defined psychological interventions as interventions that had psychotherapeutic or psychoeducational content and where the focus/aim was to reduce emotional distress or improve wellbeing or coping skills.

Table 2

Review inclusion and exclusion criteria

Criteria	Inclusion	Exclusion
Population	Adults who have a neurodegenerative disease (Alzheimer's disease, Huntington's disease, Parkinson's disease, Motor neuron disease/ Amyotrophic lateral sclerosis, Multiple system atrophy, Multiple sclerosis) Both people who have neurodegenerative diseases and their family caregivers.	Children or young adults with neurodegenerative diseases Adults with acute, non-progressive neurological illness (e.g. stroke, brain injury) Mixed population of older adults, some with dementia or mild cognitive impairment and some without. Professional health or care perspectives
Intervention	Psychotherapeutic or psychoeducational interventions aimed at managing the emotional impact of neurodegenerative diseases/aimed at reducing distress or improving psychological wellbeing. Interventions can be in any format or mode of delivery Evaluations of psychological support services that aim to improve emotional wellbeing (e.g. counselling service)	Interventions designed to improve management of physical symptoms cognitive impairments, and behaviour difficulties. Multidisciplinary or multicomponent interventions that do not have a substantial psychotherapeutic or educational component about coping with stress and difficult emotions. Interventions that only provide social support, or that use creative therapies (e.g. art, dance, music) or adaptations to the environment.
Outcome	Experiences and views of using psychological interventions, reported from the patient and/or family caregiver perspective.	
Study design	Qualitative studies and mixed methods studies with a	Studies with quantitative data only

	substantial qualitative component	Systematic and other literature reviews
	Qualitative data and reporting that includes adequate depth and detail of methods and findings to be able to interpret meaning about the experience of using psychological interventions.	Open-ended questionnaire data, or brief researcher commentary on the acceptability and feasibility of interventions.
Types of publication	Primary research including peer-reviewed articles, theses, and dissertations.	Commentaries, book chapters, editorials, clinical guidelines, or recommendations.

3.3.3 Screening and selection

The search results were exported to EndNote and duplicates were removed. All titles and abstracts were screened for eligibility by one author (CP). 15% were screened independently by a second reviewer (CM) and discrepancies were resolved through discussion and consensus. All full text screening was conducted by CP and CM independently, any conflicts were resolved through discussion, involving a third reviewer (LD) where necessary.

3.3.4 Data extraction and quality appraisal

The data was extracted into a table and included data about study characteristics, participants, and the interventions. All qualitative data (participant quotes and interpretations of findings) from the results and discussion sections were imported to Nvivo for analysis and synthesis.

All included studies were assessed for quality using the Critical Appraisal Skills Program (CASP) qualitative research checklist (Campbell et al., 2012; Mays & Pope, 2000). The CASP is a well-recognized checklist for assessing quality in qualitative studies and has been previously used to assess qualitative studies which seek to understand experiences of interventions in other disease groups (Cox et al., 2017; Maund et al., 2019). Details of the quality assessment for each included study can be found in Appendix E. We did not exclude low quality studies but took quality ratings into account when describing and interpreting the confidence in the findings.

3.3.5 Analysis and synthesis

Data was analysed using a thematic synthesis (Thomas & Harden, 2008). We selected this method because it allowed us to stay close to the original context of the primary studies, and draw conclusions about participants' experiences across a heterogeneous group of neurodegenerative diseases and intervention types through developing analytic themes which go beyond the interpretations presented in the primary studies. Extracted data was managed and coded using Nvivo software. An inductive line-by-line coding of the data was carried out for each primary study. The list of 'free' codes for each study was then systematically compared, similar codes were combined and grouped into descriptive themes, paying attention to and recording whether the theme was represented in interventions with patients, caregivers or both groups. The wider review team then discussed the descriptive themes in relation to the review question. The descriptive themes were compared with each other and diagrams were used to identify specific issues with engagement and acceptability. Abstract or analytic themes that went beyond the content of original studies were developed, the descriptive themes were examined in light of these analytic themes, and changes to the analytic themes were made iteratively. This process was repeated until the analytic themes sufficiently captured the descriptive themes and answered the review question. The analytic themes were further refined during the process of writing up the results.

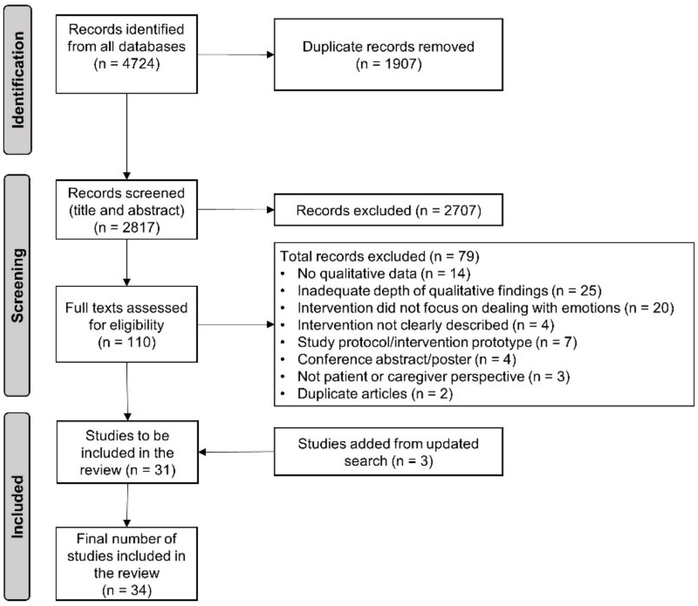
We used the GRADE CERQual approach (Lewin et al., 2018) as a systematic and transparent way to evaluate confidence in our principal review findings (i.e. our analytic themes). GRADE CERQual considers 4 components: methodological limitations, coherence, adequacy of data, and relevance; ratings were assigned through discussions between CP and LD.

3.4 Results

Figure 12 shows the studies identified and included and excluded at different stages of the screening and selection process. 34 papers met the inclusion criteria and were included in the review. The different neurodegenerative diseases included were dementia (n=19), MS (n=9), Parkinson's disease (n=3), MND/ALS (n=2), and Huntington's disease (n=1). 13 papers included people with neurodegenerative diseases as participants, 14 included caregivers as participants, and 7 papers included both people with neurodegenerative diseases and caregivers together. The different types of interventions covered were mindfulness (n=15), ACT (n=6), multicomponent psychoeducational interventions (n=6), CBT (n=5), dignity therapy (n=1), and compassion-focussed therapy (n=1). Further details about the included papers can be found in Table 3.

Figure 12

PRISMA diagram



3.4.1 Assessment of quality and confidence in review findings

Individual study quality ratings can be found in Appendix E. Overall, studies were of high quality, with papers scoring 7 or higher on a scale from 1-10 on the CASP checklist. Studies tended to be weaker on reporting the relationship between researchers and participants, and reporting data analysis methods sufficiently. Included studies scored well on clearly stating the aims of the research and appropriate use of qualitative methodology.

The GRADE CERQual evaluation found that in addition to the included studies being of high quality, a large number of studies contributed to the development of each analytic theme, the studies had rich data, and the themes were relevant across different diseases and intervention types (see Appendix F). The evaluation process therefore concluded that there was high confidence in three analytic themes and moderate confidence in one analytic theme. Overall, we are confident that the review findings are a reasonable representation of the experiences of psychological interventions among people with neurodegenerative diseases and caregivers.

3.4.2 Themes

We developed 4 analytic themes; Figure 13 outlines the descriptive and analytic themes and the relationship between them. These themes were common for people with neurodegenerative diseases and caregivers. Where specific differences occur between these two participant groups, they are discussed within each theme.

Figure 13

Analytic and descriptive themes

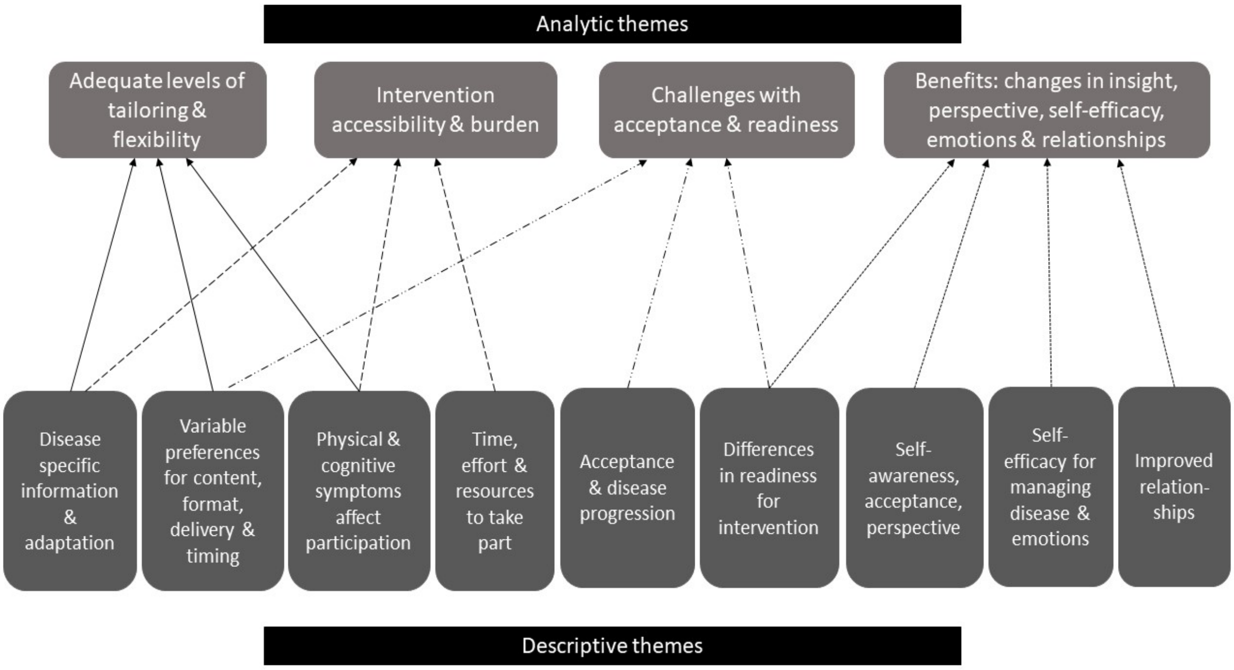


Table 3

Summary table of included studies

Author, year, location	Disease	Participants & (sample size)	Data collection and analysis	Intervention details (intervention type, delivery, duration)
(Adams, 2018) UK	Dementia	Patients and caregivers (6 – 3 patients and 3 caregivers)	Semi-structured interviews Thematic analysis and constant comparative method	Mindfulness based stress reduction Group, face-to-face 8 sessions, 2 hours each
(Berk et al., 2019) Netherlands	Dementia	Patients and caregivers (12 – 6 patients and 6 caregivers)	Semi-structured interviews Deductive content analysis	Mindfulness based stress reduction Group, face-to-face 8 sessions, 2.5 hours each

Author, year, location	Disease	Participants & (sample size)	Data collection and analysis	Intervention details (intervention type, delivery, duration)
(Bogosian, Hughes, Norton, Silber, & Moss-Morris, 2016) UK	Multiple sclerosis	Patients (15)	Semi-structured interviews Thematic analysis	Mindfulness based stress reduction Group, video conference 8 sessions, 1 hour each
(Bogosian et al., 2021) UK	Parkinson's disease	Patients (26)	Interviews Thematic analysis	Mindfulness based cognitive therapy Group, video conference 8 sessions, 1 hour each
(Craig, Hiskey, Royan, Poz, & Spector, 2018) UK	Dementia	Patients and caregivers (5)	Semi-structured interviews Thematic analysis	Compassion focused therapy Group, face-to-face 10 sessions, 1 hour each

Author, year, location	Disease	Participants & (sample size)	Data collection and analysis	Intervention details (intervention type, delivery, duration)
(de Wit, Vervoort, et al., 2019) Netherlands	MND/ALS	Caregivers (23)	Semi-structured interviews Thematic analysis	Psychoeducation based on ACT Individual, blended (face-to-face, online and telephone) 6 online modules completed in own time, 1 face-to-face & 1 phone session
(Dennison, Moss-Morris, Yardley, Kirby, & Chalder, 2013) UK	Multiple sclerosis	Patients (30)	In-depth interviews Thematic analysis with some grounded analytic techniques	Cognitive behaviour therapy Individual, blended (face-to-face and telephone) 8 sessions, 1 hour each
(Douglas et al., 2021) UK	Dementia	Patients and caregivers (14 – 8 patients and 6 caregivers)	Semi-structured interviews Thematic analysis	Mindfulness based cognitive therapy Group, face-to-face 8 sessions, 1.5 hours each

Author, year, location	**Disease**	**Participants & (sample size)**	**Data collection and analysis**	**Intervention details (intervention type, delivery, duration)**
(Eccles et al., 2020) UK	Huntington's disease	Patients (11)	Interviews Framework analysis	Mindfulness based cognitive therapy Group, face-to-face 8 sessions, 2 hours each
(Fitzpatrick, Simpson, & Smith, 2010) UK	Parkinson's disease	Patients (12)	Semi-structured interviews Interpretative Phenomenological Analysis	Mindfulness based cognitive therapy Group, face-to-face 8 sessions, 2.5 hours each
(Giovannetti et al., 2020) Italy	Multiple sclerosis	Patients (30)	Interviews Content analysis	Acceptance and commitment therapy Group, face-to-face 8 sessions, 2.5 hours each

Author, year, location	Disease	Participants & (sample size)	Data collection and analysis	Intervention details (intervention type, delivery, duration)
(Glueckauf et al., 2012) USA	Dementia	Caregivers (10)	Semi-structured interviews Open coding procedure and categorization into themes	Cognitive Behavioural therapy Group and individual, blended (face-to-face and telephone) 12 sessions, 1 hour each
(Gottberg et al., 2016) Sweden	Multiple sclerosis	Patients (12)	Semi-structured interviews Content analysis	Cognitive Behavioural Therapy Individual, face-to-face 15-20 sessions, 50 mins each
(Han, Yuen, Jenkins, & Yun Lee, 2021) USA	Dementia	Caregivers (7)	Semi-structured interviews Interpretive Phenomenological Analysis	Acceptance and commitment therapy Individual, video conference 10 sessions, 1 hour each

Author, year, location	**Disease**	**Participants & (sample size)**	**Data collection and analysis**	**Intervention details (intervention type, delivery, duration)**
(Hind et al., 2010) UK	Multiple sclerosis	Patients (17)	In-depth interviews Framework analysis	Cognitive Behavioural Therapy Individual, computer/online 5-8 weekly sessions, completed in own time
(Hoppes, Bryce, Hellman, & Finlay, 2012) USA	Dementia	Caregivers (11)	Semi-structured interviews Thematic analysis with grounded theory open coding	Mindfulness based stress reduction Group, face-to-face 4 sessions, 1 hour each
(Johannessen, Bruvik, & Hauge, 2015) Norway	Dementia	Caregivers (20)	Interviews Content analysis	Multicomponent psychoeducational Group and individual, face-to-face 15 sessions (2 half day seminars, 5 hour long counselling sessions and 8 group meetings)

Author, year, location	Disease	Participants & (sample size)	Data collection and analysis	Intervention details (intervention type, delivery, duration)
(Johnston et al., 2016) UK	Dementia	Both patients and caregivers (27)	Interviews and focus group Framework analysis	Dignity therapy Individual, face-to-face Not mentioned, time spent to undertake interview with person and then edit the dignity document.
(Kazmer, Glueckauf, Schettini, Ma, & Silva, 2018) USA	Dementia	Caregivers (7)	Semi-structured interviews Open coding, axial coding, categorization via concept mapping	Cognitive Behavioural therapy and spiritual counselling Individual, face-to-face 12 sessions, 1 hour each
(Larochette, Wawrziczny, Papo, Pasquier, & Antoine, 2020) France	Dementia	Caregivers (23)	Semi-structured interviews and questionnaire Thematic analysis	Acceptance and commitment therapy Individual, face-to-face 7 sessions, 90 mins each

Author, year, location	Disease	Participants & (sample size)	Data collection and analysis	Intervention details (intervention type, delivery, duration)
(Lavoie et al., 2005) Canada	Dementia	Caregivers (30)	Semi-structured interviews Content analysis	Psychoeducation Group, face-to-face 15 sessions, 2 hours each
(Marconi et al., 2016) Italy	MND/ALS	Both patients and caregivers (44 – 26 patients, 18 caregivers)	Semi-structured interviews Grounded theory approach to analysis	Mindfulness based stress reduction Group, face-to-face 8 sessions, (duration not mentioned)
(Martin et al., 2015) UK	Dementia	Patients (6)	Focus groups and interviews Thematic analysis	Psychoeducation Group, face-to-face 6 sessions, 2.5 hours each

Author, year, location	**Disease**	**Participants & (sample size)**	**Data collection and analysis**	**Intervention details (intervention type, delivery, duration)**
(Nehrig & Chen, 2019a) USA	Dementia	Caregivers (15)	Semi-structured interviews Thematic analysis	Psychoeducation Individual, face-to-face 12 sessions, duration not mentioned
(Nehrig, Gillooly, Abraham, Shifrin, & Chen, 2019b) USA	Dementia	Caregivers (14)	Semi-structured interviews Thematic analysis	Psychoeducation Individual, face-to-face 12 sessions, duration not mentioned
(Nehrig, Shifrin, Abraham, & Chen, 2019c) USA	Dementia	Caregivers (22)	Semi-structured interviews Thematic analysis	Psychoeducation Individual, face-to-face 12 sessions, duration not mentioned

Author, year, location	Disease	Participants & (sample size)	Data collection and analysis	Intervention details (intervention type, delivery, duration)
(Pegler, 2017) UK	Dementia	Caregivers (8)	Feedback surveys and focus group Framework analysis	Acceptance and commitment therapy Group, face-to-face 3 sessions, 2.5 hours each
(Potter, Golijana-Moghaddam, Evangelou, Mhizha-Murira, & Das Nair, 2021) UK	Multiple sclerosis	Caregivers (7)	Semi-structured interviews Framework analysis	Acceptance and commitment therapy Individual, remote (self help text and telephone) 8 sessions, duration not mentioned
(Sessanna et al., 2021) USA	Multiple sclerosis	Patients (14)	Focus group Thematic analysis	Mindfulness based stress reduction Group, blended (face-to-face and online) 8 sessions, 1 hour each

Author, year, location	**Disease**	**Participants & (sample size)**	**Data collection and analysis**	**Intervention details (intervention type, delivery, duration)**
(Simpson, Byrne, Wood, Mair, & Mercer, 2018) UK	Multiple sclerosis	Patients (33)	Semi-structured interviews Thematic analysis and framework approach	Mindfulness based stress reduction Group, face-to-face 8 sessions, duration not mentioned
(Simpson, Simpson, Wood, Mercer, & Mair, 2019) UK	Multiple sclerosis	Patients (33)	Semi-structured interviews Deductively using normalisation process theory constructs	Mindfulness based stress reduction Group, face-to-face 8 sessions, duration not mentioned
(Swannell, 2017) UK	Dementia	Both patients and caregivers (8 – 4 patients and 4 caregivers)	Semi-structured interviews Thematic analysis	Mindfulness based stress reduction Group, face-to-face 8 sessions, 1.5 hours each

Author, year, location	**Disease**	**Participants & (sample size)**	**Data collection and analysis**	**Intervention details (intervention type, delivery, duration)**
(Tahsin, Stanyon, Sun, & Gamble, 2021) Canada	Dementia	Caregivers (6)	Interviews Thematic analysis	Mindfulness Group, face-to-face 1 session, 15 mins
(Vandenberg et al., 2019) Australia	Parkinson's disease	Patients (16)	Semi-structured interviews Thematic analysis	Mindfulness based stress reduction Group, face-to-face 6 sessions, 2 hours each

3.4.2.1 Adequate levels of tailoring and flexibility

Engagement with psychological interventions was either facilitated or hindered by adequate intervention tailoring and flexibility. Disease-specific tailoring was often used to adapt the intervention content and activities to the specific disease or the specific needs of caregivers. People with neurodegenerative diseases and caregivers found disease-specific information and advice helpful (Adams, 2018c; Bogosian et al., 2021; de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Eccles et al., 2020; Gottberg et al., 2016; Hind et al., 2010; Kazmer et al., 2018; Nehrig, Shifrin, et al., 2019c; Simpson et al., 2018; Simpson et al., 2019) and discussed the importance of therapists or facilitators having disease-specific knowledge (Craig et al., 2018; de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Gottberg et al., 2016; Kazmer et al., 2018; Martin et al., 2015; Simpson et al., 2019). Caregivers also appreciated interventions that were targeted to their own unique needs and difficulties (de Wit, Vervoort, et al., 2019; Larochette et al., 2020; Nehrig, Shifrin, et al., 2019) and appreciated the ability to choose intervention modules that were relevant to their situation (de Wit, Vervoort, et al., 2019; Larochette et al., 2020).

> I don’t think you can have mixed abilities walking round and everybody happy... I just felt the delivery needed to be a bit more sensitive to the needs of people with MS and MS is very different from chronic fatigue or any other thing like that.. it might have been better to have somebody who has actually been ill or not been well to have an input into the delivery on the course’ [person with MS, mindfulness intervention, (Simpson et al., 2019)]

People with the same neurodegenerative disease can have different physical and cognitive symptoms and consequently, intervention activities and delivery formats need to be adjusted. People with neurodegenerative diseases stressed the importance of adapting interventions so that they were user-friendly for people with varying levels of ability and cognition (Adams, 2018; Martin et al., 2015; Sessanna et al., 2021; Simpson et al., 2018; Simpson et al., 2019; Swannell, 2017) and also went a step further to adapt the activities to their own individual routines and circumstances (Adams, 2018; Berk et al., 2019; Eccles et al., 2020; Larochette et al., 2020; Martin et al., 2015; Swannell, 2017; Vandenberg et al., 2019).

Similarly, intervention formats and delivery methods need to be flexible to accommodate people with different symptoms and needs or levels of ability. This could be through ensuring flexibility in intervention duration. Some participants preferred shorter intervention durations

(Berk et al., 2019; Giovannetti et al., 2020; Simpson et al., 2018; Simpson et al., 2019), whereas others wanted more time to enable learning, practising and group work (Berk et al., 2019; Bogosian et al., 2016; Bogosian et al., 2021; Craig et al., 2018; de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Eccles et al., 2020; Giovannetti et al., 2020; Larochette et al., 2020; Martin et al., 2015; Nehrig & Chen, 2019a; Nehrig, Gillooly, et al., 2019b; Nehrig, Shifrin, et al., 2019c; Pegler, 2017; Potter et al., 2021; Sessanna et al., 2021; Simpson et al., 2018; Simpson et al., 2019; Tahsin et al., 2021). Interventions also require some flexibility in terms of mode of delivery. Face-to-face options were valued because of the bond and rapport created between participants or between participants and therapists which made it easier to talk about difficult issues (de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Gottberg et al., 2016; Potter et al., 2021; Sessanna et al., 2021). However, face-to-face sessions were difficult if people had problems with mobility, transportation or had other conflicting commitments (Berk et al., 2019; Eccles et al., 2020; Gottberg et al., 2016; Marconi et al., 2016; Simpson et al., 2018). Accessing the intervention remotely (online or by telephone/at home) was convenient, easy to use, and could be completed at the individual's own pace (Bogosian et al., 2021; de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Hind et al., 2010; Johnston et al., 2016; Sessanna et al., 2021) but difficulties with concentration, technology, and the lack of human interaction could make engagement challenging (de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Hind et al., 2010; Sessanna et al., 2021).

> With MS you can become very isolated because of your disability ... So, I think when working with something that is a computer programme it makes you feel even more like you're not speaking to someone face to face. You don't get the empathy there. [person with MS, computerised CBT, (Hind et al., 2010)]

Being at different disease stages could also impact intervention preferences. Group settings generally facilitated intervention engagement (Adams, 2018; Berk et al., 2019; Bogosian et al., 2021; Johannessen et al., 2015; Lavoie et al., 2005; Pegler, 2017; Swannell, 2017); however, some participants found it difficult to engage with the group if they were not similar to other participants in terms of symptoms or disease stage (Berk et al., 2019; Bogosian et al., 2016; de Wit, Vervoort, et al., 2019; Eccles et al., 2020; Fitzpatrick et al., 2010; Giovannetti et al., 2020; Johannessen et al., 2015; Lavoie et al., 2005; Simpson et al., 2018; Simpson et al., 2019). Some preferred the intervention early on in their disease trajectory (Adams, 2018; de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Johannessen et al., 2015; Nehrig, Gillooly, et al., 2019b; Simpson et al., 2018), but others felt that the intervention was too early and they faced fewer problems that they needed help with (de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Gottberg et al., 2016; Hind et al., 2010; Pegler, 2017; Simpson et al., 2018).

> I think the program came at a good time. That everything is still relatively new for you, and can put your own things into perspective and that you receive support. Otherwise, you will keep going in circles for too long. [caregiver for person with MND/ALS, ACT intervention, (de Wit, Vervoort, et al., 2019)]

Tailoring and flexibility are also needed because of variability in coping preferences and preferences for therapy. Some participants preferred practical and structured support because it was more focused and suited people who had a more pragmatic approach to coping (de Wit, Vervoort, et al., 2019; Giovannetti et al., 2020; Gottberg et al., 2016; Johannessen et al., 2015; Kazmer et al., 2018; Lavoie et al., 2005; Martin et al., 2015; Nehrig, Gillooly, et al., 2019b; Nehrig, Shifrin, et al., 2019c). Others preferred opportunities to connect with the peers or a therapist, have open discussions, and access emotional support (Bogosian et al., 2016; de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Eccles et al., 2020; Johannessen et al., 2015; Nehrig & Chen, 2019a; Nehrig, Gillooly, et al., 2019b; Nehrig, Shifrin, et al., 2019c). There were also different preferences for working in patient-caregiver dyads and for including other family members in the intervention (Adams, 2018; Berk et al., 2019; Giovannetti et al., 2020; Johannessen et al., 2015; Johnston et al., 2016; Swannell, 2017).

3.4.2.2 Intervention accessibility and burden

Some psychological interventions or some components of the intervention were experienced as inaccessible or burdensome, and this could act as a barrier to engaging with the intervention. People with neurodegenerative diseases found it difficult to take part in and complete intervention activities because of physical and cognitive symptoms (Adams, 2018; Berk et al., 2019; Craig et al., 2018; Marconi et al., 2016; Martin et al., 2015; Sessanna et al., 2021; Swannell, 2017). Difficulties with cognition (in particular, concentration or memory difficulties) impacted their ability to sustain attention during the intervention, learn and practice new techniques/strategies (Adams, 2018; Berk et al., 2019; Craig et al., 2018; Douglas et al., 2021; Hind et al., 2010; Marconi et al., 2016; Martin et al., 2015; Swannell, 2017). Cognitive difficulties also made information processing difficult if information is too much or too technical (Adams, 2018; Berk et al., 2019; Martin et al., 2015); therefore, having adequate time and clear instructions when learning new activities was important (Douglas et al., 2021; Martin et al., 2015; Sessanna et al., 2021).

> Participants discussed the impact cognitive decline had on being able to practice MBSR (Mindfulness-based stress reduction). This varied from remembering to practice, to being unable to hold on to, or understand, the concept of mindfulness. Some participants felt it improved their concentration, while others felt it was not beneficial for people with dementia. [study author, dementia, mindfulness intervention, (Swannell, 2017)]

Physical symptoms can also make participation in intervention activities difficult (Berk et al., 2019; Dennison et al., 2013; Eccles et al., 2020; Gottberg et al., 2016; Hind et al., 2010; Marconi et al., 2016; Martin et al., 2015; Simpson et al., 2018; Simpson et al., 2019). For example, mindfulness activities such as mindful movement or mindful walking could sometimes prove unhelpful and insensitive to the participants who had difficulties with mobility (Simpson et al., 2018; Simpson et al., 2019). Symptoms such as pain, weakness and fatigue made it difficult to sit for long periods of time, to write, hold a phone for a long period of time, or lift a heavy manual (Dennison et al., 2013; Hind et al., 2010; Simpson et al., 2018; Simpson et al., 2019). Those with mobility issues or difficulties with balance and fatigue found it difficult to get to face-to-face sessions, and were frustrated by the lack of disability-friendly facilities (Berk et al., 2019; Eccles et al., 2020; Gottberg et al., 2016; Marconi et al., 2016; Martin et al., 2015; Simpson et al., 2018; Simpson et al., 2019).

> My brain just couldn’t cope with it because it was so busy just trying not to fall over ... and of course sometimes there’s no feeling in your feet ... so no, that was very challenging. [person with MS, mindfulness intervention, (Simpson et al., 2018)]

Additionally, the time and effort involved in taking part in a psychological intervention could sometimes be burdensome. Both caregivers and people with neurodegenerative diseases found it difficult to carve out time to participate due to other conflicting commitments and caring responsibilities (Adams, 2018; Bogosian et al., 2021; de Wit, Vervoort, et al., 2019; Douglas et al., 2021; Eccles et al., 2020; Hind et al., 2010; Pegler, 2017; Potter et al., 2021; Simpson et al., 2018; Simpson et al., 2019; Swannell, 2017). Interventions were sometimes viewed as effortful or demanding (Bogosian et al., 2016; Craig et al., 2018; Dennison et al., 2013; Gottberg et al., 2016; Hind et al., 2010; Nehrig, Shifrin, et al., 2019c; Potter et al., 2021; Swannell, 2017). Activities such as thought restructuring, goal setting, or mindfulness, that brought up difficult emotions or thoughts about physical impairment/disability, were difficult and some felt they could even be detrimental (Bogosian et al., 2016; Dennison et al., 2013; Gottberg et al., 2016; Hind et al., 2010; Swannell, 2017).

> The participants became aware that CBT is essentially a continuous process, with no pause between the sessions, but they also became aware that they did not always have enough time and energy in their everyday lives to focus on changing themselves. [study author, MS, CBT intervention, (Gottberg et al., 2016)]

Both patients and caregivers expressed that committed practice and time were required to apply what they had learned in the intervention to their own lives (Adams, 2018; Berk et al., 2019; Craig et al., 2018; Douglas et al., 2021; Giovannetti et al., 2020; Nehrig, Gillooly, et al., 2019b; Nehrig, Shifrin, et al., 2019c; Simpson et al., 2018; Simpson et al., 2019; Swannell, 2017) and it was sometimes difficult to find the time and energy to practise intervention strategies (particularly completing homework) or to find time for self-care (Adams, 2018; Berk et al., 2019; de Wit, Vervoort, et al., 2019; Douglas et al., 2021; Gottberg et al., 2016; Hind et al., 2010; Kazmer et al., 2018; Potter et al., 2021; Simpson et al., 2018; Swannell, 2017). Having support from work or family gave people more time and facilitated their participation in interventions (Douglas et al., 2021; Gottberg et al., 2016; Nehrig, Shifrin, et al., 2019c).

> Participants requested further flexibility to adapt the spacing of the reading they had to complete each week to maximise their ability to engage with the text in the light of their caring responsibilities, which sometimes changed dramatically week to week. [study author, MS, ACT intervention, (Potter et al., 2021)]

3.4.2.3 Challenges with acceptance and readiness

With neurodegenerative diseases, acceptance of the diagnosis and progressive symptoms can happen gradually and at different stages, and this influences how willing and how well participants can engage with interventions. Participants described how accepting the diagnosis and thinking about the future and symptom progression is difficult and anxiety provoking (Bogosian et al., 2016; de Wit, Vervoort, et al., 2019; Douglas et al., 2021; Fitzpatrick et al., 2010; Gottberg et al., 2016; Johannessen et al., 2015; Johnston et al., 2016; Larochette et al., 2020; Nehrig, Gillooly, et al., 2019b; Nehrig, Shifrin, et al., 2019c; Vandenberg et al., 2019). Intervention activities that triggered thoughts about the future and progression could also be challenging. For example, participants reported being anxious about meeting people with same condition or finding out about other people’s experiences, as this may show them what might happen to them in the future (Berk et al., 2019; Bogosian et al., 2016; de Wit, Vervoort, et al., 2019; Eccles et al., 2020; Fitzpatrick et al., 2010; Giovannetti et al., 2020; Simpson et al., 2018).

> Everybody seemed to be worse than me and I thought ‘I don’t think this is going to work for me’ I’ve said before, ‘if I don’t see it I haven’t got it’. There was one lady who said ‘come and join us at the Parkinson’s society’ and I thought ‘I’m not ready for that’. [person with Parkinson’s disease, mindfulness intervention, (Fitzpatrick et al., 2010)]

Similarly, some caregivers found interventions that discussed end of life issues and aggressive or disruptive behaviours in the case of dementia difficult and tried to avoid that section of the intervention (de Wit, Vervoort, et al., 2019; Johannessen et al., 2015). Activities like mindfulness can trigger difficult thoughts about one’s own disability or impairment (Johnston et al., 2016; Martin et al., 2015; Simpson et al., 2018; Simpson et al., 2019). Some interventions tried to avoid provoking distress by framing the intervention activities in more positive and less confronting ways (Johnston et al., 2016; Martin et al., 2015). For example, a “memory box” activity was presented as a place to put happy memories and a goal setting activity was presented as a way to focus on doing positive things in the future, and participants in this particular intervention found these two activities useful (Martin et al., 2015). Even though some intervention components such as group support or mindfulness were described as challenging, participants also reported that they were useful in increasing their acceptance of the disease and encouraged them to be more open about their situation (Bogosian et al., 2016; Fitzpatrick et al., 2010; Johannessen et al., 2015; Lavoie et al., 2005; Pegler, 2017; Vandenberg et al., 2019).

> The ‘memory box’ activity, which was demonstrated by the course champion, was something some participants already did. Others felt worried this was linked to thinking about death. The tutors reemphasized that the focus of the activity was on having a place where photographs or items relating to happy memories are kept and used as a resource to enjoy. Additionally, the activities around increasing happiness and sharing successes were seen positively as they both drew attention to previous achievements and current strengths. [study author, dementia, psychoeducation intervention, (Martin et al., 2015)]

Being aware of the impact of the disease can be a necessary step to engaging with psychological interventions. Participants who were struggling to cope with the disease, felt a stronger need for the intervention (Giovannetti et al., 2020; Gottberg et al., 2016; Kazmer et al., 2018; Simpson et al., 2019; Tahsin et al., 2021). Those who did not currently experience too many problems as a result of the disease, did not feel the need for psychological support (de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Gottberg et al., 2016; Hind et al., 2010; Pegler, 2017; Simpson et al., 2018). This was particularly true for people at the early stages of disease, but there were some exceptions where the intervention at an early stage helped prepare participants for future challenges (Adams, 2018; de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Simpson et al., 2018). Some participants at a more advanced stage also felt that they would have benefitted from accessing the intervention earlier on (de Wit, Vervoort, et al., 2019; Han et al., 2021; Johannessen et al., 2015; Nehrig, Gillooly, et al., 2019). Some suggested introducing the intervention early and then re-examining content as they encountered specific difficulties (de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Eccles et al., 2020; Han et al., 2021; Johnston et al., 2016).

> Those 6 weeks, they really helped to sort things out again. But you gradually notice that you start to forget things. Things change so much with ALS. When I look at how I experienced it in the beginning and in the final phase, it is so different. So I would like to give it as advice to offer the program several times. It simply helps you to make conscious choices. [caregiver for person with MND, ACT intervention, (de Wit, Vervoort, et al., 2019)]

It was also important to be open-minded, willing to try something new, and be willing to address difficult thoughts and feelings (Bogosian et al., 2021; Dennison et al., 2013; Giovannetti et al., 2020; Gottberg et al., 2016; Larochette et al., 2020; Nehrig, Shifrin, et al., 2019c; Pegler, 2017; Sessanna et al., 2021). Although important, this can be difficult to do and some feared that addressing difficult situations and emotions might make them unhappy (Berk et al., 2019; Bogosian et al., 2016; Larochette et al., 2020). There was some scepticism about the benefit of a psychological intervention when there was little to do to change the course of the disease and participants felt that this needed to be explained more clearly (Adams, 2018; Berk et al., 2019; Douglas et al., 2021; Eccles et al., 2020; Giovannetti et al., 2020; Gottberg et al., 2016; Martin et al., 2015; Simpson et al., 2019).

> One factor that appeared to influence whether a participant benefited from and was satisfied with their therapy experience was the degree to which they related to and engaged with the approach. Participants' identified this as 'buying into' therapy and described being open-minded, willing to open up to the therapist and give the approach a fair chance as important. [study author, MS, CBT intervention, (Dennison et al., 2013)]

Introductory sessions can be useful for preparing participants for the intervention and setting expectations about the time, commitment, likely benefits, and doing intervention activities in the context of disability/impairment (Bogosian et al., 2021; de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Douglas et al., 2021; Gottberg et al., 2016; Johannessen et al., 2015; Martin et al., 2015; Simpson et al., 2018; Simpson et al., 2019).

3.4.2.4 Benefits: changes in insight, perspective, self-efficacy, emotions, and relationships

People with neurodegenerative diseases and caregivers reported a number of benefits from psychological interventions. The interventions helped participants become more open and accepting of the impact of the disease (Adams, 2018; Berk et al., 2019; Bogosian et al., 2016; Bogosian et al., 2021; Craig et al., 2018; de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Douglas et al., 2021; Fitzpatrick et al., 2010; Giovannetti et al., 2020; Gottberg et al., 2016; Hoppes et al., 2012; Johannessen et al., 2015; Kazmer et al., 2018; Larochette et al., 2020; Lavoie et al., 2005; Nehrig, Shifrin, et al., 2019c; Pegler, 2017; Simpson et al., 2018; Simpson et al., 2019; Swannell, 2017; Vandenberg et al., 2019). They were more self-aware, particularly of their thoughts and how this impacted their feelings and responses to situations (Berk et al., 2019; Bogosian et al., 2016; Dennison et al., 2013; Douglas et al., 2021; Eccles et al., 2020; Fitzpatrick et al., 2010; Giovannetti et al., 2020; Gottberg et al., 2016; Han et al., 2021; Swannell, 2017; Vandenberg et al., 2019). This awareness or insight was key to changing their perspective on the disease and their situation. They thought about their disease or situation differently, and considered other perspectives (Adams, 2018; Bogosian et al., 2016; Bogosian et al., 2021; de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Giovannetti et al., 2020; Gottberg et al., 2016; Hoppes et al., 2012; Johnston et al., 2016; Larochette et al., 2020; Marconi et al., 2016; Nehrig, Gillooly, et al., 2019b; Nehrig, Shifrin, et al., 2019c; Vandenberg et al., 2019). This perspective taking was an important part of coping especially when there was no cure or treatment. Participants developed a more self-compassionate and positive approach to coping with neurodegenerative diseases. They placed more value on themselves and carved time out for self-care (Berk et al., 2019; Bogosian et al., 2016; Craig et al., 2018; de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Douglas et al., 2021; Giovannetti et al., 2020; Glueckauf et al., 2012; Han et al., 2021; Kazmer et al., 2018; Larochette et al., 2020; Lavoie et al., 2005; Nehrig, Gillooly, et al., 2019b; Nehrig, Shifrin, et al., 2019c; Pegler, 2017), were less critical or kinder to themselves (Adams, 2018; Craig et al., 2018; Dennison et al., 2013; Douglas et al., 2021; Han et al., 2021; Simpson et al., 2018; Simpson et al., 2019; Swannell, 2017), and focused on the things they could still do, appreciated things more, and overall developed a more positive outlook (Bogosian et al., 2016; Dennison et al., 2013; Douglas et al., 2021; Hoppes et al., 2012; Martin et al., 2015; Pegler, 2017; Sessanna et al., 2021; Simpson et al., 2018).

> Yes, I'd say I'm more at ease with my situation. I can accept things more the way they are and think it's OK. [person with MS, CBT intervention, (Gottberg et al., 2016)]

> I think I remind myself now... that I'm actually doing a good job. Looking after my mum and everything else I'm dealing with…and sometimes I forget and I take a step back and I think "damn, you've been through a lot." So cut yourself some slack... [caregiver for person with dementia, compassion-focused therapy, (Craig et al., 2018)]

Participants felt more in control of their lives, more confident about caring tasks, and expressed that they were 'coping better'. They valued learning skills and strategies and having the tools to manage difficult thoughts and feelings when they came up (Adams, 2018; Berk et al., 2019; Bogosian et al., 2016; de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Douglas et al., 2021; Fitzpatrick et al., 2010; Giovannetti et al., 2020; Glueckauf et al., 2012; Gottberg et al., 2016; Han et al., 2021; Hoppes et al., 2012; Kazmer et al., 2018; Larochette et al., 2020; Lavoie et al., 2005; Marconi et al., 2016; Martin et al., 2015; Nehrig, Gillooly, et al., 2019b; Nehrig, Shifrin, et al., 2019c; Potter et al., 2021; Sessanna et al., 2021; Simpson et al., 2019; Swannell, 2017; Tahsin et al., 2021; Vandenberg et al., 2019). This included being able to give negative thoughts less attention (Berk et al., 2019; Bogosian et al., 2016; Fitzpatrick et al., 2010; Giovannetti et al., 2020; Han et al., 2021; Marconi et al., 2016; Vandenberg et al., 2019) and being able to choose how they respond to situations (Bogosian et al., 2016; Dennison et al., 2013; Douglas et al., 2021; Fitzpatrick et al., 2010; Han et al., 2021; Hoppes et al., 2012; Marconi et al., 2016; Simpson et al., 2018; Swannell, 2017; Vandenberg et al., 2019). Some also reported being better able to deal with certain physical symptoms like fatigue, pain, tremors (Bogosian et al., 2021; Dennison et al., 2013; Fitzpatrick et al., 2010; Gottberg et al., 2016; Simpson et al., 2018) and improved sleep (Bogosian et al., 2021; Giovannetti et al., 2020; Gottberg et al., 2016; Marconi et al., 2016; Sessanna et al., 2021; Simpson et al., 2018; Tahsin et al., 2021).

> Through learning to be mindful, participants gained freedom to choose how they wished to respond to situations. This choice was empowering as people gained control over themselves and their responses. [study author, MS, mindfulness intervention, (Bogosian et al., 2016)]

People with neurodegenerative diseases felt confident about dealing with disease-related challenges (Adams, 2018; Dennison et al., 2013; Giovannetti et al., 2020; Gottberg et al., 2016; Johnston et al., 2016; Sessanna et al., 2021; Vandenberg et al., 2019) and caregivers felt confident about problem-solving and preparing for future challenges (de Wit, Vervoort, et al., 2019; Glueckauf et al., 2012; Han et al., 2021; Johannessen et al., 2015; Johnston et al., 2016; Kazmer et al., 2018; Larochette et al., 2020; Nehrig, Gillooly, et al., 2019b; Nehrig, Shifrin, et al., 2019c; Pegler, 2017). Caregiver participants recognized when they needed help and felt able to find or ask for help in these situations (de Wit, Vervoort, et al., 2019; Glueckauf et al., 2012; Han et al., 2021; Johannessen et al., 2015; Kazmer et al., 2018; Larochette et al., 2020; Lavoie et al., 2005; Nehrig, Gillooly, et al., 2019b; Nehrig, Shifrin, et al., 2019c; Swannell, 2017). Participants were able to identify what was important or meaningful to them and pursue these goals or activities (de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Giovannetti et al., 2020; Han et al., 2021; Martin et al., 2015).

> The findings show that the intervention contributed to making the caregivers feeling safer, giving them a better understanding of the disorder. Also, the intervention helped them to be more open about their situation and more prepared for the future. [study author, dementia, psychoeducation intervention, (Johannessen et al., 2015)]

Other benefits included feeling acknowledged and understood (Craig et al., 2018; de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Douglas et al., 2021; Giovannetti et al., 2020; Kazmer et al., 2018; Lavoie et al., 2005; Marconi et al., 2016; Martin et al., 2015; Potter et al., 2021), feeling calm and less stressed or anxious (Berk et al., 2019; Bogosian et al., 2021; Craig et al., 2018; Douglas et al., 2021; Fitzpatrick et al., 2010; Giovannetti et al., 2020; Hoppes et al., 2012; Marconi et al., 2016; Nehrig, Shifrin, et al., 2019c; Potter et al., 2021; Simpson et al., 2018; Tahsin et al., 2021), positive effects on self-esteem (Giovannetti et al., 2020; Martin et al., 2015), reduced caregiver burden and feelings of guilt (Han et al., 2021; Hoppes et al., 2012; Johannessen et al., 2015; Lavoie et al., 2005).

Some reported an improvement in their relationships, where there was more understanding between the person with the neurodegenerative condition and family (de Wit, Vervoort, et al., 2019; Douglas et al., 2021; Hoppes et al., 2012; Kazmer et al., 2018; Larochette et al., 2020; Nehrig, Shifrin, et al., 2019c; Vandenberg et al., 2019), less conflict (Berk et al., 2019; Giovannetti et al., 2020; Han et al., 2021; Hoppes et al., 2012; Kazmer et al., 2018; Larochette et al., 2020; Lavoie et al., 2005; Nehrig, Gillooly, et al., 2019b; Nehrig, Shifrin, et al., 2019c; Pegler, 2017; Simpson et al., 2018), less isolation (Berk et al., 2019; Douglas et al., 2021; Swannell, 2017), more connection and communication (Adams, 2018; Berk et al., 2019; Craig et al., 2018; Larochette et al., 2020; Lavoie et al., 2005; Marconi et al., 2016; Swannell, 2017), more openness (Bogosian et al., 2021; de Wit, Vervoort, et al., 2019; Giovannetti et al., 2020; Sessanna et al., 2021; Vandenberg et al., 2019), and overall better interactions with others (Berk et al., 2019; Giovannetti et al., 2020; Hoppes et al., 2012; Simpson et al., 2018; Vandenberg et al., 2019). Participants felt more confident with social interactions, and did not avoid social situations (Dennison et al., 2013; Fitzpatrick et al., 2010; Giovannetti et al., 2020; Johannessen et al., 2015; Vandenberg et al., 2019).

> Some participants reported that the training had influenced their relationship as a couple. Caregivers were better able to prevent or deal with quarrels. They felt more connected. [study author, dementia, mindfulness intervention, (Berk et al., 2019)]

Although majority of the studies reported intervention benefits, few participants in some studies said that they did not notice any changes post-intervention (Berk et al., 2019; Craig et al., 2018; Dennison et al., 2013; Douglas et al., 2021; Potter et al., 2021).

> A minority of participants reported little or no change from therapy. This tended to be more common for the SL (supportive listening – control group) participants and those who disagreed with the logic of their intervention. Other participants felt that they were already coping very well with MS, and so found they had little

to gain from therapy. [study author, MS, CBT intervention, (Dennison et al., 2013)]

3.5 Discussion

We synthesised the experiences of people with neurodegenerative diseases and caregivers and identified barriers and facilitators that are relevant to engagement and acceptability of psychological interventions. Many participants valued the post-intervention changes in insight, perspective, self-efficacy, emotions, and relationships. However, important things to consider include adequate levels of intervention tailoring and flexibility, intervention accessibility and burden, and challenges with acceptance and readiness among participants. The four themes presented in this review need to be considered in conjunction when developing or evaluating interventions, as they can interact with each other. For example, adequate tailoring might help reduce intervention burden; insight and perspective may be valued outcomes but may depend on acceptance or readiness from participants; or accessibility can be improved with dedicated tailoring strategies. Table 4 summarises key considerations when developing engaging and acceptable interventions for people with neurodegenerative diseases and caregivers based on the findings from our review.

With neurodegenerative diseases, it is important to tailor intervention content to the specific disease and adapt activities or advice so that people with various physical and cognitive symptoms can participate and feel that the intervention is designed for the problems and context they are facing. Disease-specific therapy manuals have been developed and used to improve psychological outcomes for patients and caregivers in some neurodegenerative diseases like Parkinson’s disease, dementia, and MS (Chan, Churcher Clarke, Royan, Stott, & Spector, 2017; Dobkin, Allen, & Menza, 2007; Moss-Morris et al., 2013). However, psychological support interventions need to move beyond a one-size-fits-all approach for specific disorders and personalise interventions based on individual and contextual factors (Purgato, Singh, Acarturk, & Cuijpers, 2021). Our findings show that additional tailoring and flexibility is required with interventions for neurodegenerative diseases because of the variability in symptoms, disease stages, coping preferences, and preferences for therapy. Individual differences in terms of levels of acceptance and readiness are also important considerations, particularly with neurodegenerative conditions, where thinking about disease progression can be distressing (Moss-Morris et al., 2013). People with neurodegenerative diseases can experience rapid changes in their medical and psychological condition, and therefore their needs within different phases in psychological interventions may also change. Although this level of personalisation of interventions is important, it may also be challenging to execute in very structured, manualised therapies or in interventions that are conducted in a group setting, compared to one-to-one interventions facilitated by trained professionals. Our review also found very few interventions that offered individuals a choice regarding the type of therapy and therapeutic activities. Having options for different kinds of psychological support could be a way forward to further personalise interventions by allowing people to choose content and activities that match their needs and preferences.

Recent reviews of the effectiveness of psychological interventions for wellbeing in neurodegenerative diseases noted that so far there is more evidence for certain therapeutic approaches (e.g. CBT and mindfulness) and less research on other therapeutic approaches (Zarotti, Eccles, et al., 2020; Zarotti et al., 2021). Our current synthesis of intervention experiences mirrors the types of interventions covered in these reviews with mindfulness-based interventions being most common. Noteworthy in our review is the use of therapeutic approaches that originate from clinical psychology, and few studies that use health psychology theory and frameworks. Health psychology approaches often have a focus on beliefs about the disease or symptoms, and cognitive and behavioural strategies in relation to illness-related threats (e.g. Leventhal's common sense model of self-regulation or Moss-Morris' model of adjustment to chronic illness) (Leventhal, Leventhal, & Contrada, 1998; Moss-Morris, 2013). Our review showed that some beliefs about the illness, particularly how people thought about future disease progression and the level of acceptance of the impact of the illness on their life can also play a role in how people engage with interventions. Integrating theoretical frameworks from health psychology and psychological therapy models can potentially improve engagement and make intervention programmes more effective (Karekla, Karademas, & Gloster, 2019). The included studies were typically conducted as intervention evaluation studies and focused on participants' experiences of the different intervention components and how the intervention may have helped them. Therefore, the review themes are limited to these aspects and do not cover how participants' experiences relate to psychological frameworks or models of coping and adjustment.

There also needs to be greater flexibility with how psychological interventions are delivered for people with neurodegenerative diseases and caregivers. Therapists working with long-term conditions have expressed a need for a model of therapy that has greater flexibility in terms of session frequency, attendance policies, mode of delivery, and session location because of issues with mobility, unpredictable symptoms, time involved, and fatigue (Carroll, Moss-Morris, Hulme, & Hudson, 2021). Additionally, physical and cognitive symptoms can make engaging with therapy difficult or burdensome for people with neurodegenerative diseases (Dennison & Moss-Morris, 2010; Dobkin et al., 2013; Van Groenestijn et al., 2015). With psychological interventions, the structure and timing of sessions, and aspects such as homework can act as barriers to starting or continuing therapy (Barnes et al., 2013; Rice et al., 2020). Being flexible with the structure of psychological services or providing necessary resources/support can help reduce this treatment burden (May et al., 2014; Rice et al., 2020). Carefully considering a biopsychosocial framework when developing interventions (biological aspects – e.g. changes in symptoms, psychological aspects – e.g. level of acceptance, social aspects – e.g. support from services or family) could also guide interventions to be more flexible in terms of their delivery.

Based on our review findings, we have made some recommendations for future research. Going forward researchers need to focus on issues with engagement and acceptability when developing interventions for people with neurodegenerative diseases and caregivers. User needs should be evaluated using qualitative methods by adopting a more user-centred or co-production approach. There is a gap in our understanding of how engagement differs with different types of psychological interventions and with different methods of intervention delivery, and how different levels of engagement can impact outcomes. This needs to be explored in future research. We also recommend more detailed and consistent reporting of intervention development, content, and delivery, including the steps taken to adapt or tailor the intervention to specific populations as this can impact the acceptability and engagement with the intervention. Our review highlighted the range of benefits people with neurodegenerative diseases and caregivers experience from psychological interventions, which go beyond changes usually anticipated in trials such as improvements in mood or well-being. It would be useful to measure these broader changes (e.g. changes in self-awareness, thinking and attitudes, self-efficacy) quantitatively as outcomes of intervention trials (Berk et al., 2019; Eccles et al., 2020; Meek et al., 2021; Nehrig, Gillooly, et al., 2019; Tang & Chan, 2016) as well as undertake research to determine what outcomes are valued or appropriate to use as indicators of therapeutic success, especially in progressive diseases where symptoms will get worse with time.

Table 4

Key considerations when developing psychological interventions for people with neurodegenerative diseases and caregivers

Key things to consider	
Intervention personalisation	It is important not to treat people with a particular neurodegenerative disease or their caregivers as a homogenous group as there can be differences in symptoms experienced, coping styles, levels of readiness to engage with interventions, and preferences for therapy. Interventions could be personalised by asking people about their needs and preferences and then tailoring interventions, or allowing individuals to self-tailor the intervention to meet their needs (e.g. choosing or prioritising from a choice of content and activities).
Intervention accessibility and burden	Intervention content and activities need to be accessible to people with different physical and cognitive symptoms. This includes thinking about and improving the accessibility of intervention materials, session durations, homework tasks, different modes of intervention delivery. To make interventions less burdensome, intervention durations can be minimised, some activities can be optional, and information and suggestions need to be easy to integrate into users' daily routine.
Intervention flexibility and responsiveness	As symptoms change users' needs may also change, and interventions need to take this into account and respond to these new needs. Offering support at different points in the disease trajectory allows for people with neurodegenerative diseases and caregivers to take up the offer when they feel ready or as symptoms and needs change.
Introduction and setting expectations	Familiarity with and understanding the benefits of psychological therapy can improve engagement (Liu & Gellatly, 2021; Theed, Eccles, & Simpson, 2018), and explaining this through introductory sessions or intervention promotion materials or testimonies from peers may help engage users and alleviate anxieties in individuals who are unsure of what therapy would entail or do not feel ready to take part. Explaining the time and commitment involved with taking part in the intervention can also help users' understand what to expect from the intervention.
Focus on engagement and acceptability when developing and evaluating interventions	It is important to examine how users engage with the intervention and be open to exploring issues with acceptability. Adopting user-centred or co-production approaches when designing interventions for people with neurodegenerative diseases and caregivers may be useful for anticipating potential barriers to engagement and evaluating the acceptability of intervention adaptations.

3.5.1 *Strengths and limitations*

Analysing data from qualitative studies provided a unique insight into issues of acceptability and engagement, and we were able to identify common factors that impact acceptability and engagement across different neurodegenerative diseases. We also need to be conscious that these studies may represent experiences of those who have agreed to take part in a psychological intervention and agreed to give feedback. There may be other barriers or views among those who did not take part, or who dropped out or did not benefit from the intervention. It is not always easy to capture these perspectives through research studies, however we need to be cognisant that the review findings may present a more optimistic view of participants' experiences.

The majority of the interventions were mindfulness-based, conducted face-to-face and in a group setting. Some of the facilitators and barriers identified by this review may have been influenced by this. For example, difficulties with accessibility and burden may reflect the group and face-to-face format as well as the relatively longer duration of sessions. Additional research with other intervention types and formats may yield different insights into engagement issues. The majority of the studies included people/caregivers of people with dementia or MS and this may have influenced review findings. However, after assessing confidence in each of the review themes, we are confident that our findings did not come solely from particular diseases but were derived from experiences across the different neurodegenerative diseases. Combining the results and discussion sections of included studies for our synthesis could have put an emphasis on the study authors' interpretations of the results rather than the actual findings. However, we were reflexive and checked that the themes did not only come from the study authors' interpretations. We also presented quotes within each theme to demonstrate how themes were derived from both, the study findings (participant quotes) and discussion sections (author quotes).

3.5.2 *Conclusion*

We have identified specific barriers and facilitators to using psychological interventions with people with neurodegenerative diseases and their family/informal caregivers. It was important to tailor information and advice beyond the specific neurodegenerative disease and be flexible to participants' needs and preferences. This was particularly important so that interventions were accessible to people with different physical and cognitive symptoms, and so that interventions were not experienced as burdensome. Different levels of acceptance and readiness can make intervention engagement and acceptability challenging and interventionists need to be sensitive to this and adapt the intervention or their approach accordingly. Once these factors are considered, individuals can experience wide ranging benefits from psychological interventions including changes in insight, perspective, self-efficacy, emotions, and relationships. It is useful to consider and measure the range of potential benefits from psychological interventions, especially with neurodegenerative diseases where physical deterioration and limited prognoses can make interpreting the outcomes of psychological interventions more challenging.

Chapter 4 How do people with MND and caregivers experience a digital mental health intervention? A qualitative study

4.1 Abstract

Objective: There is an urgent need for psychological interventions to support people with Motor Neurone Disease (MND) and caregivers. We used the person-based approach to develop a digital mental health intervention and conducted two studies to explore people's experiences of using it.

Methods: In Study 1, we conducted think-aloud interviews with 9 people with MND and 8 caregivers, and used findings to refine the intervention. In Study 2, 18 people with MND and 9 caregivers used the intervention for 6 weeks after which in-depth interviews were conducted. Data from both studies were combined and analysed using thematic analysis.

Results: We developed 3 main themes around intervention acceptability, engagement, and usefulness. Participants highlighted the importance of accessibility and realistic presentation of information and support. Tailoring and timing intervention use to suit own needs, preferences, and disease stage was also important. Participants used the strategies presented to develop a positive outlook and regain some control. They also faced some challenges using these strategies in the context of dealing with progressive loss.

Conclusion: People with MND and caregivers can find digital mental health interventions useful. Intervention accessibility and flexibility are important for developing acceptable and engaging interventions for MND.

4.2 Introduction

Motor Neurone Disease (MND) is a progressive, neurological disease which can lead to muscle weakness in the limbs, difficulties communicating, difficulties with swallowing and eating, breathlessness as well as other sensory symptoms, cognitive impairment and emotional lability. (Moore et al., 2008). MND is a fatal disease with life expectancy typically 2-3 years from diagnosis (Moore et al., 2008). Currently there is no cure for MND, and treatment and care are centred around optimising quality of life. People with MND and their family members or caregivers can experience high levels of psychological distress and burden (Averill et al., 2007; Burke et al., 2017; Chen et al., 2015; Pagnini, 2013). Psychological support can help improve quality of life, however research evidence for psychological interventions to support people with MND and caregivers is sparse, and there is an urgent need to develop and evaluate psychological interventions (Aoun et al., 2013; Burke et al., 2019; Gould et al., 2015; Zarotti et al., 2021). Some barriers have been identified with engaging with traditional face-to-face therapeutic interventions because of physical disability, and the time and commitment required to participate (Marconi et al., 2016; Van Groenestijn et al., 2015). Digital mental health interventions (DMHIs) can provide an alternative format for psychological support and could be an accessible way to provide psychological support to people with MND and caregivers, particularly for those who have difficulties with mobility or speech, or for caregivers who need support that can fit around care tasks and schedules. In places like the United Kingdom (UK) where demand for psychological support exceeds available resources, DMHIs can also be important for improving access to resources (Davies, 2014; Hollis et al., 2018).

Research on DMHIs for people with MND and caregivers is in its infancy. So far, one online non-meditative mindfulness intervention for people with MND and caregivers, and another blended face-to-face and online acceptance and commitment therapy intervention for caregivers have been reported. Both studies were randomised controlled trials and showed promising results for improving wellbeing and quality of life, however there were also some problems with adherence and drop out (De Wit et al., 2020; de Wit, Vervoort, et al., 2019; Pagnini et al., 2022). Engagement with psychological interventions can be difficult with neurodegenerative diseases, and specifically with MND because of the variability of individual needs and symptoms, uncertainty about the disease and its progression, and challenges with acceptance and readiness to engage with interventions (Pinto et al., 2022; Weeks et al., 2019). Therefore, understanding the experiences of people who might consider using DMHIs is important, and needed to develop engaging and acceptable psychological interventions for people with MND and caregivers.

We developed a DMHI that contained self-guided strategies and practical tips to reduce psychological distress and improve wellbeing. The aim of the current study was to explore how people with MND and caregivers used the intervention we developed which would help us refine and optimise it. We also wanted to develop a broader understanding of how people with MND and caregivers engage with and use self-guided DMHIs.

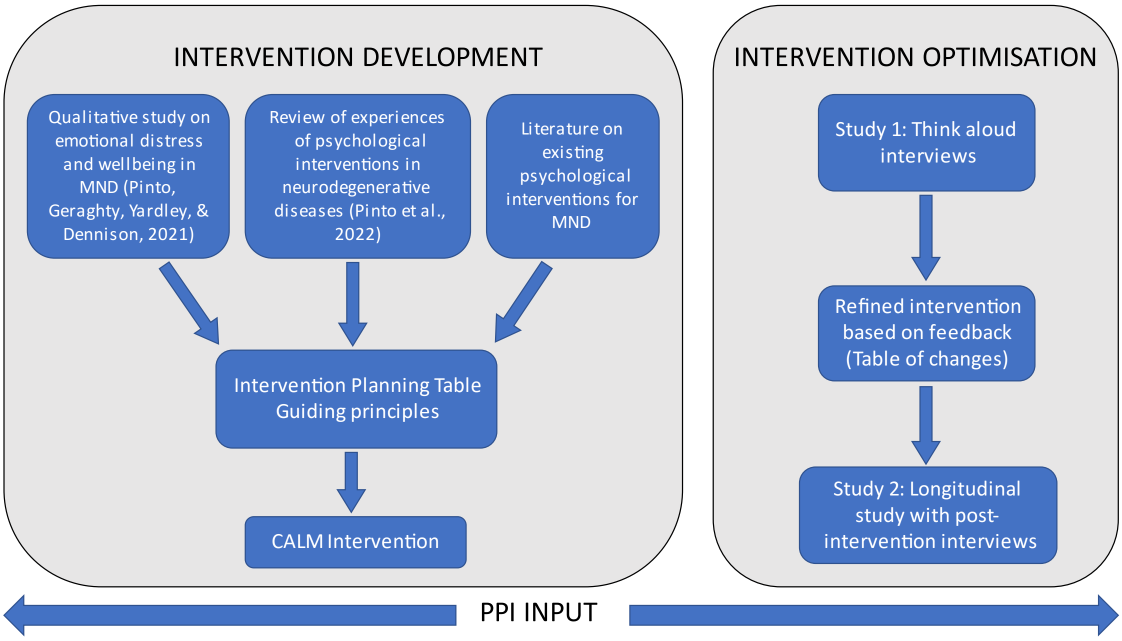
4.3 Methods

4.3.1 Design

We used the person-based approach to develop and optimise the intervention. This approach largely uses qualitative, iterative methods, and incorporates in-depth feedback from target users at different stages of development, optimisation and evaluation (Morrison et al., 2018; Yardley, Ainsworth, et al., 2015). Our overall intervention development and optimisation process is shown in Figure 14. The two studies presented in this paper were nested within the intervention optimisation stage. Both studies were designed to seek user feedback in order to refine and optimise the intervention prior to a more rigorous evaluation. In study 1, we conducted think-aloud interviews with people with MND and caregivers to examine the acceptability of the intervention content and presentation, and findings were used to refine the intervention. In study 2, different people with MND and caregivers used the intervention for 6 weeks and were subsequently interviewed about their experiences.

Figure 14

Process of intervention development and optimisation

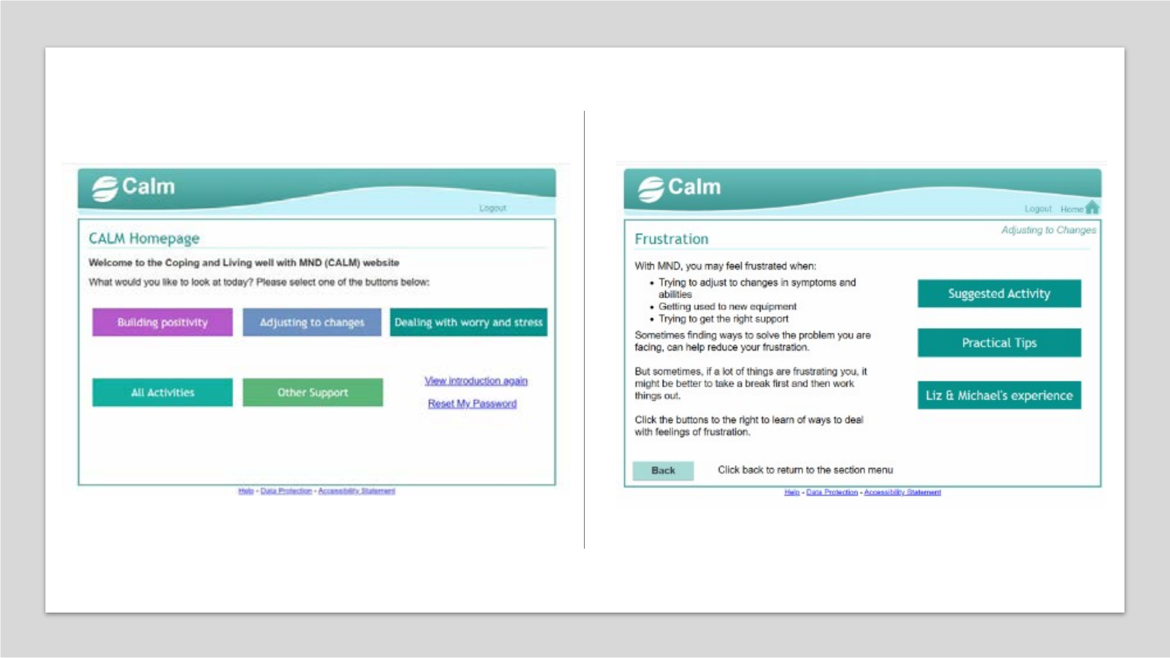


4.3.2 Intervention

We created a digital mental health intervention in the form of an interactive website called Coping And Living well with MND (CALM). The intervention planning was guided by previous research on effective psychological interventions for MND (e.g. Díaz et al., 2016; Pagnini et al., 2017), and qualitative research on coping strategies, preferences for and engagement with psychological interventions (Pinto, Geraghty, Yardley, & Dennison, 2021; Weeks et al., 2019). See Appendix G for the intervention planning table and guiding principles. The website contained activities from cognitive behavioural therapy, mindfulness, acceptance and commitment therapy, and compassion-focused therapy. A detailed description of the intervention development process and the main intervention components can be found in Chapter 1.7.4. The website was meant to be used independently by people with MND and caregivers without any professional facilitation or support. It did not have a guided format and allowed people to choose relevant sections or activities. The information and activities were adapted to be short and accessible to people with various levels of physical ability. The layout and navigation were designed to be simple and easy to use for people who may have different physical and cognitive difficulties (e.g. large font and buttons, limited information per page, compatible to use with screen readers). The design was based closely on an intervention for managing distress in primary care settings (Geraghty et al., 2016). Examples or quotes from people with MND and caregivers were used throughout the website to demonstrate how the information and activities were relevant to MND. See Figure 15, Figure 16, Figure 17 and Figure 18 for examples of pages from the website.

Figure 15

Example 1 of pages from the CALM website



CALM Homepage

Welcome to the Coping and Living well with MND (CALM) website

What would you like to look at today? Please select one of the buttons below:

Frustration

With MND, you may feel frustrated when:

- Trying to adjust to changes in symptoms and abilities
- Getting used to new equipment
- Trying to get the right support

Sometimes finding ways to solve the problem you are facing, can help reduce your frustration.

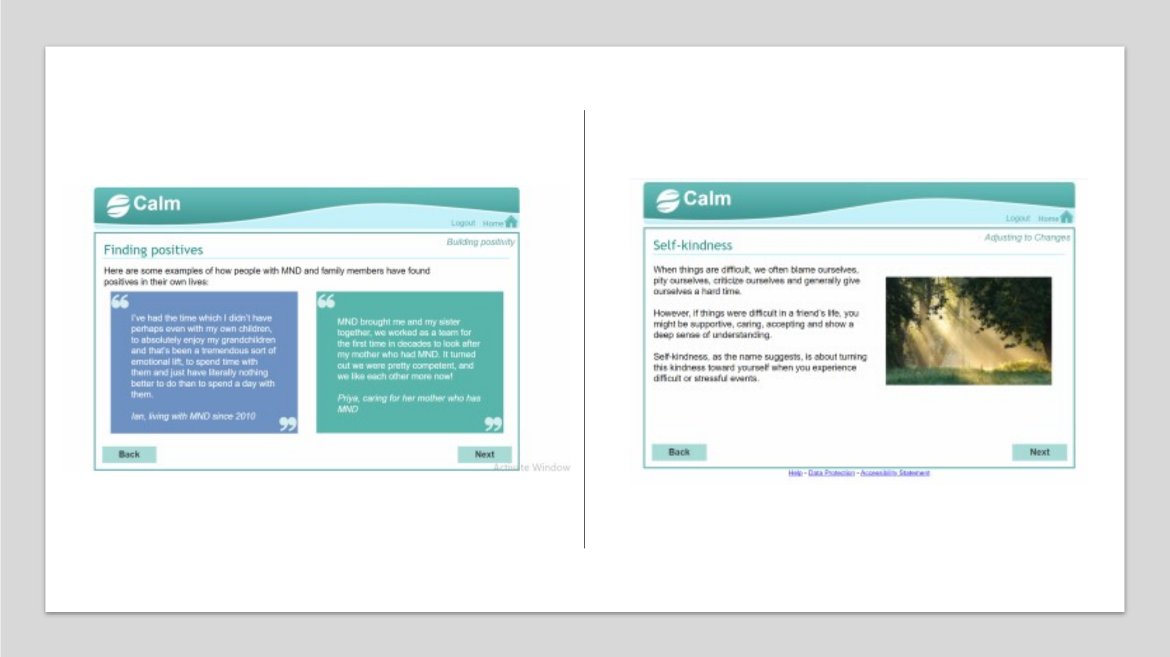
But sometimes, if a lot of things are frustrating you, it might be better to take a break first and then work things out.

Click the buttons to the right to learn of ways to deal with feelings of frustration.

Click back to return to the section menu

Figure 16

Example 2 of pages from the CALM website



Finding positives

Here are some examples of how people with MND and family members have found positives in their own lives:

"I've had the time which I didn't have perhaps even with my own children, to absolutely enjoy my grandchildren and that's been a tremendous sort of emotional lift, to spend time with them and just have literally nothing better to do than to spend a day with them.

Ian, living with MND since 2010"

"MND brought me and my sister together, we worked as a team for the first time in decades to look after my mother who had MND. It turned out we were pretty competent, and we like each other more now!

Priya, caring for her mother who has MND"

Self-kindness

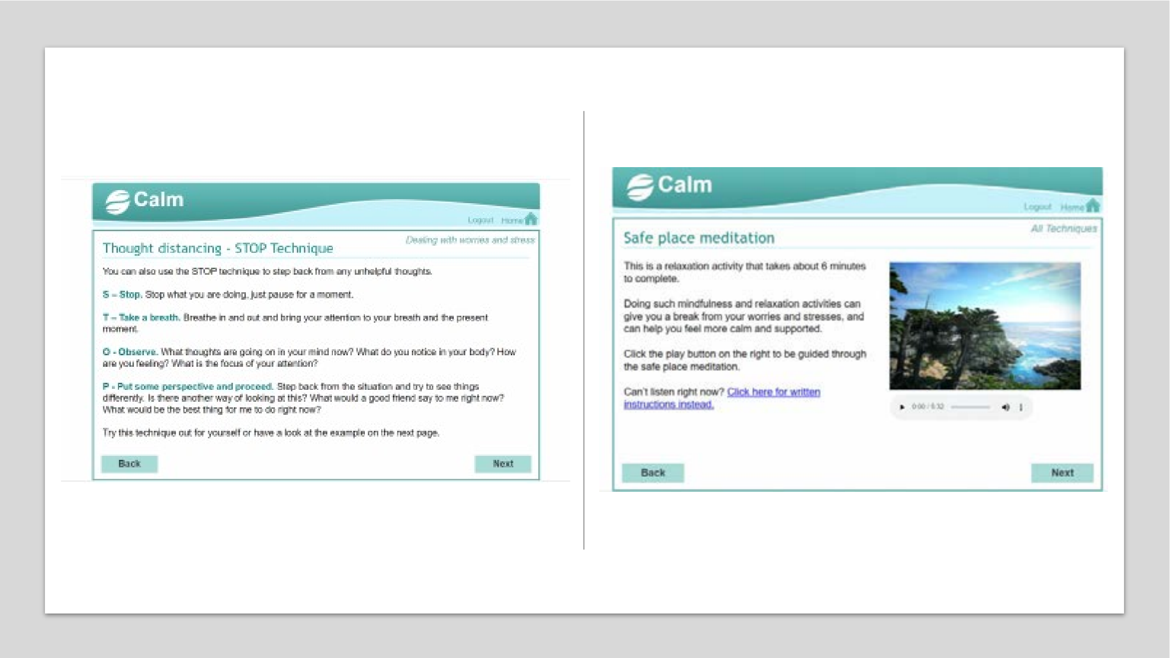
When things are difficult, we often blame ourselves, pity ourselves, criticize ourselves and generally give ourselves a hard time.

However, if things were difficult in a friend's life, you might be supportive, caring, accepting and show a deep sense of understanding.

Self-kindness, as the name suggests, is about turning this kindness toward yourself when you experience difficult or stressful events.

Figure 17

Example 3 of pages from the CALM website



Thought distancing - STOP Technique

You can also use the STOP technique to step back from any unhelpful thoughts.

S – Stop. Stop what you are doing, just pause for a moment.

T – Take a breath. Breathe in and out and bring your attention to your breath and the present moment.

O - Observe. What thoughts are going on in your mind now? What do you notice in your body? How are you feeling? What is the focus of your attention?

P - Put some perspective and proceed. Step back from the situation and try to see things differently. Is there another way of looking at this? What would a good friend say to me right now? What would be the best thing for me to do right now?

Try this technique out for yourself or have a look at the example on the next page.

Safe place meditation

This is a relaxation activity that takes about 6 minutes to complete.

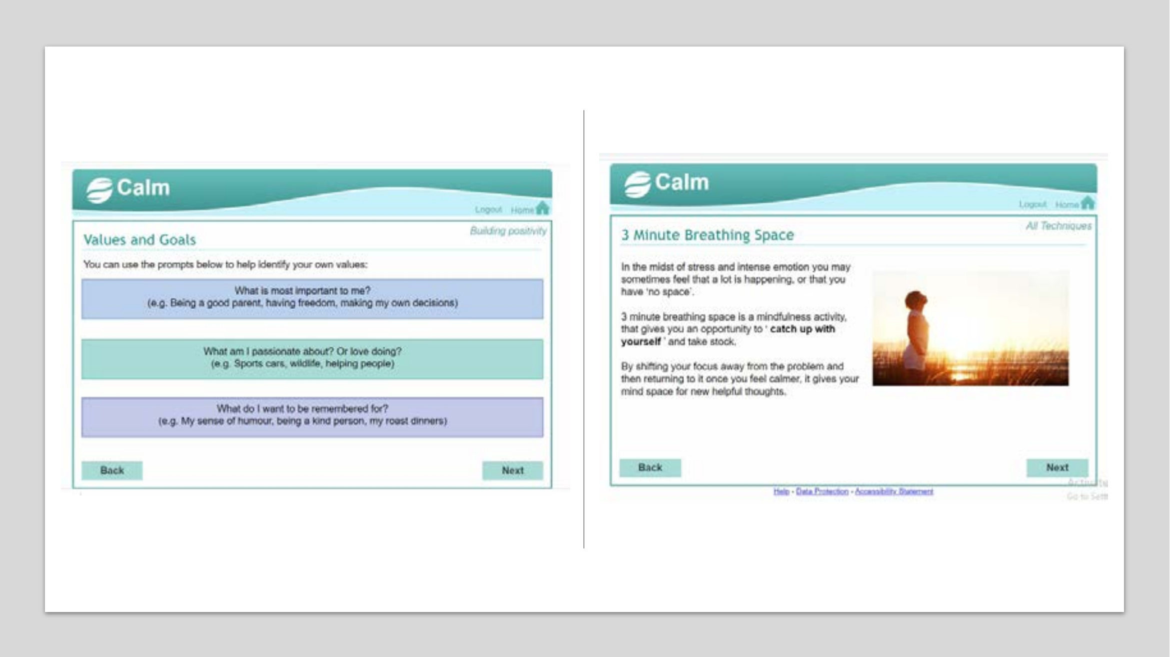
Doing such mindfulness and relaxation activities can give you a break from your worries and stresses, and can help you feel more calm and supported.

Click the play button on the right to be guided through the safe place meditation.

Can't listen right now? Click here for written instructions instead.

Figure 18

Example 4 of pages from the CALM website



Values and Goals

You can use the prompts below to help identify your own values:

What is most important to me?
(e.g. Being a good parent, having freedom, making my own decisions)

What am I passionate about? Or love doing?
(e.g. Sports cars, wildlife, helping people)

What do I want to be remembered for?
(e.g. My sense of humour, being a kind person, my roast dinners)

3 Minute Breathing Space

In the midst of stress and intense emotion you may sometimes feel that a lot is happening, or that you have 'no space'.

3 minute breathing space is a mindfulness activity, that gives you an opportunity to ' **catch up with yourself** ' and take stock.

By shifting your focus away from the problem and then returning to it once you feel calmer, it gives your mind space for new helpful thoughts.

4.3.3 Ethics

We obtained ethical approval from the University of Southampton ethics committee (ERGOII-61216).

4.3.4 Participants and recruitment

Participants were eligible if they had a diagnosis of MND or were a caregiver for someone with MND and had mental capacity to give informed consent. They needed to be able to participate in either a videocall or written feedback and have access to an electronic device and the internet to complete online surveys and use the CALM website. Recruitment was carried out through the UK Motor Neurone Disease Association (MNDA), a charity that supports people with MND. Information about the research was placed in their newsletters, website, and social media platforms. Interested individuals contacted the researcher to discuss participation, were given an information sheet and filled a consent form to take part.

For people with MND, we purposively selected people with a range of symptoms and varying lengths of time since diagnosis. We recruited caregiver participants who were currently caring for someone with MND and wished to use the intervention for their own emotional needs.

4.3.5 Data collection

Data collection took place between October 2020 and July 2021, and was conducted remotely due to the COVID-19 pandemic. The researcher discussed with participants the most appropriate and comfortable way of conducting the interviews (e.g. via phone, video call, or written responses). Where requested or necessary due to physical impairments, participants were interviewed as a patient-caregiver dyad.

4.3.5.1 Study 1

Participants completed a brief online questionnaire that captured demographic and clinical information which was initially used to guide purposive sampling and subsequently used to report characteristics of the sample. This questionnaire included two validated scales. Participants with MND completed the Amyotrophic Lateral Sclerosis Assessment Questionnaire (ALSAQ-40), a self-report of health status capturing physical mobility, activities of daily living, eating and drinking, communication, and emotional reactions (Jenkinson, 2000). Caregivers completed the Zarit burden interview (ZBI-12) (Higginson et al., 2010). Participants were also asked to self-report any cognitive difficulties and caregivers reported if their family member experienced any cognitive difficulties. Interviews were conducted at a date and time convenient to participants via videocall or phone. All interviews were conducted by a researcher (CP), who had training and experience in qualitative methods and had worked clinically with people with neurodegenerative diseases. During the interview, participants viewed the CALM website on their own devices and simultaneously answered interview questions. Think-aloud interviews followed a semi-structured format with a set of standard questions, and follow-up questions based on the particular webpages participants looked at and their initial reactions (see Appendix J). In addition to being analysed for the current paper, feedback was logged in a 'table of changes', discussed regularly with the research team and used to refine the intervention (see Appendix K).

4.3.5.2 Study 2

As with Study 1, participants completed a brief questionnaire online capturing demographic and clinical details (ALSAQ-40 and ZBI-12). Participants were sent a link to the website with brief instructions about logging in and using the website, and a researcher checked in with them after 2 weeks. After 6 weeks, the researcher contacted participants to arrange the videocall or phone interviews. Participants who had difficulties with speech were sent a questionnaire with open-ended questions so they could type their feedback; follow-up questions were emailed if responses needed more elaboration. Interviews were semi-structured and covered questions about participants' overall thoughts and feelings about the intervention, with more detailed questions about using specific advice or strategies (see Appendix L for the interview topic guide).

4.3.6 Patient and Public Involvement

Patient and public involvement (PPI) members had input throughout the entire process of development of the intervention. Initial versions of the intervention were shared with PPI members and feedback was used to improve presentation and navigation. Data collection procedures were reviewed by PPI members, especially the open-ended questionnaire for people who had difficulties with speech, and feedback was used to make the process smoother and interview questions clearer. Some PPI members also gave their input on the preliminary findings.

4.3.7 Data analysis

All interviews were transcribed verbatim and identifying characteristics such as names and locations removed. Data from the think-aloud interviews in study 1 was logged in the 'table of changes' and used by the researcher to refine the intervention (see Appendix K.2). This included specific feedback about the presentation and phrasing of information on the webpages. Interview data from study 2 was used to understand participants' experiences of using the intervention. Data from the think-aloud interviews where participants expressed their views and perceptions about using and engaging with the intervention was added to the data set for analysis. Patient and caregiver interviews were coded separately and compared at the theme development stage. As there were very minor differences, themes were developed drawing on data from both patient and caregiver codes.

A reflexive thematic analysis was conducted to draw themes in relation to experiences of engaging with and using DMHIs and strategies to reduce distress and improve wellbeing in MND (Braun & Clarke, 2006, 2019). After familiarisation with the interviews through repeated reading of transcripts, CP began coding the data inductively. A list of codes was developed and reviewed within the research team to identify interesting aspects and patterns of the data. At this stage, diagrams were used to map out the relationship between different codes across the dataset and identify candidate themes relevant to the research question. Descriptions of each candidate theme were written out and codes were revisited to make sure the descriptions reflected the data. The themes were reviewed by the research team and changes were made to the themes and subthemes. This process involved adding some subthemes that were more descriptive but which formed an important part of the narrative of participants' experience. Themes were defined and theme names were refined iteratively through discussions within the research team and through engaging with the wider literature. Presenting preliminary findings through project dissemination activities also helped further refine the essence of each theme and ensure the theme names reflected this. The findings were written for publication, together with appropriate data extracts that helped convey the story of how participants engaged with the intervention and used the advice and strategies.

4.4 Results

27 people with MND and 17 caregivers were interviewed in total. 14 think-aloud interviews were conducted with early intervention prototypes. 26 interviews were conducted after participants had used the refined intervention for 6 weeks. Most of the interviews were conducted via video call, 2 were conducted via phone, and 7 answered interview questions by giving written feedback. 5 interviews were conducted with both the person with MND and caregiver together, the rest were conducted individually. Demographic and clinical details of the sample can be found in Table 5.

Table 5

Demographic and clinical details of the sample

Characteristic	**People with MND (n=27)**	**Caregivers (n=17)**
Age		

Characteristic	People with MND (n=27)	Caregivers (n=17)
Mean (range)	63.96 (48-92)	54.47 (20-73)
Gender		
Man	14	5
Woman	13	12
Ethnicity		
White British/Irish/Other	27	16
Asian/Asian British	0	1
Relationship to person with MND		
Spouse/partner		11
Son/daughter		5
Sibling		1
Education		
Up to GCSE or equivalent	7	1
A levels or equivalent	5	2
Graduate level	9	10
Postgraduate level	6	4
Diagnosis		
ALS	17	
Primary Lateral Sclerosis	3	
Progressive Bulbar Palsy	3	
Progressive Muscular Atrophy	2	
Kennedy’s disease	2	

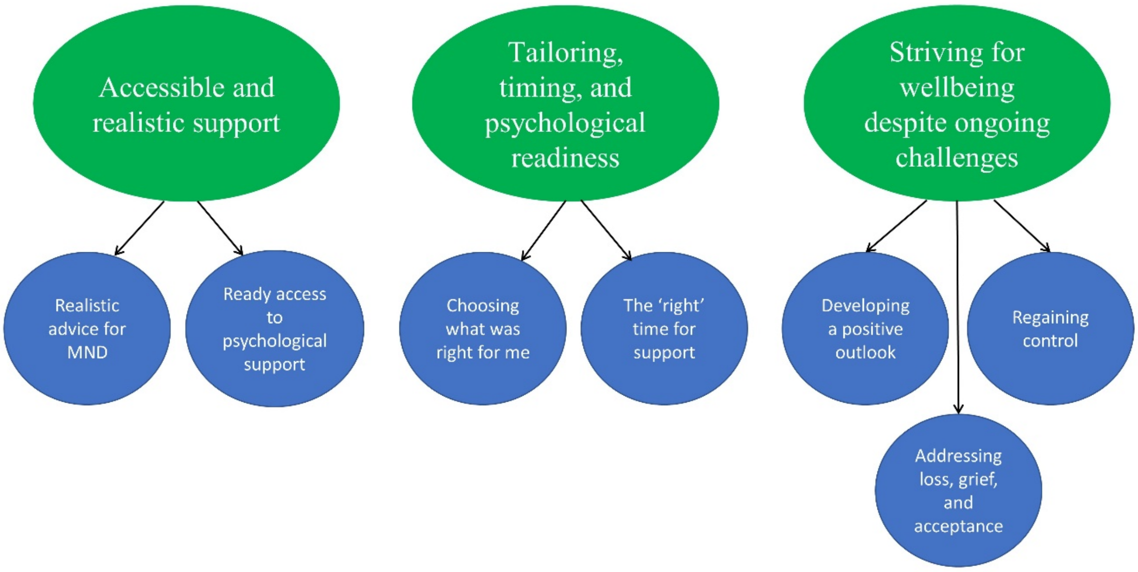
Characteristic	**People with MND (n=27)**	**Caregivers (n=17)**
Time since diagnosis		
Less than 1 year	9	6
1-5 years	8	6
More than 5 years	10	5
Health Status (ALSAQ-40)* Median (IQR)		
Physical mobility	45 (28.75)	
Activities of daily living/independence	50 (28.75)	
Eating and drinking	16.67 (45.83)	
Communication	14.29 (71.43)	
Emotional functioning	40 (27.50)	
Caregiver burden (Zarit Burden Interview)		
No to mild burden (0-10)		1
Mild to moderate burden (10-20)		5
High burden (>20)		11
Cognitive Impairment (self-reported)	5	6

*ALSAQ 40: scores on each subscale range from 0-100 with a higher score representing a greater degree of impairment

We developed 3 main themes and 7 subthemes which reflect key aspects of engaging with and using the intervention and the specific strategies presented (see Figure 19)

Figure 19

Thematic map of main themes and corresponding subthemes



4.4.1 Accessible and realistic support

This theme and respective subthemes highlight the importance and meaning of accessibility of support for people with MND and caregivers. It includes a discussion of the accessibility and realistic presentation of specific advice and strategies, and of having access to psychological support for MND.

4.4.1.1 Realistic advice for MND

It was important for people with MND and caregivers that the information and activities were presented in a realistic way that matched their experience of living with MND and the realities of MND care. The information and strategies needed to be presented in a way that was sensitive to the needs of people with MND and caregivers.

> It didn't tell you just to sort of pick yourself up and go for a walk, up and down the stairs or something when you might not well be able to do that, you'd love to do that but you can't do that, and so it was a bit more appropriate I think to people with motor neuron. (Rachel, person with MND)

Participants stressed the importance of framing advice and strategies in an accessible way particularly for people with very limited physical ability or for those who required assistance or practical support to be able to follow some of the advice on the website. This was discussed with particular reference to sections that encouraged people to do more activities that were meaningful and brought them joy.

> It may be helpful to say when you are planning your list, it may be helpful to discuss with your friend or family member so they can help with any preparations or adaptations. (Priya, caregiver)

Participants also felt that some advice could be phrased more realistically and acknowledge that it may sometimes be difficult to follow because of the realities of living with or caring for someone with MND. One example of this was the thought distancing or mindfulness activities that may provide temporary relief, but with MND worries often crop up again. Participants felt this could be phrased in a way where people could anticipate the benefits of the activity but were also aware of the effort it may take. Similarly, for caregivers it may be difficult to take time out to look after themselves and take a break from caring responsibilities. Some described a tendency to neglect taking care of themselves in order to take care of the person with MND, and feeling guilty when they did take a break.

> My mum looks after my dad and is a full-time carer and it’s fine saying take some time for yourself, but if my dad has to go to the toilet, then she has to help with that, you’re always on call. (Tina, caregiver)

> I was struggling to find time or be able to actually leave the house to go out to an exercise class, to travel to it, to do the class, to travel back because of the time it takes. (Rose, caregiver)

4.4.1.2 Ready access to psychological support

Having a website that provides timely and continuous access to some psychological support was seen as important. Finding and accessing psychological support for MND can be difficult, and some participants described their struggle with the lack of psychological support available. They highlighted the value of being able to readily access tips and strategies and being introduced to different kinds of things that might be helpful. The availability of psychological support through the CALM website was reassuring for some participants who took comfort in knowing that support was there should they ever need it.

> It's very helpful knowing that this stuff is all there, for when you get in such a state that you can't think for yourself really. (Elaine, caregiver)

Having psychological support or advice that was readily available was seen as helpful, especially as things change with the disease. One participant described the time-bound nature of other forms of psychological support, and how having continuous access to some psychological support through a website was important.

> I've done the mindfulness course, I've done this CBT, I've seen the counsellors and then it all stops. Do you know what I mean? So having something like that… OK, I recognize a lot of the little practical tips and the ideas, but I haven't got access to them without this site so that it's helped in that respect. And anytime you do any of those things, even if it's just a quick body scan or a three-minute breathing exercise or something, it has a positive effect. (Rachel, person with MND)

4.4.2 Tailoring, timing, and psychological readiness

In this theme, we present participants' views about tailoring their use of the website to their own needs and preferences. Having the choice to decide when and how to engage with support from the website was important for people with MND and caregivers, and the diversity of views are discussed in the subthemes.

4.4.2.1 Choosing what was right for me

Participants valued being able to choose sections or activities from the website that were relevant to them. It was important for them to choose activities that were in line with their personality or coping style. For example, participants who were familiar with yoga and or cognitive behavioural therapy techniques were drawn to activities like mindfulness or goal setting respectively. Others described how they aimed to stay positive and keep busy as a way of coping with MND, and were not tempted to try activities like mindfulness or thought distancing that required introspection about thoughts and feelings.

> I did look through every section at first, but I knew what would be for me and what wouldn't and the distancing [thought distancing]… I listened to it and went away, but I thought 'no, this isn't for me, so that's fine.' (Liz, person with MND)

How participants coped and what was important to them changed based on what stage they were in the disease. Therefore, having a range of strategies to choose from allowed people to select ones that were appropriate to them at that time. Conversely, participants also decided not to use strategies at certain times. For example, one participant described not wanting to try the mindfulness activities while he still had physical ability to actively do things that would help him feel better. Similarly, one carer explained that she was not at the stage where she was ready to look at the ‘building positivity’ section. For people with MND, the body scan mindfulness activity was a particular activity whose perceived applicability and value appeared to be strongly influenced by disease stage.

> Practical tips and getting rid of the negative thoughts and things like that, that's important to me at the moment, so that's where I went to, just depends on how I felt when I was looking at the website. (Rachel, person with MND)

> No, I did not try these activities [in the building positivity section]. That is not to say they are not necessarily helpful activities, I think I am not ready for these just yet. One stage at a time... (Carmen, caregiver)

Some participants described how they preferred to learn and use the strategies from the website in their spare time, whereas others preferred using them when they were struggling with particular emotions. Not having a fixed format for the website made it easier for participants to adapt website use to these preferences. Both caregiver and patient participants suggested including more activities on the website because having options and choice was important with MND.

4.4.2.2 The ‘right’ time for support

Participants felt it was important to be able to choose and make their own decisions about the best time to engage with psychological support from the CALM website. This was a subjective decision based on participants’ own assessments of how they were coping, and participants at similar stages could have very diverse views. For some participants at an early stage of the disease, their symptoms did not have much of an effect on their daily life and they explained that the website was currently not as useful for them. However, they could see that it would be useful later on, when they were more affected by symptoms or had more caring responsibilities.

> To be quite honest because of my condition at the moment, although I'm slowly losing the ability in my leg, I'm sort of pretty positive about it all… An awful lot of it didn't really refer to how I feel if you know what I mean? I'm sort of still relatively happy and positive. I can imagine once things get worse, I might find it of more interest really. (Michael, person with MND)

There were some participants however, who would have preferred to receive support from a website like this early on, closer to when they were diagnosed. These participants wanted support to deal with the shock of diagnosis, uncertainty about what was going to happen, and the emotional impact of receiving the diagnosis. They explained their journey finding appropriate psychological support or developing their own coping strategies, and said that the advice and strategies from the website mirrored what they had learned or now do. The website served as validation or confirmation that they were on the right track, and was a useful reminder and motivator to continue to use these strategies. A few participants wanted to be prepared for the future, and the website prompted them to think of activities they could do to look after their mental health when their symptoms got worse.

> It's helped me because it has made me do those things again or allowed me to do those things, and reminded me of the positive effect of some of those exercises and ways of thinking and it just sort of, just confirms it really. (Rachel, person with MND)

Some participants felt the website came at the right time for them. They were either experiencing new symptoms or struggling to adjust to new equipment, and felt that the information and strategies could be applied to the problems they were facing.

> I particularly liked this section [self-kindness activity]. I found it helped me through a tough time I was having recently when I was introduced to RIG [radiologically inserted gastronomy] tube feeding and experienced adverse side effects. This section and activities allowed me to look at my thinking "I feel I'm not a strong person to cope with this disease, other people cope." (Jane, Person with MND)

4.4.3 Striving for wellbeing despite ongoing challenges

This theme captures how people with MND and caregivers used the strategies in the website to cope with emotional difficulties and improve wellbeing. It also sheds light on the struggles with using some of these strategies to maintain wellbeing when facing progressive loss.

4.4.3.1 Developing a positive outlook

Participants used advice and strategies from the website to develop and maintain a more positive way of coping with MND. Some participants look at activities they were currently doing, and found it helpful to realize that they were in line with their values. Others planned to do more activities that would bring them joy which allowed them to look towards the future in a more positive way.

> I think I'm someone that likes to look ahead, you know, so having some new goals and new values is very useful, it's a positive thing to focus on. (Jo, person with MND)

People with MND often gave examples of how they made adaptations to continue doing activities they enjoyed, or how they planned to do this when symptoms got worse. Caregivers also described how they adapted activities they enjoy to fit around their caring tasks.

> I like gardening and my husband will sit me down outside and bring everything to me so that I can still plant up planters and hanging baskets but everything is brought to me. (Liz, person with MND)

For other participants, exploring different ways of thinking about their situation gave them a different and more positive perspective. This different perspective came from either following the website activities or from reading examples or quotes from people with MND and caregivers explaining how they coped with difficulties.

> Just reminding yourself that there are other ways of thinking of things, and that sticks in my mind a lot there. Just stopping, taking up a breath, just taking a bit of time to say, 'OK, that's what I'm thinking, that life isn't worth or whatever' you know… But there are other ways of thinking about things and you need to just…

> ‘what are they’, there must be some other way of looking at it. And then just sitting and trying to think that through, that helped quite a bit. (Rachel, person with MND)

Some people with MND said they developed a more positive outlook through looking at themselves and their body not as failing, but in a positive and kinder way.

> From the motor neurone disease point of view, it’s accepting that your body is failing and not getting too angry with that part of your body and being accepting of it, being kind to it… So yeah, I thought that [body scan activity] was good. (Jo, person with MND)

The activities in the building positivity section were seen as relevant and useful, and in keeping with some people’s coping strategies. The activities prompted some to recognize and list positive things in their life. However, some people found that these activities were difficult to do on certain days, when they were stressed or in times of crisis. In particular, people with MND who had very limited ability or no family or social support found it difficult to follow some of the advice or strategies in the building positivity section. Caregivers who witnessed rapid decline in the health of their family member also struggled with some of the activities to promote positivity.

> Positive thoughts are easy when you can do things for yourself, but hard when you can't. (Chris, person with MND)

> It all makes sense really, to try and find the positives i’ what's happening. I did read that several times because obviously there are times when you think everything is negative and so I have been going on that and thinking about ways that I could improve my positivity. (Diana, person with MND)

4.4.3.2 Regaining control

Participants used advice and activities from the website to feel more in control over what was happening. Both caregivers and people with MND spoke about struggling with thoughts about the future, triggered particularly after a setback or symptoms getting worse. In these situations, using some of the advice and strategies from the website (e.g. mindfulness and thought distancing activities) helped them calm down and step back from the situation that was overwhelming them. This then helped them approach the problem in a different and often more constructive way.

> Fear and anxiety took over as I’ve been so independent regarding my daily hygiene and to allow a stranger into my home to help me filled me with dread instead of ease and peace of mind. With CALM [website], [STOP technique for thought distancing] I was able to take a look at and acknowledge this worry and fear, see that these new changes will benefit and make things safer and help me. Surely I want the best for myself. Stopping and observing allows a little clarity in the situation and makes for a good outcome. (Jane, person with MND)

> I do find those [mindfulness activities] really useful 'cause it means you can just focus on something else rather than just powering forward and doing everything else. I do find it really useful to have that time to just stop and think. (Maria, caregiver)

Participants also found it helpful to focus on the here and now, particularly when there were worrying thoughts about the future. The strategies helped them focus on things they can do, as opposed to things they could no longer do. However, some participants explained how it was difficult to focus on the present whilst also needing to plan for the future.

> We really do you have to think ahead of ourselves because [name of person with MND]’s not going to get better. So we have to put things in situ that will help. So it's very…it’s odd 'cause you are in the mindfulness zone and then all of a sudden you think ‘Gosh, I really do have to think of this.’ (Stacy, caregiver)

Participants felt that the website strategies gave them something positive or constructive to focus on, which gave them more control over their situation. Having access to the information and strategies from the website increased their confidence in their ability to deal with difficult emotions.

> I've done the activities when I've been feeling down or been in a quiet and reflective mood, they help me focus on positive things that I can do to help me

> feel as though I'm taking some control over what is happening to me. (Peter, person with MND)

> I'm more positive I guess, and know I have ways of, you know, when I do have bad days say I'll go and listen to the body scan or any of the audios, because I can just close my eyes and listen and just refocus. (Diana, person with MND)

4.4.3.3 Addressing loss, grief, and acceptance

Participants described a need to address and deal with grief and loss, whilst they strived to be positive and engage with strategies to improve their wellbeing. There were constant reminders of things they could no longer do or things that alluded to future loss. These triggers sometimes came from features in the website (e.g. pictures of nature or people, lists of activities, quotes from others). Reminders of loss also were also present when adapting activities or using equipment to continue to do enjoyable and meaningful activities.

> The sadness didn't leave me it stayed there, it was just temporarily overtaken by my enjoyment of either seeing close family and friends, going somewhere or doing another activity. Even though I was doing things I enjoyed my MND was still impacting on the activity. (Peter, person with MND)

Participants wanted more of an acknowledgement of the grief and loss they were experiencing. Accepting loss was seen as necessary in order to be able to do some of the activities. Some participants felt they needed more guidance on dealing with this grief and loss.

> I would like an acknowledgement earlier on in this, that the pleasant things have changed massively. So if I had been doing this pre MND obviously some of these things may be on my list, but there's many, many things that wouldn't be on that list, that can't be on that list. So there's a big sense of loss for me when I think about pleasant activities. (Wendy, person with MND)

Acceptance was also important in order to be open to receiving the information in the website. Participants described this initial struggle with acceptance, but also explained how they experienced benefit when they persisted with the advice and strategies. In some cases, the information on the website facilitated this acceptance by giving people a chance to think about or assess how they were feeling or coping, and giving people permission to think about and address their emotions.

> I think the very first time I went into the website it made me cry and I think that's because it was a real acknowledgement of what I was going through. Maybe that's the first time, I've really, kind of sat down and gone ‘gosh, this is huge.’ That was quite powerful, but I’ve gotten over that as I started using it [the website] but it did have quite a powerful effect the first time I used it. (Jo, person with MND)

4.5 Discussion

In summary, people with MND and caregivers found the intervention useful for developing a more positive outlook and regaining some control over their lives. However, participants also needed to acknowledge and deal with losses and accompanying grief whilst they attempted to use these strategies for their wellbeing. We found that accessibility and realistic presentation of information and support were important for people with MND and caregivers. It was also important for participants to decide what was right for them, based on individual needs and preferences as well as what stage of the disease they were at. Our findings relate to the CALM website, however the themes are applicable more generally to psychological support for MND and using self-guided DMHIs with similar populations.

Similar to our findings, other studies have reported an increase in acceptance and perceived control from using psychological interventions (Bilenchi, Banfi, Pagnini, & Volpato, 2022; de Wit, Vervoort, et al., 2019). People with MND and caregivers also exert control by choosing to engage with services, choosing when to accept help from others or to use assistive devices (Foley et al., 2014, 2014; Pinto et al., 2021) We found that there was considerable variability between participants about which strategies were right for them and the right time for support from a psychological intervention. In the review in the previous chapter, we found that additional personalization beyond disease-specific tailoring was important for intervention engagement and acceptability (Pinto et al., 2022). With the CALM website, we provided a variety of strategies based on different psychological approaches and encouraged users to choose and tailor strategies to their own needs and preferences. The findings showed that participants valued this ability to choose, however it could also lead to variable levels of engagement with the website. Psychological support needs to be flexible and offered at different points in the disease trajectory as symptoms and needs change (Ando et al., 2021; Weisser et al., 2015). DMHIs can offer this flexibility through self-tailoring, however variability in use is to be expected.

Strategies to improve wellbeing may also need to be adapted or repeated to help people stay positive and in control in the context of experiencing loss. Approaches like acceptance and commitment therapy can be helpful as they enable acceptance of both positive and negative experiences in the context of personal values, and the resulting psychological flexibility acts as a buffer to distress (Hulbert-Williams, Storey, & Wilson, 2015). Models of coping and adjustment in MND and other neurodegenerative diseases like MS and PD also highlight the importance maintaining equilibrium in the context of disease progression and how adjustment is a dynamic process (King et al., 2009; Moss-Morris, 2013; Wieringa et al., 2021). Therefore, strategies to improve wellbeing that are successful at one stage may not be as successful or may need to be adapted as the disease gets progressively worse. The willingness and ability to change and learn new strategies may be better indicators of wellbeing and adjustment. Therefore, it is important to capture variables such as resilience, adaptability, or psychological flexibility for progressive diseases like MND as indicators of wellbeing in addition to psychological outcomes related to distress reduction.

The findings from this study point to the relevance of psychological readiness with respect to when participants engaged with the CALM website and when they felt ready to use particular strategies. Participants who took part in this study may have been at different stages of change which could have had an impact on their willingness to engage with the intervention. According to the transtheoretical model, an individual can move between five different stages (i.e. pre-contemplation, contemplation, preparation, action, and maintenance) in the process of changing behaviour (Norcross, Krebs, & Prochaska, 2011). Although these stages of change were originally conceptualized for changing health behaviours, they can also be applied to psychotherapy and more specifically to the behaviour of engaging with the CALM website and employing the different suggested strategies. The variability in responses particularly around the ‘right’ time for psychological support could have been a result of including participants at earlier stages of change. A more in-depth understanding or assessment of a person’s stage of change might be useful for future research to help us understand when people are more likely to engage with this kind of self-guided psychological support, whether particular strategies are more useful/suited for people at different stages of change, and how best to tailor information and support to an individual’s stage of change.

In the previous chapter, the results of the review also highlighted the challenges with acceptance and readiness to engage with psychological interventions, particularly for neurodegenerative diseases as it involves an acceptance of the progressive and terminal nature of the disease. In the CALM website, we took this into account and did not present any information that was too confronting or dealt with the progressive and terminal nature of MND. We also framed information and examples such that it focused more on things that the individual could do to feel better and less on the different problems that people might face. In the intervention introduction, we set expectations about what the intervention included, explained that there were no right ways of coping with MND, and that users could choose how and when to engage with the strategies presented in the website. We still found that there were some challenges with acceptance and readiness for some participants, where people reported that certain activity suggestions or pictures triggered thoughts about future progression. However, for some participants the website and suggested activities helped increase acceptance of the problems they were facing and allowed them to acknowledge the emotions they were experiencing. Therefore, although differences in acceptance and readiness may account for variability in how and when people engage with psychological interventions, it is still worthwhile to pay attention to how interventions are introduced and framed/designed so that people who might struggle with acceptance can also experience some benefit.

Engagement with DMHIs is facilitated when interventions can be integrated into people's daily routines (Borghouts et al., 2021). With MND, additional practical support (e.g. respite for caregivers or assistive devices for people with MND) may be required to enable people to use the self-guided strategies. Although DMHIs can improve access to psychological support, they also have some limitations. They can bring up difficult emotions that may need professional support, or they may not be an appropriate tool to use at certain points (e.g. in crisis or very stressful times). Providing choice about support in online and face-to-face formats can help overcome some of these challenges (Bentley et al., 2020). Self-guided support may lead to people choosing strategies they are comfortable with and not trying out new ways of coping. In these instances, a therapist-supported or hybrid or blended approach to DMHIs may be valuable, where individuals can be guided through helpful strategies and where strategies and feedback can be personalised (de Wit, Vervoort, et al., 2019).

4.5.1 Strengths and Limitations

The user-centred approach to intervention development and evaluation was a strength and enabled us to anticipate some of the issues with engagement and acceptability. Additionally, we obtained views of participants with a range of symptoms, participants at different stages in the disease, and those who looked at the intervention and decided not to use it. This gave us a broader understanding of the limits and challenges of using DMHIs with MND.

Intervention development and the studies conducted to optimise the intervention were conducted by the same researcher and this may have influenced the findings. The researcher took steps to be reflexive about researcher influence through careful framing of the interview questions, asking participants for both positive and negative feedback on the intervention, using field notes to reflect on data collection processes and interviewees' responses, discussing feedback from the interviews with the research team and making changes to the intervention collaboratively, regularly discussing codes and developing themes with the research team. The 6-week period may not have been sufficient for some participants to learn and apply the strategies to problems they encountered. The one-off interviews at the end of the 6-week period were subject to participants' recall and may not have provided a complete picture of intervention engagement.

4.5.2 Conclusion

People with MND and caregivers can find DMHIs useful, particularly for developing a positive outlook and regaining control. It is important to pay attention to accessibility, choice, and flexibility when designing DMHIs for MND so people can tailor their use to meet their own needs. DMHIs can help improve access to psychological support for people with MND and caregivers. However, self-guided psychological support has its limitations and should be provided as an option alongside other forms of psychological support.

Chapter 5 General Discussion

This chapter presents a discussion of the major findings across the three papers in this thesis, elaborating on and extending issues raised in the discussion sections of Papers 1, 2 and 3. Findings about emotional distress and wellbeing in MND and the use of DMHIs are discussed in the context of the wider literature. The implications of the findings for clinical practice have been integrated throughout the discussion, and specific suggestions for future research and DMHIs are outlined. The chapter concludes with a discussion of the strengths and limitations of the thesis and next steps for future research.

5.1 Summary of major findings from the thesis

The three papers in this thesis contribute to knowledge about distress and wellbeing in MND, and the acceptability, engagement, and usefulness of psychological interventions, especially self-guided DMHIs. Paper 1 highlighted the importance of finding hope and positivity, exerting control, having self-compassion, and receiving compassion and support from others, for wellbeing among people with MND and caregivers. Paper 2 looked more broadly at neurodegenerative diseases and found that people with neurodegenerative diseases and caregivers experienced many benefits from psychological interventions. However, to enhance acceptability and engagement, interventions needed to be sensitively tailored to the individual's needs and levels of readiness, and adequately adapted to ensure that using the intervention was accessible and not burdensome. Through the development and initial evaluation of the DMHI developed in this thesis (i.e. the CALM website), Paper 3 demonstrated that self-guided psychological interventions can be useful for people with MND and caregivers, particularly for developing a more positive outlook and regaining control. Participants found accessing support in a digital format easy and accessible, and it helped increase access to psychological support. There was considerable variation in perspectives around what aspects of the intervention were useful and when they were useful, which reiterated the importance of intervention flexibility. Realistic presentation of information and support, especially in the context of experiencing progressive losses was also vital.

5.2 Towards a more dynamic and balanced view of wellbeing in MND

5.2.1 Dealing with grief – confronting and avoiding loss

A consistent finding from both papers 1 and 3 was the importance of hope and positivity for wellbeing in MND. Participants used the CALM website to develop a more positive outlook, however some struggled with being positive whilst also facing progressive losses. In the CALM website, the activities that were presented to promote positivity (i.e. benefit finding, and doing more pleasant activities and value-based activities) were seen as important but some participants felt they needed additional guidance to come to terms with loss. Experiences of loss and positivity appear to be intertwined, and both can be part of the process of adaptive coping. This can be further understood through the dual process model of coping with bereavement which explains that coming to terms with loss involves a dynamic process of oscillation between loss-oriented and restoration-oriented coping (Stroebe & Schut, 1999). At times people will need to confront the loss and process it, and at other times they may avoid the loss, get distracted, or attend to practical things to cope with the consequences of the loss. Both these tasks or processes are necessary, but they do not need to be done relentlessly, the dosage is important (Stroebe & Schut, 1999). This can be applied to caregivers’ experiences as well, where dealing with loss involves periods of coping with losses emotionally as well as performing practical care tasks to cope with the consequences of the functional losses their family member is experiencing, and their experience may oscillate between these two processes. During this grieving process, meaning-making processes (i.e. making sense of the loss and finding benefits from the loss) are also important and can be useful for staying positive (Hall, 2014). Therefore, there will be times when people want to confront the loss and make sense of it, and times when avoiding the loss is adaptive, and psychological interventions are needed to support people as they move through these processes.

Grieving is a highly individualised and dynamic process (Hall, 2014; Stroebe & Schut, 2001) and people with MND and caregivers are constantly going through the process of grieving and coming to terms with progressive losses. This also requires constant adaptation to changing needs and circumstances (King et al., 2009; Pinto et al., 2021). Studies with other similar neurodegenerative diseases like MS and PD have also found that coping requires ongoing adaptation to loss and people change their adjustment modes or coping strategies as a result dealing with continuous loss (Bogosian, Morgan, Bishop, Day, & Moss-Morris, 2017; Haahr, Groos, & Sørensen, 2021). With MND, the rapid disease progression may also mean that some coping strategies that are useful for dealing with change in other progressive diseases like MS and PD, may not be appropriate for people who are dealing with overwhelming change. As pointed out in paper 2, different levels of readiness and acceptance can affect engagement with interventions, but some amount of avoidance may be adaptive for people with MND and caregivers who have to cope with the rapid decline. Fluidity between different ways of adjusting or coping is important. For example, resignation or avoidance which can often be seen as a maladaptive coping strategy, can serve a protective function for a short period of time (Bogosian et al., 2017). Therefore, wellbeing in progressive diseases like MND needs to be seen in terms of adaptability and flexibility, where people shift to different ways of coping over time or with the right support.

Moving between confronting and avoiding loss can also affect how people access and engage with psychological support and how ready they are to seek help from psychological interventions. Paper 2 in this thesis showed that levels of readiness and acceptance influenced how and when people engaged with psychological support. Similarly, with the CALM website, some people felt that it was too early in the disease stage to use psychological support, others made judgements about the type of support or activities that felt right for them and that they were ready to put into practice. This has also been seen in other studies with MND particularly with reference to peer support, where people did not feel ready to meet others or be confronted by the problems that could happen to them (de Wit, Vervoort, et al., 2019; Weeks et al., 2019). Coping mechanisms can be used differently at different stages based on readiness. For example, during the initial stages near diagnosis, hope may be used to seek recovery and hope for a cure, whereas in later stages when there are more severe problems with physical functioning hope may be used to re-define goals or obtain physical assistance to be able to achieve those goals (Madan & Pakenham, 2014). Therefore, when assessing how interventions and coping strategies are used, levels of acceptance and readiness are an important factor to consider.

5.2.2 Adapting sense of control for wellbeing

Living with MND requires constant adaptation, and people respond to changes that occur with different coping strategies that either put more control in their own hands or more control in the disease (King et al., 2009). Regaining a sense of control is important and perceived control has been linked to wellbeing in other neurodegenerative diseases (Bishop, Frain, & Tschopp, 2008; Eccles, Murray, & Simpson, 2011). There were differences in how individuals exerted this sense of control (e.g. through trying to live as normal a life as possible, or choosing when and how to engage with support), and some differences in how people with MND and caregivers exerted control (Pinto et al., 2021). People with MND and caregivers also used the CALM website to regain control over what was happening to them, and this was done by taking a step back from overwhelming situations, focusing on the present and what was in their control, and through learning strategies to deal with their emotions and difficult situations. A recent review showed that people with MND used three main strategies to retain control – developing own illness beliefs, focusing on the present, and maintaining personal values (Glennie, Harris, & France, 2022). Although control is an important factor for wellbeing, there are subjective differences in how control is expressed and used.

In addition to individual differences in using control, control strategies may be used differently or have more or less importance at different stages of the disease (Glennie et al., 2022). For example, as physical disability increases, control may involve redefining goals or values, or making decisions about care. The process of adaptation to change in MND is constant and repetitive, and periods of ‘normality’ or ‘slowing’ tend to be short-lived (King et al., 2009). When things are constantly changing and overwhelming, control strategies may also involve denial or distancing from the future by living day-to-day or adopting a mindful approach (Ando, Cousins, & Young, 2022; Glennie et al., 2022). Being flexible is important for quality of life and wellbeing, and this can be achieved through using strategies like mindfulness and savouring positive experiences (Ando et al., 2022). Therefore, interventions can maintain and enhance wellbeing through promoting a sense of control, despite the unpredictability and uncontrollability of MND symptoms and progression. However, the strategies that are used need to be adapted to the individual’s changing needs and stage of disease.

5.2.3 Wellbeing outcomes or indicators

Having a more dynamic conceptualization of wellbeing also implies that the benefits or value of psychological interventions for people with MND and other progressive diseases need to be seen in the context of these changing needs and circumstances. One way of doing this is by measuring the mechanisms that capture this responsiveness to change (e.g. psychological flexibility, resilience, adaptation) and have an impact on psychological outcomes, rather than only capturing distress or mental health status at a particular point in time (e.g. anxiety or depression). An alternative is to track distress or mood at different points of time in the disease trajectory. However, this method may be too time-consuming and some people may find it too confronting to track their mood as it may relate to their physical decline.

Another way to capture intervention benefits to wellbeing in neurodegenerative diseases is through measuring changes that are relevant and meaningful to the individual, i.e. to have more value- and goal-oriented intervention outcomes (Bogosian et al., 2021; Dennison et al., 2013). Personal values take into consideration the individual’s circumstances and goals can be changed as things change with the disease. There may also be differences in the outcomes that clinicians and people with MND value. Therefore, outcome measures should consider personal values and preferences in addition to standardized clinical outcomes (Glennie et al., 2022). More work is needed to determine how these values or goals can be defined and measured. However, if this can be achieved they may reflect more meaningful outcomes for people with progressive diseases like MND.

5.3 Importance of choice and flexibility for psychological interventions

5.3.1 Choice of therapeutic approaches and activities

The evidence for recommending a particular type of therapy or psychological intervention for MND was limited. However, there was promising evidence for different types of therapeutic techniques including CBT, hypnosis, ACT, and mindfulness (De Wit et al., 2020; Díaz et al., 2016; Pagnini et al., 2017; Palmieri et al., 2012). The findings from Paper 1 pointed to the potential usefulness of mindfulness, self-compassion, and meaning-based activities (Pinto et al., 2021). Therefore, the CALM website contained a mix of different self-guided therapeutic activities. Integrating different therapeutic techniques and approaches can be beneficial for individualising psychotherapy and adapting it to the needs of each client, and it can fit different patients, problems, and contexts (Norcross & Goldfried, 2005; Zarbo, Tasca, Cattafi, & Compare, 2016). The findings from Papers 2 and 3 also highlight the importance of individualizing psychological interventions to account for the variability of needs, preferences, and coping styles. In the CALM website, this individualization was facilitated through providing choice of information and strategies from a variety of psychological approaches, and in Paper 3 participants explained how this enabled them to tailor intervention use to their own needs.

Providing a choice of therapeutic activities and approaches can have some benefits. There is some evidence to suggest that matching treatment and therapy options to patient preferences can facilitate better engagement with therapy and sometimes even improve treatment outcomes (Kwan, Dimidjian, & Rizvi, 2010; Lin et al., 2005; Swift & Callahan, 2009). People often make judgements about the ‘suitability’ or ‘fit’ of therapy with their own needs or preferences which can impact engagement with therapy or interventions (Dennison et al., 2013; Liu & Gellatly, 2021). Actively engaging patients in decisions about what type of intervention they receive can be important to ensure their ‘buy-in’ to therapy which can potentially impact the efficacy of therapy (Marziliano et al., 2021). This ‘buy-in’ can be achieved by introducing the therapy or intervention in a way where people can see the benefit or applicability to their situation and are also aware of the realities of the therapy process or effort involved. The necessity-concerns framework postulates that adherence to a treatment is influenced by the individual’s judgement of personal need for the treatment and concerns about the adverse consequences of the treatment (Horne et al., 2013). This framework has been largely used to understand how patients’ beliefs influence adherence to medicine, but it can also be used to understand how beliefs may impact engagement with psychological support. Understanding people with MND and caregivers’ beliefs about psychological support may help clinicians and service providers tailor the intervention introduction to address people's beliefs about need and concerns, which may encourage buy-in and continued intervention use.

There are some drawbacks to presenting people with a choice of therapeutic approaches or activities, particularly when choice is in the context of self-guided, remotely delivered interventions such as DMHIs. Limited guidance about what activities are best to do and when, may not suit some people who may be confused or stressed and want support in where and how to engage. There is also some evidence to suggest that guided DMHIs have higher engagement than unguided ones (Borghouts et al., 2021). A potential solution would be to use a hybrid approach where a therapist or healthcare professional guides people through self-help resources and can personalise strategies for individuals. When choice is provided, people often choose therapeutic activities or approaches they are either familiar with or ones that suit their preferences, which does not always mean the activity will be useful or beneficial to them. Therapy can involve trying new strategies or challenging existing ways of thinking or coping. Therapist support or guidance can help people through this process and can offer support or feedback when people encounter challenges with these activities. However, adding therapist support to DMHIs does have additional cost and practical implications. An alternative method is to use artificial intelligence (AI) to tailor interventions or provide guidance or feedback. However, using AI for tailoring DMHIs is still in its infancy and further research is needed to determine the value and acceptability of this approach (Gual-Montolio, Jaén, Martínez-Borba, Castilla, & Suso-Ribera, 2022).

5.3.2 Flexible therapy or intervention delivery

It would also be beneficial to provide choice and flexibility about how therapy or interventions are delivered for people with MND and caregivers. The variability of symptoms, disease progression, and needs in MND suggest that the delivery of any psychological intervention needs to be flexible (Weeks et al., 2019). The findings from papers 1 and 2 similarly highlight the need for having flexible psychological support options, particularly as symptoms change and the disease progresses. Potential solutions include having options of receiving psychological support in clinic, at home, or via video-conference (Weeks et al., 2019). Providing choice of therapy timing, location and delivery mode is important in terms of providing patient-centred care and can influence patients' views of the helpfulness of therapy (Irvine et al., 2021; Williams et al., 2016). In the context of an IAPT service for long term conditions, therapists strongly felt that working flexibly helped overcome barriers that people with long-term conditions may have that make it difficult to attend sessions or adhere to therapy. In terms of in-session flexibility adaptations included taking breaks or adjusting session length. However, service and system level constraints could limit the flexibility that could be offered (Carroll et al., 2021). For MND as well, greater flexibility is needed in terms of session frequency, attendance policies, mode of delivery, and session location. Discussions with the person at the beginning of the intervention can help healthcare professionals adapt and customise treatment delivery to the needs of the individual (Weeks et al., 2019).

Recently, telehealth has been used in MND clinics to provide multidisciplinary care (Hobson et al., 2019; Knox, McDermott, & Hobson, 2022). People with MND and caregivers found telehealth solutions acceptable and the uptake and adherence to telehealth systems was good (Helleman, Kruitwagen, van den Berg, Visser-Meily, & Beelen, 2020; Hobson et al., 2019). These findings can be extended to psychological interventions as well, and telehealth may be a good solution to offer this flexibility to people with MND and caregivers. Similarly, DMHIs can offer a flexible and convenient means of psychological support (as seen in Paper 3). However, these digital solutions need to be offered alongside other forms of psychological support so that they meet the variable needs of people with MND and caregivers and so that certain groups who do not use digital technology are not excluded.

Intervention delivery may also need to be adapted to individuals’ disease stage and symptoms, perceived need for psychological support, and levels of acceptance and readiness. In the CALM website, this flexibility was offered by not having a prescribed intervention dose or guided format, and ensuring that the suggested activities were appropriate and sensitive for people at different stages of the disease and with different symptoms. This was also because the nature of progression and symptoms are not always predictable with MND and tailoring to specific diseases stages and symptoms would be difficult. Offering a choice of information and strategies also enabled participants to tailor intervention use based on what they needed at their particular disease stage, and the intervention was reported as useful by participants who were at different stages and severities of MND. The findings from Paper 3 showed that there can be considerable variability in the perceived need for psychological support and this resulted in variable levels of engagement with the CALM website. This perceived need for psychological support does not always correlate with the stage of the disease or symptom severity. Therefore, it may not be pragmatic to tailor interventions and support to the particular disease stage and symptoms as there can still be considerable variability in levels of psychological readiness. Other studies have also found a similar unpredictability and inconsistency in assessing who would need and/or take up an intervention (de Wit, Vervoort, et al., 2019; Dennison et al., 2013). It may be useful to integrate questions about how people are coping (in addition to questions about distress) into assessments in order to gage the perceived need for psychological support.

Providing different options does have cost and logistic implications for services, however it provides a sense of hope and control for service users which can impact their wellbeing.

5.4 Implications for using digital mental health interventions in MND

There are several benefits to using DMHIs to support people with MND and caregivers. DMHIs offer a flexible form of support as they can be accessed and used at any time and at people's convenience. People can dip in and out of this kind of support, use it alongside other forms of psychological support, or decide to not use it all if they are not ready or do not feel they need it. In Paper 3, there was considerable variability in participants' judgment about the 'right' time for psychological support. Self-guided DMHIs can be a well-suited option especially as people's needs change. Findings from Paper 3 also point to the benefit of DMHIs in validating existing coping strategies and offering reassurance that support is available should needs and circumstances change.

Providing access to psychological support is another important feature and benefit of DMHIs. They can be made freely available and people can avoid waiting for access to psychological support. This is particularly important given the short prognosis of MND. People with communication difficulties can often feel forgotten with respect to the provision of psychological support (Weeks et al., 2019). DMHIs can offer an accessible means of providing some psychological support to people who have communication difficulties. The findings from the evaluation of the CALM website in Paper 3 showed that it was accessible and easy to use for people with a range of symptoms and at different stages of the disease. Therefore, DMHIs can be a feasible and acceptable option for psychological support for people with MND and caregivers.

However, there are also certain challenges to developing and using DMHIs for people with MND and caregivers. Making advice and suggestions in DMHIs accessible to people with a range of symptoms and levels of acceptance and readiness can be tricky. With the CALM website, offering suggestions that could be applicable to everyone, dramatically limited the number of suggestions or activities that could be presented. For example, mindfulness activities had to be selected carefully such that they did not focus heavily on breathing or sensations in the body. Additionally, providing options for some groups of people may make them less relevant to other groups of people. For example, by offering activities that were appropriate for people with higher levels of physical disability, those with lower levels of physical disability felt that the activities were not ambitious enough or seemed too limited. Similarly, providing information about dealing with grief and loss may be useful for some users, but not for others who are not ready to confront the loss and want to continue focusing on the positive. Tailoring the suggestions and activities to individual's level of physical ability and levels of acceptance and readiness could be a way forward. However, more research is needed to determine how to tailor information and therapeutic activities to subgroups of people with different needs and characteristics.

DMHIs may not always be an appropriate means of providing psychological support (e.g. when people are in crisis, or if someone expresses a need for one-to-one professional support). Therefore, DMHIs must not be the only available form of psychological support and other forms of psychological support (e.g. face-to-face therapy or peer support) are also needed to meet the variable needs of people at different stages of the disease. DMHIs may be used alongside these other forms of support, however the use of DMHIs in conjunction with other forms of psychological support still needs to be explored.

5.5 Implications for research and future directions

The 3 papers in this thesis have looked at experiences and views of people with MND and caregivers at particular points of time in their disease trajectory, and more research is needed to understand how people engage with and use psychological support or interventions over time. Paper 1 portrayed the experiences of distress and wellbeing of people with MND and caregivers at a particular point in time; in some interviews participants did recall how distress and wellbeing changed over time. There has been some research to suggest that coping strategies and wellbeing may change for people with MND and caregivers in relation to disease progression (Cipolletta et al., 2017; Gauthier et al., 2007; Jakobsson Larsson et al., 2017). However, this research is limited and longitudinal research to gain a better understanding of how people's needs and perspectives change over time would be useful to determine how psychological support can be tailored as these changes occur. Similarly, in Paper 3 participants' experiences of the CALM website over 6 weeks were examined to allow for people to learn the different strategies and apply them to problems they were facing. However, findings did not demonstrate how people adapted intervention use to changes in their symptoms or circumstances. There is some promising research on the lasting long-term effects of psychological interventions, including interventions delivered using telehealth, in other neurodegenerative diseases like PD and dementia (Livingston et al., 2014; Swalwell, Pachana, & Dissanayaka, 2018). However, further research is needed to understand the use and effects of psychological support more longitudinally, as things change with MND.

More research that focuses on MND caregivers is also needed. From the start, the researcher wanted to explore experiences of both people with MND and caregivers. There was a recognition that illnesses are often an intersubjective experience, where people with MND and caregivers go through the illness experience together, whilst also holding different perspectives about the experience (Sakellariou, Boniface, & Brown, 2013). Additionally, there is very little research on psychological interventions for caregivers (Cafarella et al., 2022), and the researcher (and indeed the funder of the PhD project) did not want to exclude caregivers if they could potentially benefit from the same intervention. Therefore, both types of participants were included for all 3 studies and findings are presented in relation to both patient and caregiver experiences. The intervention developed mainly focused on issues that people with MND faced, but also included caregivers as participants as they could benefit from similar strategies. This is reflected in the sample sizes and sampling criteria where there were fewer caregivers who participated and fewer criteria to select caregiver participants. Although the CALM website had advice and strategies that were applicable to caregivers, MND caregivers may have additional needs that were not covered. Therefore, further research and separate interventions that focus on the specific needs of MND caregivers are warranted.

The findings from this thesis point to the importance of assessing acceptability and engagement with psychological interventions, in addition to effectiveness. More research is needed to understand how engagement differs with different types of psychological interventions and with different methods of intervention delivery, and how different levels of engagement can impact outcomes. Future research can also explore the use of behaviour change theory and techniques to promote engagement and adherence to psychological interventions.

5.6 Strengths, limitations, and reflections on methods used

5.6.1 Using the PBA and qualitative methods

One of the major strengths of this thesis was the use of the person-based approach at all stages of intervention development and evaluation. Using this approach enabled the identification and anticipation of challenges with acceptability and engagement and the implementation of solutions to overcome these challenges. Additionally, intervention components were created in response to users' needs, as opposed to approaching intervention development solely from a particular theoretical or therapeutic approach. This is particularly important for situations when there is limited evidence for the effectiveness of specific psychological interventions, and where novel solutions are being developed. As a result, all three papers present a nuanced understanding of the experiences and context of people with MND and caregivers in relation to wellbeing and using psychological interventions, that can be utilized by other researchers and psychologists working to develop or provide psychological support for MND. The PBA also provided a systematic way of recording and using participant views and feedback, and the resulting intervention benefitted from these incremental learnings.

Subsequently, using this approach also highlighted future improvements for the CALM website (see Appendix N).

Developing these insights through conducting the research studies described in this thesis did require time, and it was not possible to assess intervention feasibility and effectiveness within the scope of this PhD project. However, this in-depth intervention development work was needed to create an intervention that was acceptable and useful for people with MND and caregivers, especially given the limited intervention research. It also avoids wasting resources on trialling an intervention that may not be acceptable or engaging for users. Although some issues with engagement and acceptability were highlighted, not all of them could be addressed in the scope of a PhD project (e.g. use of more audio-visual content, more professional website design and high-tech features) because additional technical expertise and resources were needed. This is one of the challenges with using user-centred approaches, sometimes more resources may be required to respond well to user feedback and this cannot always be anticipated at the start of the project.

Across the three papers in this thesis, qualitative methods were used to explore and understand participants' experiences of wellbeing and use of psychological interventions. This resulted in the development of an intervention that was grounded in the needs and views of its target users. Using qualitative methods made it possible to capture challenges with engagement and acceptability and explore the views of participants who did not like or want to use the intervention. DMHIs often have issues with drop out and adherence, especially interventions with no human input and in real-world settings outside of the research study (Baumel, Muench, Edan, & Kane, 2019; Donkin & Glozier, 2012; Mohr, Lyon, Lattie, Reddy, & Schueller, 2017). Including the views of people who did not use the intervention or decided it was not suited for them, was a particular strength of Paper 3 as it presented a broader picture of engagement with DMHIs. Findings and themes from Paper 2 and Paper 3 highlighted similar issues with acceptability and engagement for psychological interventions with MND and other neurodegenerative diseases. This points to the broader applicability and transferability of findings to other progressive and terminal illnesses.

5.6.2 Social dimensions of wellbeing

Social factors and social support are equally important for emotional wellbeing. Paper 1 outlined how a lack of support can cause emotional distress and how support from family, peers, and healthcare professionals can contribute to wellbeing. The CALM website presented experiences and quotes from people with MND and caregivers to convey empathy and peer support. Some of the practical tips and advice on the website recommended social activities for improving wellbeing. It also encouraged people to communicate their feelings to family, friends, or healthcare professionals, and signposted people to forums for peer support. Beyond this however, the website did not integrate any peer support or social support features. The evaluation of the website indicated that social support was important for users. For example, people with MND discussed the need for support from family members to be able to do activities that were in line with their values, or the positive things that people appreciated about their lives and wanted to do more of often included social aspects such spending time with family or grandchildren or friends. Interventions like the CALM website may be useful for people who already have some social support or for those who are able to access social support. Some psychological interventions with MND that have integrated group or peer support have found that this intervention component was highly valued by participants (Cipolletta et al., 2018; Locock & Brown, 2010; Ugalde et al., 2018). Therefore, it is also important to think about how to support people with MND and caregivers who do not have social support resources and develop interventions that address these social needs as they are important for wellbeing.

5.6.3 Digital divide

One of the drawbacks in developing a DMHI was that it could limit access to support for some groups of people. Digital solutions are intended to reduce inequality in terms of access and provision of healthcare. However, these solutions may also widen the gap of health inequalities where users of digital interventions have better access to healthcare resources and better health outcomes than non-users of digital interventions. This is known as the digital divide. A 2019 report by the Office for National Statistics (ONS) revealed that although declining, the number of internet non-users is still large in the UK. In 2018, there were 5.3 million internet non-users, 10% of the adult population (ONS, 2019).

Several factors contribute to the digital divide including poverty, low literacy, lack of interest in or lack of access to technology (Latulippe, Hamel, & Giroux, 2017; Saeed & Masters, 2021). Older age and ethnicity have also been associated with the digital divide or non-internet use, as these characteristics are often linked to low income and lack of access to technology (Latulippe et al., 2017). Among older adults internet use is growing rapidly, however this is still a group that uses the internet the least because of barriers such as lower income and lower levels of physical and cognitive ability (Latulippe et al., 2017). The CALM website could only be used by people who had access to electronic devices and the internet. Additionally, recruitment and data collection procedures were mainly conducted through email, online surveys, and videocall. This may have excluded certain groups of people from taking part in this research.

If resources allowed one way to bridge this digital divide would be to provide participants where disparities are known to exist, with access to devices so they can use the intervention along with technical training or assistance to use the intervention (Latulippe et al., 2017). Another way to reduce the digital divide is to develop interventions using a user-centred approach and including diverse perspectives in the development and design, particularly those at risk of social health inequalities, so that the intervention can be as accessible as possible. A user-centred approach was used to develop and evaluate the CALM website. Specific features such as using simple language, larger fonts, limited information per screen, simple and intuitive navigation with 'next' and 'back' buttons so the website was almost like reading a book, helped increase the accessibility of the website for people with lower digital literacy. Older people, people with various levels of physical disability, and people with mild cognitive impairment were included in the sample, and their perspectives were taken into account in terms of intervention development. Although attempts were made to improve the accessibility of the CALM website, it was not suitable for people who had severe cognitive impairment or dementia as a lot of the strategies involved reading information and following a series of instructions. Additionally, it was difficult to provide suggestions and strategies for people who had severe disability because of MND (e.g. difficulties with breathing might limit ability to do certain mindfulness activities) and few participants who were in the later stages of MND reported difficulties following certain activities and suggestions. Therefore, although DMHIs need to take accessibility into account in their development, a digital solution may not always be appropriate or accessible for all people with MND especially those with more severe symptoms and disability.

People from ethnic minorities and low levels of education or literacy were under-represented in the sample. Data about socio-economic status or income was not collected, and this was a limitation of the research. The researcher was mindful of these different factors influencing digital intervention use and attempts were made to recruit a range of participants and to provide technical support during recruitment, data collection and whilst using the website. However, existing knowledge, familiarity with, and access to technology may have still influenced participation in the studies. The researcher has also acknowledged the limitations and boundaries of DMHIs and has suggested that other forms of psychological support are made available, especially for people who have difficulty accessing and using technology.

5.6.4 Sampling, inclusion, and recruitment

In Paper 3, the aim was to explore acceptability and engagement and therefore, seek perspectives from participants with a range of characteristics. Purposive sampling was used to select participants with different physical symptoms, severity of symptoms, and duration since diagnosis. This was achieved and the findings come from the experiences of a range of participants. There were two sampling criteria which could have benefitted from input from clinical records or healthcare professional assessment – identifying people with cognitive impairment and identifying people at a late stage of MND. This could have been collected by recruiting participants from the National Health Service (NHS) and conducting detailed assessments of cognitive functioning. However, recruiting via the NHS was not practical after the Covid-19 lockdowns and restrictions, and the cognitive assessments would have been too burdensome for participants especially if they were conducted remotely.

Paper 2 showed that levels of readiness and acceptance can affect participation and use of psychological interventions (Pinto et al., 2022). Participants were mainly recruited through the MNDA. The researcher originally planned to also recruit participants via the NHS which would have given access to a more diverse pool of participants. However, recruitment methods had to be adjusted in light of the COVID-19 pandemic. Participants who may be willing to review research study adverts and respond to them, might already be proactive in seeking help and may already had some coping strategies in place so that they can look for relevant interventions or research opportunities. Recruitment via the NHS would have benefitted the study in terms of being able to invite participants with a variety of clinical characteristics and at various stages of being ready to seek psychological support.

5.6.5 Reflections on data collection methods

In all studies in this thesis, data collection methods were adapted to be flexible and respond to the needs of participants. This involved offering different ways to conduct the interviews (e.g. via written responses, phone or video call interviews), asking participants about adjustments that needed to be made so that they could take part (e.g. timing and length of interviews, or involving caregivers), being sensitive to participants' symptoms during the interview and adjusting interview questions accordingly, and checking in with participants at regular intervals to make sure they were comfortable. These methods helped facilitate participation from people with a range of physical symptoms, with different severities of these symptoms, and it also communicated to participants that their experiences and needs were important. These research practices aimed to be respectful and inclusive to participants, and also facilitated the collection of rich and in-depth qualitative data.

There were some challenges with using data collection methods flexibly. For example, with written interviews, participants did not always provide rich explanations or accounts, and there were limited ways the interviewer could follow up without burdening participants with too many interview questions. Participants who had difficulties with speech could also have difficulties using their fingers to type answers or need assistive equipment or support from caregivers to type answers to the interview questions. Therefore, it was important balance the need to collect rich, thick data whilst being sensitive to participants' needs and levels of ability. Similarly, when both patient and caregiver participants wanted to be interviewed together, there may not have always been equal representation of both patient and caregiver views in response to all interview questions, or responses may have been influenced by the presence of both participants together. The researcher reflected on and described these difficulties through field notes. However, being flexible with and adjusting these methods was important to enable participation from a diverse group of people and helped keep the research and intervention development user-centred.

The interviews conducted for the development and evaluation of the CALM website (i.e. in Paper 3) were mainly conducted remotely due to restrictions during the COVID pandemic. One of the strengths of conducting interviews remotely was that people could take part from all over the UK. Interviews via video call have also been reported by participants to be easy, convenient, accessible, and an acceptable way to discuss personal and private topics (Archibald, Ambagtsheer, Casey, & Lawless, 2019; Gray, Wong-Wylie, Rempel, & Cook, 2020; Jenner & Myers, 2019). Additionally, video-recording the think-aloud interviews made it easier for the researcher to clearly view how participants navigated the website and their responses to each part of the website were recorded.

As work and social activities moved online due to lockdown restrictions, participants were more familiar with using the internet and participating in video calls. Many aspects of care (e.g. MND clinic appointments or attending local support groups) had also moved online. This meant that participants who may not have otherwise accessed online interventions took part in the study (e.g. older people, people not as familiar with using technology). There is still a limitation in relation to the digital divide where participants who did not use or have access to technology could not take part in this research. However, the relative ease and inclination for accessing support online was influenced by the context of the COVID-19 pandemic and participants' increased familiarity with using technology.

5.6.6 Other practical limitations

Developing a DMHI within the context of a PhD project had certain challenges. Some of the intervention content was adapted from an existing intervention (Geraghty et al., 2016). However, other content needed to be developed by the researcher and designed to fit the overall structure of the website. The researcher had input from the supervision team and some assistance with designing and creating the website using appropriate software. However, additional assistance in terms of software development and graphic design would have been beneficial to the timescales and overall quality of the intervention.

There were also some challenges with obtaining PPI input at different stages of the project. PPI members (mainly people with MND) wanted to provide assistance but fluctuations with health and disease progression meant that it was not always possible to obtain input from all members or hold meetings where everyone could attend. The researcher liaised with different PPI members on an individual basis and for different kinds of input depending on the PPI members' capacity to provide feedback at the time. This also meant that new PPI members had to be recruited at various stages of the project and a larger group was required to accommodate for drop out or lack of engagement during certain periods. Due to tight timelines, it was not always possible for PPI members to be involved in all aspects of study design and this could have been improved with better planning. A detailed reflection of the role and contribution of PPI to this PhD thesis has been outlined in Appendix M using the GRIPP2 (guidance for reporting involvement of patients and the public) checklist (Staniszewska et al., 2017).

5.7 Dissemination of findings and next steps

The researcher presented findings from the 3 studies at various academic conferences and lay group meetings. The audiences included academics, healthcare professionals, people with MND, caregivers, and people working with MND charities. From these meetings and discussions, it was clear that there was a lack of available psychological support for people with MND and caregivers, and a need or demand for provision of psychological support. This led to the researcher applying for funding to conduct some public engagement workshops with healthcare professionals and volunteers working to support people with MND to discuss the application and implementation of the CALM website. Discussions from these workshops confirmed that healthcare professionals and volunteers were supportive of the CALM website for MND. They felt that a self-guided DMHI had a place alongside other forms of support and could fit in with how care was provided in clinical practice. The variability in how participants responded to support from the CALM website mirrored their experiences of providing informational support in clinical practice. They also felt that the self-guided format had the additional benefit of giving people permission to acknowledge and express how they were feeling in their own time and space, and that this could encourage them to reach out to healthcare professionals and other resources for help.

These dissemination and public engagement activities show that there is a clear local and national need for psychological support in MND. The CALM website can be a useful means of providing some support and discussions will continue about how CALM (either in its current or a further refined state) could become part of MND service provision.

5.8 Conclusion

The research in this thesis has been carried out in response to the need for both a better understanding of what psychological interventions can help in MND and the need to find practical and cost-effective ways to reach families affected by this disease. This was an under-researched area. The findings from this thesis have provided a better understanding of emotional distress and wellbeing in MND and have identified important issues with acceptability and engagement with psychological interventions for MND and other similar neurodegenerative diseases.

The first paper identified the importance of hope, control, and compassion for wellbeing in MND and the need for interventions and support to harness these factors. The second paper highlighted the benefits of psychological interventions for neurodegenerative diseases but also stressed the importance of considering adequate intervention tailoring, intervention accessibility and burden, and individual’s levels of readiness when designing psychological interventions for neurodegenerative diseases. The development of the CALM website was guided by findings from these two papers. Evaluation of the CALM website showed that self-guided psychological interventions can be useful for people with MND and caregivers, particularly for developing a more positive outlook and regaining control. Findings from paper 3 also pointed to the importance of accessibility, choice, and flexibility when developing DMHIs to meet the varying needs of people with MND and caregivers at different stages of the disease.

Findings from the three papers in this thesis have important implications for clinical practice and psychological intervention development for MND. Any intervention or support for MND needs to pay attention to concepts such as hope, control, and compassion. Intervention accessibility and flexibility are key to acceptability and engagement with interventions for MND. Future research is needed to determine the effectiveness of DMHIs such as the CALM website, for improving wellbeing outcomes and the contexts and mechanisms that are important. It would also be useful to investigate the long-term effects of using self-guided strategies to improve wellbeing for people with MND and caregivers, and explore how these self-guided strategies are used alongside other forms of psychological support.

Appendix A Consolidated criteria for reporting qualitative studies (COREQ)

Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care*. 2007. Volume 19, Number 6: pp. 349 – 357

No. Item	**Guide questions/description**	**Reported on Page # or section**
Domain 1: Research team and reflexivity		
Personal Characteristics		
1. Interviewer/facilitator	Which author/s conducted the interview or focus group?	CP – Methods section
2. Credentials	What were the researcher's credentials? E.g. PhD, MD	PhD student – Methods section
3. Occupation	What was their occupation at the time of the study?	PhD student – Methods section
4. Gender	Was the researcher male or female?	Female
5. Experience and training	What experience or training did the researcher have?	Methods section
Relationship with participants		
6. Relationship established	Was a relationship established prior to study commencement?	No
7. Participant knowledge of the interviewer	What did the participants know about the researcher? e.g. personal goals, reasons for doing the research	Participants knew that the researcher was a PhD student at the University of Southampton, doing her PhD on emotional distress and wellbeing in Motor Neurone Disease (MND). This information was described on the participant information sheets and study adverts.
8. Interviewer characteristics	What characteristics were reported about the interviewer/facilitator? e.g. Bias, assumptions, reasons and interests in the research topic	The interviewer did not have much prior knowledge of MND, but had previously worked

		with and interviewed people with Parkinson’s disease and people receiving palliative care.
Domain 2: study design		
Theoretical framework		
9. Methodological orientation and Theory	What methodological orientation was stated to underpin the study? e.g. grounded theory, discourse analysis, ethnography, phenomenology, content analysis	The study used methods (in-depth semi-structured interviews and reflexive thematic analysis) in line with an interpretivist approach.
Participant selection		
10. Sampling	How were participants selected? e.g. purposive, convenience, consecutive, snowball	Purposive sampling – Methods section
11. Method of approach	How were participants approached? e.g. face-to-face, telephone, mail, email	Methods section
12. Sample size	How many participants were in the study?	Methods section
13. Non-participation	How many people refused to participate or dropped out? Reasons?	Participants contacted the researcher if they wanted to take part in an interview. No one dropped out.
Setting		
14. Setting of data collection	Where was the data collected? e.g. home, clinic, workplace	Methods section
15. Presence of non-participants	Was anyone else present besides the participants and researchers?	No
16. Description of sample	What are the important characteristics of the sample? e.g. demographic data, date	Methods section
Data collection		
17. Interview guide	Were questions, prompts, guides provided by the authors? Was it pilot tested?	Methods section and Appendix B
18. Repeat interviews	Were repeat interviews carried	N/A

	out? If yes, how many?	
19. Audio/visual recording	Did the research use audio or visual recording to collect the data?	Methods section
20. Field notes	Were field notes made during and/or after the interview or focus group?	Field notes were made after each interview.
21. Duration	What was the duration of the interviews or focus group?	Average duration reported – Methods section
22. Data saturation	Was data saturation discussed?	No. We were not aiming for theoretical data saturation, but rather to look at patterns of experience across participants with diverse characteristics.
23. Transcripts returned	Were transcripts returned to participants for comment and/or correction?	No
Domain 3: analysis and findings		
Data analysis		
24. Number of data coders	How many data coders coded the data?	One – Methods section
25. Description of the coding tree	Did authors provide a description of the coding tree?	N/A
26. Derivation of themes	Were themes identified in advance or derived from the data?	Inductive thematic analysis, Methods section
27. Software	What software, if applicable, was used to manage the data?	NVivo
28. Participant checking	Did participants provide feedback on the findings?	Not participant checking but a summary of findings was sent to all participants.
Reporting		
29. Quotations presented	Were participant quotations presented to illustrate the themes/findings? Was each quotation identified? e.g. participant number	Yes, Results section

30. Data and findings consistent	Was there consistency between the data presented and the findings?	Yes, illustrative quotations have been provided for each theme and sub-theme – Results section
31. Clarity of major themes	Were major themes clearly presented in the findings?	Results section and figures
32. Clarity of minor themes	Is there a description of diverse cases or discussion of minor themes?	Description of diverse cases embedded within the explanation of themes – Results section

Appendix B Interview topic guide for Paper 1

- Can you tell me about your experience with MND?
    - wherever the participant would like to start, could be about diagnosis or their experience now.
- Have your thoughts and feelings changed since you were diagnosed or are they the same? (If different, in what way have they changed?)
- What's it like to live with MND? Can you tell me a bit more about that?
- What are your main concerns/worries/emotions you face on a day-to-day basis?
- What are your thoughts and feelings about living with MND?
- Can you tell me about how you manage MND on a daily basis? If you find yourself having emotional concerns or worries, do you have any ways of managing them?
- Can you tell me about any support you feel you have? Or people/ things/ processes that have helped you?
- (In addition to this support) Do you have any thoughts about what else might support you to cope with MND?
    - particularly additional support with any emotional concerns/worries that participants have previously mentioned.

Appendix C Search terms used for all databases

Psychological Interventions	**Neurodegenerative disease**	**Qualitative or mixed methods**
Psychological intervention	Neurodegenerative diseases/conditions/illness	Qualitative
Psychosocial intervention	Progressive neurological diseases	Interview
Psychotherapy	Motor neuron disease	Focus group
Psychological therapy	Amyotrophic lateral sclerosis	Experience
Cognitive behavioural therapy	Parkinson’s disease	Perspective
Acceptance and commitment therapy	Huntington’s disease	View
Mindfulness	Alzheimer’s disease	Feedback
Meaning centred therapy	Dementia	Ethnography
Psychoeducation	Multiple sclerosis	Thematic analysis
Self help		Theme
Education program		Usability
Support program		Feasibility
		Evaluation
		Mixed methods

Appendix D Example of modified terms and search strategy for MEDLINE

1. “psych* intervention”.tw
2. Psychotherap*.tw
3. “psychologic* therap*”.tw
4. “cognitive behavio#r therapy”.tw
5. “CBT”.tw
6. “cognitive behavio#ral therapy”.tw
7. “acceptance and commitment therapy”.tw
8. Mindful*.tw
9. “self help”.tw
10. Psychoeducation.tw
11. “psycho-education”.tw
12. “education program*”.tw
13. “support program*”.tw
14. “meaning cent*”.tw
15. Psychotherapy/
16. exp Cognitive Behavioral Therapy/
17. exp mindfulness/
18. 1 OR 2 OR 3 OR 4 OR 5 OR 6 OR 7 OR 8 OR 9 OR 10 OR 11 OR 12 OR 13 OR 14 OR 15 OR 16 OR 17
19. “progressive neuro*”.tw
20. “neurodegenerative dis*”.tw
21. “neurodegenerative ill*”.tw
22. “neurodegenerative condition*”.tw
23. Neurodegenerative diseases/
24. “motor neuron* disease”.tw
25. exp Motor neuron disease/
26. “amyotrophic lateral sclerosis”.tw
27. “Parkinson’s disease”.tw
28. exp Parkinson disease/
29. “Huntington’s disease”.tw
30. exp Huntington disease/
31. exp Multiple system atrophy/
32. “Alzheimer’s disease”.tw
33. Dementia.tw
34. exp dementia/
35. “multiple sclerosis”.tw
36. exp Multiple sclerosis/
37. 19 OR 20 OR 21 OR 22 OR 23 OR 24 OR 25 OR 26 OR 27 OR 28 OR 29 OR 30 OR 31 OR 32 OR 33 OR 34 OR 35 OR 36
38. Qualitative.tw
39. Interview.tw
40. “focus group”.tw
41. Experience.tw

42. Perspective.tw
43. View.tw
44. Feedback.tw
45. Ethnograph*.tw
46. Thematic.tw
47. Theme.tw
48. Usability.tw
49. Feasibility.tw
50. Evaluation.tw
51. “mixed method*”.tw
52. exp Qualitative research/
53. 38 OR 39 OR 40 OR 41 OR 42 OR 43 OR 44 OR 45 OR 46 OR 47 OR 48 OR 49 OR 50 OR 51 OR 52
54. 18 AND 37 AND 53

Appendix E Quality assessment of included studies using CASP checklist

Author (date)	Aims	Method	Design	Recruitment	Data collection	Relationships	Ethical issues	Data analysis	Findings	Value of research	Total score
(Adams, 2018)	Yes	yes	no	yes	yes	yes	yes	no	yes	yes	8
(Berk et al., 2019)	yes	yes	yes	yes	yes	Can't tell	yes	yes	yes	yes	9
(Bogosian et al., 2016)	yes	yes	yes	yes	yes	No	yes	yes	yes	yes	9
(Bogosian et al., 2021)	yes	yes	yes	Can't tell	yes	yes	yes	yes	yes	yes	9
(Craig et al., 2018)	yes	yes	No	yes	no	yes	yes	no	yes	yes	7
(de Wit, Vervoort, et al., 2019)	yes	yes	Yes	yes	yes	yes	yes	yes	yes	yes	10
(Dennison et al., 2013)	yes	yes	Yes	yes	yes	yes	yes	yes	yes	yes	10

(Douglas et al., 2021)	yes	yes	yes	yes	yes	yes	yes	yes	Can’t tell	yes	9
(Eccles et al., 2020)	yes	yes	yes	yes	yes	Can’t tell	yes	no	yes	yes	8
(Fitzpatrick et al., 2010)	yes	yes	yes	yes	yes	yes	Can’t tell	yes	yes	yes	9
(Giovannetti et al., 2020)	yes	yes	No	yes	yes	yes	yes	yes	yes	yes	9
(Glueckauf et al., 2012)	yes	yes	Yes	yes	yes	yes	Can’t tell	no	yes	yes	8
(Gottberg et al., 2016)	yes	yes	yes	Can’t tell	yes	Can’t tell	yes	yes	yes	yes	8
(Han et al., 2021)	yes	yes	yes	yes	yes	Can’t tell	yes	Can’t tell	yes	yes	8
(Hind et al., 2010)	yes	yes	yes	yes	yes	yes	no	yes	yes	yes	9
(Hoppes et al., 2012)	yes	yes	yes	yes	no	yes	yes	yes	Can’t tell	yes	8
(Johannessen et al., 2015)	yes	yes	yes	yes	yes	Can’t tell	yes	yes	yes	yes	9

(Johnston et al., 2016)	yes	yes	yes	yes	Can’t tell	no	yes	yes	Can’t tell	yes	7
(Kazmer et al., 2018)	yes	yes	yes	yes	yes	yes	yes	yes	Can’t tell	yes	9
(Larochette et al., 2020)	yes	yes	yes	yes	yes	no	yes	Yes	yes	Can’t tell	8
(Lavoie et al., 2005)	yes	yes	yes	yes	yes	no	no	Yes	yes	yes	8
(Marconi et al., 2016)	yes	yes	yes	yes	yes	no	yes	no	yes	yes	8
(Martin et al., 2015)	yes	yes	yes	yes	yes	no	yes	yes	yes	yes	9
(Nehrig & Chen, 2019a)	yes	yes	yes	yes	yes	yes	no	yes	yes	yes	9
(Nehrig, Gillooly, et al., 2019b)	yes	yes	yes	yes	yes	yes	no	yes	yes	yes	9
(Nehrig, Shifrin, et al., 2019c)	yes	yes	yes	yes	yes	yes	no	yes	yes	yes	9

(Pegler, 2017)	yes	yes	yes	yes	yes	yes	yes	no	no	yes	8
(Potter et al., 2021)	yes	yes	yes	yes	yes	yes	yes	yes	no	yes	9
(Sessanna et al., 2021)	yes	yes	yes	Can’t tell	yes	Can’t tell	yes	yes	yes	yes	8
(Simpson et al., 2018)	yes	yes	yes	Yes	yes	yes	yes	yes	yes	yes	10
(Simpson et al., 2019)	yes	yes	yes	Yes	Can’t tell	no	yes	no	yes	yes	7
(Swannell, 2017)	yes	yes	yes	yes	yes	yes	yes	yes	yes	yes	10
(Tahsin et al., 2021)	yes	yes	yes	yes	yes	yes	yes	yes	yes	yes	10
(Vandenberg et al., 2019)	yes	yes	yes	yes	yes	Can’t tell	yes	yes	yes	yes	9

Appendix F GRADE CERQual assessment of analytic themes and findings

Review themes and findings	Studies contributing to this finding	Methodological limitations	Coherence	Relevance	Adequacy of data	Overall CERQual assessment	Explanation of assessment decision
Adequate levels of tailoring and flexibility: Tailoring the intervention content and activities to the participants' specific disease and context was identified as important. Interventions also need to be flexible	(Adams, 2018; Berk et al., 2019; Bogosian et al., 2016; Bogosian et al., 2021; Craig et al., 2018; de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Eccles et al., 2020; Fitzpatrick et al., 2010; Giovannetti et al., 2020; Gottberg et al., 2016; Hind et al., 2010; Johannessen et al., 2015; Johnston et al., 2016; Kazmer et al., 2018; Larochette et al., 2020; Lavoie et al., 2005; Marconi et al., 2016; Martin et	**Minor concerns** Out of the 30 studies, 3 studies had some concerns with respect to methods, 22 studies had very minor concerns and 5 studies had no concerns	**Minor concerns** Evidence from the primary studies covered some aspects and diverse views about tailoring and flexibility,	**Minor concerns** Studies reported this finding across different diseases, reported by both patients and caregivers, and from	**Minor concerns** Some minor concerns about the richness of data from individual studies to support this finding.	**High confidence**	Although there were some minor concerns, this review finding was consistent across the different studies. Any discrepancies in the findings were accounted for by the different types of interventions and the different

Review themes and findings	Studies contributing to this finding	Methodological limitations	Coherence	Relevance	Adequacy of data	Overall CERQual assessment	Explanation of assessment decision
in terms of format, structure, and delivery because of individual variability in terms of symptoms, stage of disease, coping preferences and therapy preferences.	al., 2015; Nehrig & Chen, 2019a; Nehrig, Gillooly, et al., 2019b; Nehrig, Shifrin, et al., 2019c; Pegler, 2017; Potter et al., 2021; Sessanna et al., 2021; Simpson et al., 2018; Simpson et al., 2019; Swannell, 2017; Tahsin et al., 2021; Vandenberg et al., 2019) (n=30)	in terms of methods.	which were brought together in this theme.	studies that had different intervention types/ formats			levels of tailoring and flexibility that were employed.
Intervention accessibility and burden: Physical and cognitive symptoms made it difficult for people	(Adams, 2018; Berk et al., 2019; Bogosian et al., 2016; Bogosian et al., 2021; Craig et al., 2018; de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Douglas et al., 2021; Eccles et al., 2020;	**Minor concerns** Out of the 23 studies, 2	**Minor concerns** Evidence about intervention	**Moderate concerns** Different disease groups, and	**Minor concerns** Few concerns about the	**Moderate confidence**	There were some moderate and minor concerns mainly with the relevance of the finding to different

Review themes and findings	**Studies contributing to this finding**	**Methodological limitations**	**Coherence**	**Relevance**	**Adequacy of data**	**Overall CERQual assessment**	**Explanation of assessment decision**
with neurodegenerative diseases to take part in certain interventions. Additionally, participants expressed concern about the time and effort involved in taking part in some psychological interventions.	Giovannetti et al., 2020; Gottberg et al., 2016; Hind et al., 2010; Kazmer et al., 2018; Marconi et al., 2016; Martin et al., 2015; Nehrig, Gillooly, et al., 2019b; Nehrig, Shifrin, et al., 2019c; Pegler, 2017; Potter et al., 2021; Sessanna et al., 2021; Simpson et al., 2018; Simpson et al., 2019; Swannell, 2017) (n=23)	studies had some concerns with respect to methods, 17 studies had very minor concerns and 4 studies had no concerns in terms of methods.	burden was consistent across the data from primary studies.	patient and caregiver participants contributed more or less equally to this finding. The concern about the time and effort was dependent to an extent on the type of therapy and format.	richness of data and the quantity of studies supporting this finding.		intervention types and formats, but overall there is reasonable confidence in this review finding.

Review themes and findings	**Studies contributing to this finding**	**Methodological limitations**	**Coherence**	**Relevance**	**Adequacy of data**	**Overall CERQual assessment**	**Explanation of assessment decision**
Challenges with acceptance and readiness: Acceptance of disease impact and readiness for psychological support can influence engagement. Setting expectations about the intervention, being open and willing to address difficult thoughts and feelings was	(Berk et al., 2019; Bogosian et al., 2016; Bogosian et al., 2021; de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Douglas et al., 2021; Eccles et al., 2020; Fitzpatrick et al., 2010; Giovannetti et al., 2020; Gottberg et al., 2016; Han et al., 2021; Hind et al., 2010; Johannessen et al., 2015; Johnston et al., 2016; Kazmer et al., 2018; Larochette et al., 2020; Lavoie et al., 2005; Martin et al., 2015; Nehrig, Gillooly, et al., 2019b; Nehrig, Shifrin, et al., 2019c; Pegler, 2017; Sessanna et al., 2021; Simpson et al.,	**Minor concerns** Out of the 26 studies, 2 studies had some concerns with respect to methods, 20 studies had minor concerns and 4 studies had no concerns in terms of methods	**Minor concerns** Very minor concerns about data fit between the primary studies and the review findings	**No concerns** This finding was relevant to various diseases, patients and caregivers, reported with different interventions and across different countries.	**Minor concerns** Minor concerns about richness of data in primary studies and smaller samples.	**High confidence**	There were some minor concerns, but overall there is high confidence in this review finding.

Review themes and findings	Studies contributing to this finding	Methodological limitations	Coherence	Relevance	Adequacy of data	Overall CERQual assessment	Explanation of assessment decision
reported as challenging but necessary.	2018; Simpson et al., 2019; Tahsin et al., 2021; Vandenberg et al., 2019) (n=26)						
Benefits: changes in insight, perspective, self-efficacy, emotions, and relationships: A range of benefits were experienced including increased self-awareness and acceptance of the disease, seeing other perspectives	(Adams, 2018; Berk et al., 2019; Bogosian et al., 2016; Bogosian et al., 2021; Craig et al., 2018; de Wit, Vervoort, et al., 2019; Dennison et al., 2013; Douglas et al., 2021; Eccles et al., 2020; Fitzpatrick et al., 2010; Giovannetti et al., 2020; Glueckauf et al., 2012; Gottberg et al., 2016; Han et al., 2021; Hoppes et al., 2012; Johannessen et al., 2015;	**Minor concerns** Out of the 32 studies, 3 studies had some concerns with respect to methods, 24 studies had minor concerns	**No concerns** All primary studies had data that was in line with the review finding	**No concerns** This finding was covered in studies with different diseases, intervention types, and reported by	**Minor concerns** Very minor concerns, on the whole there was rich data to support the range of	**High confidence**	This review finding has good coherence, adequacy, methodological quality and relevance with data in the primary studies. Almost all studies contributed to this finding and the pattern of

Review themes and findings	Studies contributing to this finding	Methodological limitations	Coherence	Relevance	Adequacy of data	Overall CERQual assessment	Explanation of assessment decision
and developing a more positive outlook, increased sense of self-efficacy and confidence. Benefits also included an improvement in mood and relationships.	Johnston et al., 2016; Kazmer et al., 2018; Larochette et al., 2020; Lavoie et al., 2005; Marconi et al., 2016; Martin et al., 2015; Nehrig, Gillooly, et al., 2019b; Nehrig, Shifrin, et al., 2019c; Pegler, 2017; Potter et al., 2021; Sessanna et al., 2021; Simpson et al., 2018; Simpson et al., 2019; Swannell, 2017; Tahsin et al., 2021; Vandenberg et al., 2019) (n=32)	and 5 studies had no concerns in terms of methods		patients and caregivers.	benefits across the different studies.		results was consistent.

*Methodological limitations were assessed with the help of the CASP quality ratings. Studies with a score of 7 were labelled as having some concerns, scores of 8 or 9 were labelled as minor concerns, and a score of 10 indicated no concerns in terms of methods

Appendix G Intervention development process

G.1 Intervention planning table

Key issue/ barrier/facilitator to target behaviour (source)	Intervention component or section	Suggested intervention ingredients	Agreed intervention ingredients	Rationale (source)
Intervention Content				
Patients and caregivers feel that hope and positivity helps them cope with things emotionally (Pinto et al., 2021) Barrier: People might find positivity difficult because of increasing symptom burden but stressed that being positive was still important for coping and wellbeing (Pinto et al., 2021)	Section on building positivity and meaning Aim: To help users frame negative situations more positively, highlight positive aspects of experiences and help users create	– Recognizing positive things, using positive reframing to identify the positive aspects of a negative situation. – Give examples from other people with MND. Acknowledge the difficulty in doing this or reality of situation but say how positive reframing can help – Build hope and meaning by talking about the importance of values and goals, meaningful or value based goals	Building positivity section Activities in this section: Pleasant activities, Benefit finding, Values and Goals	Some evidence for the effectiveness of positive savouring or benefit finding on psychological outcomes in neurological conditions (Lai et al., 2019) and in other chronic illness found to be more useful for dealing with boosting positive affect than dealing with negative affect (De Ridder, Geenen, Kuijer, & van Middendorp, 2008). Focusing on meaning and values is important with palliative care patients (Breitbart et al., 2015) and

Key issue/ barrier/facilitator to target behaviour (source)	Intervention component or section	Suggested intervention ingredients	Agreed intervention ingredients	Rationale (source)
	or emphasize meaningful aspects of life			emphasizing meaning and goals along with positive reappraisal has been useful with older adults (Nowlan, Wuthrich, & Rapee, 2015).
Patients and caregivers find it difficult dealing with distressing thoughts about the past and future and associated negative feelings (e.g. worry, sadness) (Pagnini, 2013; Pinto et al., 2021)	Section on coping with difficult thoughts and feelings Aim: To equip users with techniques and tips to manage distressing thoughts about the past and future and associated feelings.	– Could divide into emotions (worry, depression/sadness, anger, fear, hopelessness/disappointment, frustration) and relevant strategies – Information about each emotion with relevant MND examples Examples of strategies are: – Encourage focus on the present (Mindfulness) – Strategies for dealing with thinking traps (e.g. predicting future, rumination) – Relaxation, distraction	Two sections on dealing with difficult thoughts and emotions: 1. Adjusting to changes – information and strategies to deal with anger, sadness and frustration 2. Dealing with worry and stress Emotion regulation strategies: Thought distancing (ACT), mindfulness and relaxation exercises	Some promising evidence of the effectiveness of mindfulness in improving psychological outcomes with neurological diseases (Lai et al., 2019; Pagnini et al., 2017) and some evidence of the effectiveness of ACT (Hulbert-Williams et al., 2015). CBT using goal setting and relaxation/visualisation was effective for managing anxiety in advanced cancer (Uitterhoeve et al., 2004).

Key issue/ barrier/facilitator to target behaviour (source)	Intervention component or section	Suggested intervention ingredients	Agreed intervention ingredients	Rationale (source)
Patients and caregivers express feeling tired/overwhelmed with MND and MND care, difficulty coping with the constant changes/losses (Pinto et al., 2021)	Section on dealing with stress or coping with many changes and feelings of burden Aim: To give people tools/tips to deal with stress and feelings of being burdened or overwhelmed	– Relaxation exercises (providing an escape from current situation) – Pacing self and activities – Practical suggestions for dealing with stress (e.g. taking a break, distracting self, exercising) – Encouraging self-compassion/kindness	Cover feeling overwhelmed in the 'dealing with worry and stress' section. Mindfulness exercises (e.g. safe place meditation) and strategies from compassion-focused therapy (compassion break, befriending yourself, self-kindness letter)	Mindfulness and relaxation has some evidence of effectiveness (see studies in previous row) Self compassion is helpful in improving adaptive coping (Sirois et al., 2015; Terry & Leary, 2011) and also psychological outcomes in cancer patients (Pinto-Gouveia et al., 2014).

G.2 Guiding principles

Key issue/barrier	Design objective	Key features of intervention
Intervention Tone and Presentation		
People with MND have different levels of disability, particularly with using their hands and voice, difficulties with cognition, and symptoms and ability deteriorate rapidly	To make the intervention easy to use and navigate for people with MND with varying levels of disability	– Make sure suggested techniques or tips take into account varying levels of disability and offer alternative suggestions. – Reassure people that they can modify tasks/suggestions and that there is no best way of doing things – Navigation should be simple and easy to understand, and activities/exercises should not involve too much typing (only clicking)
Difficulty finding hope and positivity because of no cure and progressive nature of disease (Pinto et al, 2021)	To make the intervention tone positive and empowering	– Emphasis on what CAN be done to deal with difficult thoughts and emotions – Make sure language used is optimistic but also realistic – Use messages of positive reinforcement at different stages – Not to sound confronting about death/future symptoms

Key issue/barrier	Design objective	Key features of intervention
People talk about the usefulness of being understood and empathy, from professionals and friends and family (Pinto et al, 2021)	To demonstrate empathy with people with MND and the difficulties associated with coping with MND	– When making suggestions of techniques or activities, acknowledge difficulties/effort with doing them – Make sure examples and information are MND specific – Provide quotes from other people with MND and caregivers to accompany information and strategies
People are already burdened with the effort of living with MND and MND care (Pinto et al, 2021, 2022)	The intervention should not be an additional burden for people with MND and carers	– Intervention should not have too much information or reading to reduce cognitive effort – Intervention should not seem like a chore – no prescribed way of using it, no fixed sessions or tasks. If there are exercises or tasks to try out on their own, these should be simple to do and there should be an option not to do them for people who don’t want to – Intervention should not involve the mastery of difficult skills, instead information and activities should be short and easy to do

Key issue/barrier	Design objective	Key features of intervention
People may have different psychological issues that they are dealing with at different stages (e.g. at diagnosis or later coping with new losses). The way people cope/preference for coping with these issues are also highly individual. (Pinto et al, 2021, 2022)	To be flexible to use at different points of the disease, for different psychological issues and for people with different preferences for coping	– Allow people to choose content based on what is most relevant to them – Emphasize that people cope differently and there is no 'right' way – Give people options of different techniques/different practical ways to handle the situation/emotion – Cover a variety of issues that people go through at different stages – Provide links to additional psychological support in case people need this

Appendix H Description of the Coping And Living well with MND (CALM) website according to the Template for Intervention Development and Replication (TIDieR) checklist

1. Brief name: CALM website - a self-guided psychological support website for MND
2. Why: There is limited evidence for psychological interventions for MND and a strong need to develop interventions to support people with people with MND and caregivers. Online support via a website could offer an accessible way of providing some psychological support. The goal of the CALM website was to reduce psychological distress and improve wellbeing for people with MND and caregivers.
3. What: The CALM website contained information and self-help strategies that was presented in 5 main sections – building positivity, adjusting to changes, dealing with worry and stress, other support, and all activities. Each section presented information or practical tips or self-help strategies or quotes from peers (see Chapter 1.7.4 for more details).
4. Procedure: The user could choose to visit any of the website sections and select information or activities that were relevant to them. Some activities required listening to audio clips or thinking through situations or thoughts or values; whilst other activities served as prompts for people to do more meaningful activities in their daily life or engage in activities that might help them feel better.
5. Who: The link to the CALM website was sent directly to participants by the researcher, and the study was introduced through the Motor Neurone Disease Association (MNDA) charity.
6. How: The entire intervention was online and meant to be used individually by either people with MND or their caregivers. It was a self-guided website, only technical support was provided by the researcher if needed.
7. Where: The CALM website could be accessed by individuals in any location that was convenient to them, this would typically be at their own home. Participants needed access to the internet and a device such as a computer, tablet or mobile to use the CALM website.

8. When and how much: As this was a study exploring the use of the intervention, participants with a range of symptoms, different levels of psychological distress, and at different stages of MND took part. There was no specific intervention dose or guided sequence, and users could choose how much or how often to refer to the website and when to use the strategies and tips. All participants had access to the CALM website for 6 weeks.
9. Tailoring: The information and strategies presented in the website were tailored to be relevant to the needs of people with MND and caregivers and framed in an accessible way for people with different symptoms and levels of ability. Participants self-tailored their use of the CALM website to their own needs and preferences.
10. Modifications: Modifications were made based on think-aloud interview feedback and presented in an iterative way to participants to improve and refine the CALM website. No further modifications were made for the next study as participants used the CALM website for 6 weeks on their own.
11. How well (planned and actual): The CALM website was used in a variety of ways (including no use) based on whether participants thought the information and strategies were relevant to their situation (i.e. symptoms, disease stage), whether they felt ready to try out the advice and strategies, and whether the strategies matched their coping preferences. These findings have been presented in Chapter 4 (paper 3).

Appendix I Demographics questionnaire

1. Age: ______________ years

2. Gender:

   ☐ Man ☐ Woman ☐ Prefer not to say

   ☐ I identify in a different way (please specify_____________________)

3. Education:

   ☐ Up to GCSE or equivalent ☐ A-levels or equivalent

   ☐ Graduate or equivalent ☐ Postgraduate

   ☐ No formal education ☐ Other (please specify): _______________

4. Employment:

   ☐ Full-time employment ☐ Retired

   ☐ Part-time employment ☐ Other (please specify) ________

5. Ethnicity:

   ☐ White British/Irish/Other

   ☐ Black/African/Caribbean/Black British

   ☐ Mixed/multiple ethnic groups

   ☐ Asian/Asian British

   ☐ Other ethnic group: ________________

6. Do you live alone?

   ☐ Yes ☐ No

7. Do you have someone who helps and supports you, who is not a healthcare professional?

   ☐ Yes ☐ No

8. Do you receive support from a professional carer?

   ☐ Yes ☐ No

9. Where do you live in the UK?

   ____________________________________ (City/town)

Please fill in the information below about your MND. Please do not worry about the list of problems, some of these may not be relevant to you, but try to fill in the information as accurately as possible. We have created these lists to make sure we hear from people with different symptoms and different experiences.

10. What is your specific diagnosis?

☐ ALS (Amyotrophic lateral sclerosis, most common form of MND)

- Did your symptoms start in your arms or legs? Yes/No
- Did your difficulties start with swallowing or speech? Yes/No

☐ PBP (Progressive Bulbar Palsy)

☐ PMA (Progressive muscular atrophy)

☐ PLS (Primary lateral sclerosis)

☐ Other (please specify): __________________

11. How long ago were you diagnosed with MND? If you cannot remember exactly, please estimate or give the approximate date of diagnosis.

    ________________ years and ____________months ago

12. Have you been given a formal diagnosis of cognitive impairment or dementia?

☐ Mild cognitive impairment ☐ Dementia ☐ Neither of these

13. Do you have any concerns about your ability to remember things, pay attention, plan or problem solve?

☐ Yes ☐ No

If yes, could you please specify what difficulties you have experienced?

____

14. Have your friends/family had any concerns about your ability to remember things, pay attention, plan or problem solve?

☐ Yes ☐ No

If yes, could you please specify what concerns your friends and family have expressed? ____

[Insert either ALSAQ-40 or ZBI based on participant]

Please answer a few more questions about the research study: [only for people with MND]

1. Could you please indicate who filled this questionnaire?

☐ Me, the person with MND without any help

☐ Me, the person with MND with someone else's help

☐ Somebody else filled it on my behalf

2. Roughly, how long did it take you to fill in the questionnaire?

______________ minutes

3. Did you find the questionnaire too long or burdensome?

☐ Not at all
☐ A little
☐ Somewhat
☐ A lot

4. In this study, you will be looking at a website and working through some webpages. Will you be able to do this:

☐ On your own without any help

☐ With someone's help

☐ With the help of some form of technology (If you have ticked this option, what device/technology will you be using? ___________________________________)

Appendix J Study 1 Think aloud interview topic guide

J.1 Questions about the website introduction

- What do you think about the title of the website?
- (On each page of this section) What are your thoughts about this page?
    - Is the information clear?
    - Was there anything you didn't understand?
- (At the end of the introduction section) What did you think about the introduction?
    - Was anything unclear?
    - Is there anything else you would've liked to know at this stage?

J.2 Questions for each page of the website

(ask 2-3 questions from the list based on the particular page and the participant's navigation/response)

- What are your first impressions of this page?
- What are you thinking now?
- What option would you choose?
- What made you choose that option?
- What do you think about this information/activity/idea?
- Is there anything you like/don't like about this page? What is it about that, that you like/don't like?
- I noticed you (smiled/frowned/hesitated). Can you tell me what you thought about that?

J.3 Questions at the end, after going through the website

- Overall, what did you think about the website?
- Can you tell me anything you particularly liked about the website?
- Can you tell me about anything you disliked or were less keen on?
- Having gone through the website, can you tell me how you feel about using this website?
- Are there any things we need to keep in mind/pay attention when we are designing websites like this for people with MND or family members/caregivers?
- Is there anything else you would like to mention about the website?

Appendix K Table of changes

K.1 Extract from table of changes

Location in intervention	Negative comments/ suggested changes	Positive comments	Possible change	Reason for change	Agreed change	MoScow (Must, Should, Could, Would)
Homepage	Signpost people to additional activities, make this as an introduction to mental wellbeing for people with MND and then they can try other activities to deepen their understanding. Because people may just skim the website and you don't want people don't feel this is all the		Potentially can add a section on the homepage if people need additional support. This could include both online resources or how to	EAS	Added a section directing people to resources for professional and peer support	Could

Location in intervention	Negative comments/ suggested changes	Positive comments	Possible change	Reason for change	Agreed change	MoScow (Must, Should, Could, Would)
	support or all the techniques available (F4)		access face-to-face psychological support			
Navigation	It would be good to have a link to the 'All techniques' there, rather than going to the main menu (F3, 53:22) Minimizing the amount of clicks is important if you are using an eye gaze system, so it's less frustrating, especially if they are asking someone to help them (P7, 54:00)	The rest of your website is good for eye gaze because the font size is big and easy to read, not overcrowded and the buttons are big to click (P7, 55:27)	Wherever possible, if I have mentioned something from another section, add a link to it	IMP – in line with guiding principles	Hyperlinks added throughout website to make navigation easier	Must
Section on Anger	It sounds patient centric rather than carer centric (F7, p2 6:35)		Change wording to focus on patient and carer experience	IMP, REP	Changed tense to 'you' to make it relevant to both	Must

Location in intervention	Negative comments/ suggested changes	Positive comments	Possible change	Reason for change	Agreed change	MoScow (Must, Should, Could, Would)
					patients and caregivers. Added caregiver quote on this page as well	
Practical tips	Physical activity may be difficult for some, instead saying ‘going out in nature’ Going into a park or looking at a tree, nature takes away anger (F4)	Useful to have something to take away and apply in your life (F2)	Say ‘going out into nature for exercise or to take a break’	IMP (in line with our guiding principles)	Rephrased to ‘getting some fresh air’ and ‘spending time with nature’	Must

Location in intervention	**Negative comments/ suggested changes**	**Positive comments**	**Possible change**	**Reason for change**	**Agreed change**	**MoScow (Must, Should, Could, Would)**
Stress and feeling burdened section	Feeling burdened is not clear, it's quite a negative word. Feeling overwhelmed might be better (F6, 22:13)		Change title to 'feeling overwhelmed'	EAS, NCON	'Stress and feeling overwhelmed'	Should
Pleasant activities	It may be helpful to say 'when you are planning your list, it may be helpful to discuss with your friend or family member' so they can help with any preparations or adaptations (F3, 1:24:28)		Added a phrase saying it might be helpful to include family members if you need any help with these activities	EAS, IMP	Added a phrase saying it might be helpful to include family members if you need any help with these activities	Should
Benefit finding	It's more about positive thinking or positive mindset than benefit finding.		Consider changing name of activity, currently confusing	REP	Name changed to 'Finding positives'	Must

Location in intervention	Negative comments/ suggested changes	Positive comments	Possible change	Reason for change	Agreed change	MoScow (Must, Should, Could, Would)
	Benefit finding sounds like looking for disability benefits (F6, 39:34)					

[*] IMP – Important change; EAS – easy and uncontroversial; REP – repeated by many participants; NC – Not changed (explain why);

EXP – supported by experience (PPI or experts or literature); NCON – does not contradict evidence, logic model or guiding principles

K.2 Summary of key changes made from think-aloud interview feedback

Summary of issue identified	Example quote/s	Change implemented
Perceived as less applicable or not aimed at family members	It sounds patient centric rather than carer centric. (F7) When you first read it seems more aimed at the person with MND. (F1)	A quote from a family member was added on each page where there were quotes. The wording describing the problem or activity was changed so that the information could be seen as relevant to both people with MND and family members.
The support from the website was limited to self-help activities and advice and people may be in a situation where they need more help or support.	Signpost people to additional activities to deepen their understanding. Because people may just skim the website and you don't want people to feel this is all the support or all the techniques available (F4)	An 'other support' section was added which directed users to resources for professional support, peer support and other useful websites and apps for psychological support.
Navigation between different sections and activities is difficult when activities are suggested but you have to go back and find where they are	It would be good to have a link to the 'All techniques' there, rather than going to the main menu (F3) Minimizing the amount of clicks is important if you are using an eye gaze system, so it's less frustrating, especially if they are asking someone to help them (P7)	Direct links to specific activities or pages were provided to reduce the number of clicks and make navigation easier.
Introduction is too long and information is repeated	'cause I feel like I've spent a few, a little bit of time navigating my way to this point, whereas if I just clicked the video and someone just told me this. If I'm struggling with using my fingers or my concentration isn't great and I'm coming across this for the first time, I may lose a bit of energy by that stage. (P3)	The number of pages in the introduction were significantly cut down and there were options to click to get to pages where there was optional information (e.g. details about who created the website). An additional audio clip summarising key information was provided to make it more accessible.
Some things can trigger sense of loss either for the past or an anticipated loss (e.g.	I look at the picture of the person in the wheelchair and	Did not include pictures of people with MND, only nature images in the website.

Summary of issue identified	Example quote/s	Change implemented
photographs of people with MND with certain symptoms, lists of pleasant activities)	think 'Oh that's what I've got coming at some point.' (F1)	Did not list activities or suggestions, instead provided a case study if we needed to communicate a concept via an example.
Terminology is not clear, specifically for the titles 'Building positivity and meaning', 'benefit finding' and 'compassionate letter writing'	With benefit finding, my first thought is it's where you get benefits (F3) Compassionate letter writing sounds like what they do in the hospital, when they write letters to loved ones of persons that have passed away (P9)	Changed titles to: 'Building positivity' 'Finding positives' 'Self-kindness letter'
The benefit to doing these activities is not communicated and people may not be motivated to trying out the activity	It doesn't say anywhere why this is being done? Why is it important? You could say more about the benefits of doing this (P4)	In the overall introduction and introduction of the activities, we suggested how doing the activity could help with improving mental health and their relevance to MND.
Some wording was not sensitive to people who may have different levels of ability and different symptoms	Physical exercise may be difficult for some, instead saying 'going out in nature' or 'going into a park' (F4)	Suggestions or tips that could not be carried out with people with higher levels of disability were either removed or reworded in a more accessible way.
Some examples and activities needed to be worded in a more realistic way for MND	And it seems slightly unrealistic to say 'OK, he sat down, he did all this, had a cup of tea and carried on,' you think, 'well that's not gonna happen!' You know, because it's something you'd have to do repeatedly to learn the technique or to get into a frame of mind that you could do it that simply. So to me, I would read that and go someone just made up (P1)	The wording of some activities and examples were altered to reflect a more realistic scenario where people had to practice activities to experience benefit. Overly positive statements (e.g. suggestions in the 'finding positives' activity) were removed or reworded so that they reflected the reality of living with MND

Appendix L Study 2 Interview Topic Guide

L.1 Opening question

- To start with, can you just tell me a bit about how you've got on with trying out the CALM website.

L.2 Questions about overall website use

- Can you tell me about when you used the website?
- Can you tell me a bit more about how often you logged in and what made you decide to log in?
- Can you talk me through the sections/activities you looked at? How did you decide to choose them?
- Were there any sections/activities that you did not look at? Or that you used least in the whole website? Can you tell me a bit more about that?
- Can you tell me about whether you had a go at trying any of the techniques or suggestions from the CALM website? How did that go?
- Could you tell me about any part of the website that didn't seem to work properly or that you had any problems with?
- (Based on the website usage data) I can see that you often looked at X section, or did not look at X section, or logged in X number of times. Can you tell me a bit more about this?

L.3 Questions about each section

(ask questions after looking at usage metrics, which sections/activities were used, how often)

- **What did you think of** (5 sections - the introduction, building positivity and meaning, adjusting to changes, dealing with worries and stress, all activities)
    a. Can you describe whether this section felt relevant to you?
    b. Can you describe anything you liked about this section?
    c. Can you tell me about anything you disliked about this section?
    d. Was there anything that was useful to you in this section?
    e. Was there anything that was not useful to you in this section?

f. Can you tell me about anything that you would change? Or anything that didn't make sense to you?
g. How did you find the activities or suggested techniques? [name and summarise the specific techniques here]

L.4 Questions about dealing with emotions after using the CALM website

- After having used the website for some time, how do you now feel about dealing with your emotions?
- Can you tell me about any advice or technique from the website that you think you might use now, or in the future?
- Has anything changed with you since you have been going through the website?
- How has it been for you using this website during all the disruption and limitations we've had because of COVID 19?

Is there anything else you would like to mention about the website/advice and activities?

Appendix M GRIPP2 checklist for PPI role and contribution

Section and topic	Item	Description and reflections
Aim	Report the aim of PPI in the study	PPI were recruited to share their perspective and help shape the research questions and methods in the 3 studies of this thesis. Recruiting PPI members took longer than anticipated and this was done within the scope of a PhD project. So the overall research questions and the design of the first study in this thesis was carried out without PPI input.
Methods	Provide a clear description of the methods used for PPI in the study	PPI members were recruited on an ongoing basis due to the progressive nature of MND impacting levels of involvement in PPI activities. A combination of group and individual meetings, along with email and phone communications were used throughout the PhD to involve PPI members and provide project updates.
Study results	Outcomes – report the results of PPI in the study, both positive and negative	PPI members were helpful in sharing their perspective and offering suggestions for data collection and recruitment procedures. They reviewed initial drafts of the intervention and provided valuable feedback which was used to improve the clarity and navigation of the intervention. They also gave their input on the preliminary findings from paper 1 and paper 3.

Discussion and conclusions	Outcomes – comment on the extent to which PPI influenced the study overall. Describe positive and negative effects	PPI members helped ensure that the recruitment and data collection procedures were as smooth and easy as possible for participants, particularly for those with difficulties with speech. Their perspective on the results of the studies drew my attention to areas I was not highlighting in the papers. They were a constant support reminding me of the value of the project and validating some of my findings by showing me how they compared to their own experiences.
Reflections and critical perspective	Comment critically on the study, reflecting on things that went well and those that did not	I was unable to include all PPI members at all the different stages of the studies due to short timelines. I did not get a lot of engagement from PPI members who were caregivers. Having a PPI panel review and endorse my intervention gave it some legitimacy to participants in studies 3 and 4. Having flexible methods to communicate with PPI members and having studies that fed into each other sequentially, allowed different PPI to get involved at their convenience at different stages of the overall project.

Appendix N Specific changes to refine the CALM website for future use based on participant feedback from Paper 3

Specific aspect of the CALM website	User Feedback	Suggested improvement(s)
Navigation and ease of use	Most people found the website easy to use and navigate. No technical difficulties were reported. Few people found it difficult to remember what section they had last looked at, or what activities they had already tried out.	Could have a bookmark function or indication of what section/activity has already been viewed
Use of multimedia & text	The audio and pictures were useful and calming. People said there was a lot of reading involved, which can be difficult particularly if stressed or experiencing difficulties with concentration. More audio/video content was suggested to overcome this.	Adding more audio/visual content, especially for the quotes from peers or explanation of concepts would be beneficial.
Number of activities available	People liked that the activities were quick and easy to do. People wanted more/different activities to keep it fresh and avoid repetition.	Add more activities, or variations of the same type of activity (e.g. different mindfulness activities)
Adding information about certain topics	Suggestions were made to add information about the following topics - dealing with loss and grief, being a burden to the caregiver, dealing with diagnosis, practical ways to adapt to changes in symptoms and new equipment.	These are all important topics that can be added. However, they may need to be piloted with target users first.

Applicability at different stages and for different levels of physical ability	People said the information on the website was sensitive to the needs of people with MND and caregivers. Some suggested changing some of the phrasing to make activities a little more appropriate or having more stage-based tailoring.	The final version of the website needs to be scanned to make sure all activities are presented in an accessible way. Stage-based tailoring may not be helpful as there are no clear stages with MND. However, more information about dealing with diagnosis can be added and tailoring activities and information to levels of physical ability can be explored.
Other support section	Most people knew of the other support options and said they were good resources to signpost people to. Some suggested adding information about caregiver charities and how to go about finding a therapist/psychologist.	Could add information about caregiver charities. Guidance around finding a therapist may be tricky to provide. Having a list of therapists and psychologists who have knowledge and experience working with MND would be useful.

Appendix O Link to dataset of interviews conducted as part of this PhD

Data in the form of anonymised interview transcripts can be made available on request from the University of Southampton repository by emailing c.l.pinto@soton.ac.uk or researchdata@soton.ac.uk (https://doi.org/10.5258/SOTON/D2652)

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