



# Amyotrophic Lateral Sclerosis and Frontotemporal Degeneration

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## RESEARCH ARTICLE

# Needs and preferences for psychological interventions of people with motor neuron disease

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## Abstract


**Background:** There is a lack of knowledge about what factors may impede or facilitate engagement in psychological interventions in people with motor neuron disease (pwMND) and how such interventions can be adapted to best meet the needs of this population. **Objectives:** To explore the needs and preferences of pwMND with respect to psychological interventions, and how best to adapt such interventions for pwMND. **Methods:** A series of semi-structured interviews ( $n = 22$ ) and workshops ( $n = 3$ ) were conducted with pwMND ( $n = 15$ ), informal caregivers of pwMND ( $n = 10$ ), and MND healthcare professionals ( $n = 12$ ). These explored preferences and concerns that would need to be considered when delivering a psychological intervention for pwMND. Three areas were explored: (i) perceived factors that may hinder or facilitate pwMND engaging with psychological interventions; (ii) ways in which such interventions could be adapted to meet the individual needs of pwMND; and (iii) views regarding the main psychological issues that would need to be addressed. Workshops and interviews were audio recorded and transcribed and thematic analysis was used to inductively derive themes. **Findings:** Data could be classified within four overarching themes: unfamiliar territory; a series of losses; variability and difficulty meeting individual needs; and informal support. **Conclusions:** Flexibility, tailoring interventions to the individual needs of pwMND, and encouraging autonomy are key attributes for psychological interventions with pwMND. Psychological interventions such as Acceptance and Commitment Therapy (ACT) could be acceptable for pwMND if adapted to their specific needs.

**Keywords:** Motor neuron disease, psychological interventions, qualitative research

## Introduction

Average life expectancy for people with motor neuron disease (pwMND) is 3–4 years after symptom onset and the disease progression is variable (1). As the onset of symptoms associated with motor

neuron disease (MND) and its speed of progression are unpredictable, it can be difficult to anticipate future support needs. With no known treatment to stop or reverse disease progression, those diagnosed are required to continually adjust to new losses (2).

 Supplemental data for this article can be accessed [here](#).

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This can have a negative effect on emotional well-being and self-esteem, as indicated by high prevalence rates of depression and anxiety (44 and 30%, respectively (3,4)) and greater risks of suicide in pwMND than in the general population (5,6).

Psychological interventions may be of benefit for those psychological distresses; however, there is insufficient evidence to support their effectiveness at present (7). Numerous barriers may interfere with engagement in psychological interventions, including: rate of deterioration in symptoms; hostile reactions from families and caregivers; belief that the diagnosis is incorrect; and perceptions that the therapeutic approach is not appropriate (8). Furthermore, those with communication difficulties may require the support of a communication device (e.g. light writer or Eye Gaze). Though these have great potential to improve the lives of those who experience communication difficulties, they can also present technical barriers if faulty, incorrectly set up or running low on battery (9) and could potentially interfere with therapeutic delivery. Additionally, cognitive and behavioral changes present in up to 30–50% of pwMND (10–12) may impede engagement in psychological interventions. For example, executive dysfunction could reduce the ability to follow therapeutic discussions that involve abstract thinking such as thinking about the future.

Research on attitudes and preferences of pwMND toward treatment have primarily focused on decision making with respect to medical interventions, such as noninvasive ventilation and percutaneous endoscopic gastrostomy (13), as well as assistive technology and home adaptations (14). Perceptions of control, reassurance, and trust have been found to be important in the ability of pwMND to engage with healthcare services (15). Nevertheless, how or why pwMND choose to accept and engage with psychological interventions has not been reported. Consequently, there have been calls for further research to understand how healthcare services can meet the psychological needs of pwMND (13).

At present, there is a lack of knowledge about individual preferences for psychological interventions among pwMND, what factors may impede or facilitate engagement in such interventions, and how psychological interventions can be adapted to best meet the needs of pwMND. Consequently, the aim of this study was to explore the needs and preferences of pwMND with respect to psychological interventions, and how such interventions can be adapted for this population.

## Methods

### *Inclusion/exclusion criteria*

Participants were aged 18 and over and had a diagnosis of definite, probable or laboratory-supported

probable familial or sporadic amyotrophic lateral sclerosis (ALS) using the World Federation of Neurology's El Escorial criteria (16), or variants of MND including Primary Lateral Sclerosis and Progressive Bulbar Palsy. Those with a self- or informant-reported diagnosis of dementia or lacking capacity to consent to participate in the study (as assessed by a member of the research team using standard research procedures) were excluded. Caregivers were required to be a current or former informal caregiver of a person with MND. Lastly, healthcare professionals were required to have current or previous experience of working with pwMND. All participants who took part provided fully informed written consent.

### *Recruitment of participants*

Potential participants, who met the inclusion/exclusion criteria, were identified and approached by clinicians through the UK Motor Neuron Disease Clinical Studies Group (a network of MND clinicians involved in research studies in the UK; <http://www.mndcsg.org.uk>), or responded to leaflets, posters or online advertisements *via* the Motor Neurone Disease Association, local community support groups and the Sheffield MND Research Advisory Group (a Patient and Public Involvement group). Purposive sampling was conducted as much as possible to select participants with a range of characteristics that might be expected to influence attitudes toward psychological interventions for pwMND (sex, time since diagnosis, caregiver relationship, and clinical profession). Recruitment continued until data saturation was achieved and new data were easily accommodated within the themes.

### *Data collection*

Demographic data were collected at screening (see Table 1). In-depth semi-structured interviews and workshops (i.e. a discussion group facilitated by a member of the research team) were conducted with pwMND ( $n=15$ ), informal caregivers of pwMND ( $n=10$ ), and healthcare professionals who work with this population ( $n=12$ ). The number of participants was deemed appropriate in order to address the research question and achieve saturation (17). Interviews ( $n=22$ ) and workshops ( $n=3$ ) were either conducted face-to-face (in participants' homes or in a meeting room at University College London [UCL]), or *via* telephone, individually or in a group, and verbally or *via* written responses, depending on each participant's preferences, communication needs, and geographical location.

Interviews and workshops were conducted by KW ( $n=21$ ) and RG ( $n=4$ ). The interviewers used a semi-structured topic guide for the

Table 1. Demographic characteristics of participants.

	pwMND (N = 15)	Caregivers (N = 10)	Healthcare professionals (N = 12)
Sex			
Male	9	3	3
Female	6	7	9
Age			
30–49	0	1	8
50–64	7	3	4
65+	8	6	0
Mean age (SD)	61.4 (SD 5.94)	64.7 (SD 11.94)	48.0 (SD 8.86)
Ethnicity			
White British	15	10	11
Other	0	0	1
Education			
GCSE/O levels	1	2	0
A level	1	1	0
Bachelor's degree	8	5	7
Master's degree	5	1	3
Doctorate	0	1	0
Employment status			
Paid work	3	2	12 (MND Nurse specialists n = 6, clinical consultants n = 2, speech and language therapists n = 2, counselor n = 1, physiotherapist n = 1)
No paid work	4	2	–
Voluntary	2	3	–
Retired (due to age)	6	3	–
Marital status			
Single	1	0	n/a
Married	12	5	n/a
Widowed	0	5	n/a
Divorced/separated	2	0	n/a
Diagnosis of pwMND/person cared for			
ALS	10	9	n/a
Primary lateral sclerosis	4	0	n/a
Progressive bulbar Palsy	1	0	n/a
Progressive muscular atrophy	0	1 <sup>a</sup>	n/a
Mean no. of years working with pwMND	n/a	n/a	10.4 (SD 7.37)
Mean no. of months caring for a person with MND	n/a	26.5 (SD 12.97)	n/a
Mean no. of hours per week caregiving	n/a	41.8 (SD 38.10)	n/a

SD: standard deviation.

aA former carer of a person with MND also had a recent diagnosis of progressive muscular atrophy, but preferred to answer the questions from the perspective of a carer as they considered themselves to be too early in the disease course to comment from the perspective of a person with MND.

interviews and workshops which were used flexibly, following the concerns of participants as they arose (see [Supplementary Table 1](#)). Topics explored included (i) facilitators/barriers to engagement in talking therapy for pwMND; (ii) general adaptations to talking therapy for pwMND; (iii) psychological or emotional issues for pwMND; and (iv) promoting talking therapy to pwMND. Each observed 1–2 of the other's interviews and workshops to ensure consistency. The interviewer encouraged participants to identify priorities for discussion and to use examples to illustrate their concerns. Interviews lasted approximately 45 mins (range 22–94 min), while workshops lasted approximately 65 min (range

56–72 min). All participants received a £10 voucher upon completion.

#### Data analysis

Interviews and workshops were audio recorded, transcribed verbatim, and anonymized to maintain confidentiality. Interview notes, including impressions of key topics and how participants found the interviews and workshops, were used to contextualize the analysis. Using NVivo version 11 to help manage the data, inductive thematic analysis (18) was conducted to develop a theoretical framework around the needs and preferences of pwMND with respect to psychological

interventions. Constant comparison (19) was used to delineate similarities and differences between lower level codes and grouped into themes. The themes were then verified and refined as the data collection and analysis progressed. KW and JL read and coded all transcripts, which was then checked by VL. Data are presented across the participant groups, and similarities and discrepancies are highlighted where relevant.

### *Ethical considerations*

The study was approved by the UCL Research Ethics Committee (ref. 12213/001).

## **Results**

### *Demographics*

As shown in Table 1, pwMND had a mean age of 61.4 years (range 56–75 years) and most were male ( $n = 9/15$ ). Self-reported diagnoses included ALS ( $n = 10/15$ ), Primary lateral sclerosis ( $n = 4/15$ ) and progressive bulbar palsy ( $n = 1/15$ ). The majority communicated verbally ( $n = 13/15$ ). With respect to caregivers of pwMND, half were current caregivers ( $n = 5/10$ ). They had a mean age of 64.7 years (range 37–77 years) and were mostly female ( $n = 7/10$ ). The majority were spouses of pwMND ( $n = 8/10$ ). Finally, the sample included a mix of healthcare professionals (see Table 1). They had a mean age of 48 years (range 31–64 years) and were mostly female ( $n = 9/12$ ). On average, they had worked with pwMND for 10.4 years (range 1.5–28 years).

### *Study findings*

Data were organized into four overarching themes: (i) unfamiliar territory; (ii) a series of losses; (iii) disease variability and difficulty meeting individual needs; and (iv) informal support. Table 2 presents these themes and sub-themes with example quotes from participants. Each of the themes has implications for engagement in psychological interventions, which are presented in Table 3.

### *Unfamiliar territory*

All participants across the sample felt that there is a lack of understanding and knowledge about the disease among the general population and particularly non-MND healthcare professionals, often explaining this by describing MND as a “rare disease”. Societal awareness of MND was strongly considered to be lower than for other life limiting conditions, with cancer a common reference point. PwMND reported a tendency for comparisons to be made with Stephen Hawking, while others were acutely consciously of the stigma associated with MND, noting that friends and family were often

reluctant to discuss the condition. The perceived lack of understanding about the disease was felt to compound feelings of isolation. Individuals indicated that they would value the opportunity to speak openly about living with MND with healthcare professionals who have sufficient knowledge of the illness.

A common view among pwMND and caregivers was that MND healthcare professionals did not provide adequate information regarding the condition or sources of support at diagnosis. However, others explained that they had received adequate literature, but actively avoided it as they did not feel ready to accept the diagnosis at that time. Participants across the sample acknowledged that healthcare professionals may struggle to find the right time to approach pwMND about engaging in psychological interventions due to the prevalence of denial and avoidance of the prognosis. Some pwMND and caregivers felt they would be more likely to engage in psychological services which were recommended by people with the same condition.

A number of female caregivers and healthcare professionals felt there may be additional difficulties engaging men in psychological interventions, suggesting that men in particular can struggle due to losses in status and threat to their role of “breadwinner”. Equally, it was acknowledged that this only increased the need for psychological support in this group.

A recurring theme across participant groups was the challenge of forging a new identity as someone with MND, especially in the absence of clear information or advice about the condition.

### *A series of losses*

A recurring theme was the sense of loss experienced by pwMND, which was evident in multiple domains, including physical functioning, mobility, independence, future hopes and dreams, social relationships and identity (see Table 2). It was noted that people experience different losses at different time points due to the variability of symptoms experienced in MND. Yet the losses experienced across these domains were commonly considered to reflect a loss of control over their lives and the pwMND felt they had no choice but to accept them. Some of the participants discussed living a life within the constraints of what is possible and adapting to their diagnosis. Nevertheless, it was apparent that growing fears and anxieties regarding the prognosis made it difficult for many pwMND to live in the present moment.

Anger, frustration, uncertainty, and hopelessness were strongly associated with the sense of loss in MND. Feelings of embarrassment were commonly reported and were most prevalent amongst those with speech and communication difficulties.



Table 2. A summary of the identified themes, sub-themes, and example quotes.

Themes		Quotes
Unfamiliar territory	(1a) Lack of MND awareness	<p>"It can be quite an isolating disease, people don't know much about it, people don't know what to say. It's not like cancer that lots of people have said oh, you know, my such-and-such relative had this. There's a lack of knowledge about it." (Person with MND, 014, F, 67 years)</p> <p>"I do think it has the kind of stigma that people don't want to talk about it". (Healthcare professional, 054, F, 48 years)</p>
	(1b) Value placed on healthcare professionals' knowledge	"... any therapist who goes in needs to have a basic understanding of what is available to a motor neuron disease patient to just help them". (Caregiver, 029, F, 74 years)
	(1c) Finding the right time	"You might find that people turn you down in the first instance. It's being able to keep plugging away, and also, sort of, try to be there at the right place, right time, really." (Person with MND, 026, M, 63 years)
	(1d) Value placed on testimonies from others with MND	"For me recommendations and testimonials from people who've already done it and seen what their views were and that it helped them, then I would think it's worth a go for me". (Person with MND, 014, F, 67 years)
	(1e) Difficulties accepting help	<p>"I just think men are difficult to work with in my experience. You know they don't like to talk about the problems, they like people to think they're very brave, they don't complain so much you know". (Caregiver, 030, F, 74 years)</p> <p>"MND patients, I find a lot of them have problems with accepting it. We see them right from the beginning, they're quite in denial. They'll be, like, oh, at least they're not as bad as so-and-so, trust me. They don't want to read the literature and things like that". (Healthcare professional, 015, M, 46 years)</p>
	(1f) Navigating a new identity	<p>"And seeing yourself as a disabled person it's a real wakeup, as I say, you just get a new identity really". (Person with MND, 002, F, 60 years)</p> <p>"I live with MND. That's how I use that. People say "You got MND." So I say "No. I live with MND" (Person with MND, 003, M, 75 years)</p>
A series of losses	(2a) Feelings of loss (loss of function, independence, hope, relationships, identity)	<p>"It's very hard to have people doing things for you, you know and of course the loss of dignity is a big thing as well. You have to have help going to the toilet and getting washed and things. That's a huge loss of dignity ...". (Caregiver, 030, F, 74 years)</p> <p>"... just the simple loss of opportunity, things that may not be fulfilled, things that have been put in towards retirement which will never happen". (Healthcare professional, 016, M, 44 years)</p> <p>"I think also we found that some of our friends have fallen away. We talk about loss". (Caregiver, 023, F, 74 years)</p>
	(2b) Psychological impact of losses (anger, frustration, hopelessness, embarrassment)	<p>"... they get angry and they find it hard to discuss, but they do when you get to know them, they get angry because they see people who have, they would say, frankly, abused their health, and, and they're not getting MND". (Healthcare professional, 046, F, 60 years)</p> <p>"They find it embarrassing even if it's only in front of family. I mean, there are very few of us, as much as we love our nearest and dearest, would want them to be wiping our, you know, spit off our chins". (Healthcare professional, 040, F, 64 years)</p>
	(2c) Coping strategies (denial & avoidance)	<p>"MND patients, I find a lot of them have problems with accepting it. We see them right from the beginning, they're quite in denial. They'll be, like, oh, at least they're not as bad as so-and-so, trust me. They don't want to read the literature and things like that". (Healthcare professional, 015, M, 46 years)</p> <p>"So some people have said, I don't want to meet with other people with MND, I don't want to see what I'm going to become. And that's the risk, isn't it, if you meet people with the same illness who are further on, and you think, I wish I hadn't seen that". (Healthcare professional, 034, F, 31 years)</p>
	(2d) Need to empower patients	<p>"It might be helpful, not important but helpful to do this in order to have the armoury/weaponry and tools to go through this journey". (Caregiver, 004, F, 77 years)</p> <p>"Getting the encouragement to do the goals and if they're practically possible to go for it". (Person with MND, 002, F, 60 years)</p> <p>"To me, it's really important that the person with MND is able to make their own choices and their own decisions, so that they feel free to say how they feel". (Caregiver, 048, F, 59 years)</p>
	(2e) Building on positive attitudes	"Trying to put things back into the here and now as much as possible, is to try and find value in the current experience and not to see the absence of what they once had in terms of what they called success." (Healthcare

(Continued)

Table 2. (Continued).

Variability and meeting individual needs		professional, 016, M, 44 years) “It’s important to find out what their interests and things like that are so then we can build them up for the future”. (Healthcare professional, 034, F, 31 years)
	(3a) Variability and uncertainty	“... you’ve obviously got two people here who go through the same disease but at the beginning of diagnosis time we both had quite a different attitude and perhaps different feelings”. (Person with MND, 021, M, 67 years) “It’s such a peculiar disease and it doesn’t follow a set course. What’s good for one person might not be good for another”. (Caregiver, 027, F, 56) “... you know, their personalities might change or their decision making’s changing as well”. (Healthcare professional, 054, F, 48 years) “We just don’t know with MND how things are going to progress, how quickly, how slowly and you’ve no idea, you know, what the future holds for you”. (Person with MND, 014, F, 67 years)
	(3b) Practicalities in meeting varied needs	“It’s sometimes difficult, you know, to get out the house because people are that much equipment to bring. There are so many people have ventilation. So they have lots of equipment, as I say, so that would be one reason why people couldn’t get to clinic because they were carrying ventilation”. (Healthcare professional, 010, F, 44 years) “There are a lot of people who are not computer literate, you know there are people who can’t program a television recording system. You know, you want to be careful you don’t disenfranchise those ...”. (Person with MND, 032, M, 74 years) “For me now, getting to hospital is very difficult, so having a therapist coming home would be very useful”. (Person with MND, 026, M, 63 years) “And I think if they’ve got lots of appointments going on, which they probably have, anything, text reminders, and written information, I think is very useful”. (Caregiver, 048, F, 59 years)
	(3c) Being inclusive of those with communication difficulties	“Physical problems, for example communication problems, if they can’t physically communicate then that really going to be a big barrier”. (Healthcare professional, 054, F, 48 years)
	(3d) Previous experiences	“He had some counseling from the hospice but he hated it he found it intrusive”. (Caregiver, 029, F, 74 years) “I think that the idea of talking to try and help them to do the things they want to do is pie in the sky because I think it’s very rare that they can do anything”. (Caregiver, 029, F, 74 years).
Informal support	(4a) Peer support	“You do learn an awful lot from other people about how they cope with it and, you know, what information they found out and sharing that is often useful”. (Person with MND, 025, F, 61 years) “So some people have said, I don’t want to meet with other people with MND, I don’t want to see what I’m going to become. And that’s the risk, isn’t it, if you meet people with the same illness who are further on, and you think, I wish I hadn’t seen that”. (Healthcare professional, 020, F, 45 years) “Some people absolutely love it, for everything to be online and everything to be under their control and their time frame as well so they can choose when to dip in and out of the forum when they’re feeling up to it”. (Person with MND, 002, F, 60 years)
	(4b) Caregivers and family	“Well I think for therapy in general I think it’s important to bring the significant other on board with it, they don’t necessarily need to be part of it but actually I think one of the biggest influences on habits and communication and getting involved with stuff are family or significant others and if they buy into it seems to make it a bit easier for all”. (Healthcare professional, 038, M, 53 years) “... for patients who have familial MND. And it’s the concern for their children. If they, you know, have familial MND and they’ve already got children.” (Healthcare professional, 010, F, 44 years) “MND doesn’t just affect the person it affects the entire family”. (Caregiver, 027, F, 56 years)
	(4c) Caregiver burden	“Sometimes all patients can be quite selfish in terms of, you know, look at me. I’ve decided to do this thing and, you know, not quite realize it’s trampling over their caregiver’s plan to be a person or what have you ...”. (Caregiver, 035, M, 37 years) “Well, I think that is a huge point about being a burden, especially with the men again. I think you know they want to look after their families and they want to be the one to do things and they do, they hate being a burden”. (Caregiver, 030, F, 74 years)

Table 3. Challenges and opportunities for engagement in psychological interventions, as well as implications for psychological interventions.

Challenges/opportunities for engagement in psychological interventions	Implications for psychological interventions
(1a) Lack of MND awareness	Provide information about MND and refer patients, families and caregivers to MND resources (i.e. online forums and websites) to help expand their understanding and awareness of the disease.
(1b) Value placed on healthcare professionals' knowledge	Provide MND-specific training to therapists on physical, communication, cognitive and psychological difficulties that pwMND may experience. However, be aware that finding time for staff to attend training and put learning into practice may be another potential barrier (35). Give therapists the opportunity to familiarize themselves with equipment (e.g. communication devices) used by people with MND before engaging them in psychological interventions.
(1c) Finding the right time	Make MND healthcare professionals aware of the fact that refusals to engage in psychological interventions may change as an individual's circumstances change, particularly for men.
(1d) Value placed on testimonies from others with MND	Provide testimonies of others who have undergone psychological interventions and how/why they found it helpful.
(1e) Difficulties accepting help	Make MND healthcare professionals aware of the fact that men in particular may refuse to engage in psychological interventions. Gently explore the consequences of refusing to accept help in terms of the costs and benefits with respect to quality of life.
(1f) Navigating a new identity	Explore issues around identity with the person with MND and what living with MND means to them as a person.
(2a) Feelings of loss (loss of function, independence, hope, relationships, identity)	Acknowledge, validate and normalize the series of losses a person with MND may experience.
(2b) Psychological impact of losses (anger, frustration, hopelessness, embarrassment)	Acknowledge, validate, and normalize the thoughts, emotions and physical sensations a person may experience as a result of the losses. Gently explore how the person with MND responds to these internal experiences (i.e. what they do), and whether this is helping to improve their quality of life in the long-term.
(2c) Coping strategies (denial & avoidance)	Explore current coping strategies with the person with MND, the consequences of these strategies (i.e. the costs and benefits of them), and whether they could be doing anything differently. Emphasize the person with MND learning new skills to manage difficult thoughts, emotions and physical sensations in order that they can engage in the things that are important and that matter to them (i.e. their values) throughout the psychological intervention.
(2d) Need to empower patients	Emphasize that the person with MND always has a choice about whether to engage in particular exercises or discussions in order to reinforce perceptions of autonomy.
(2e) Building on positive attitudes	Help the person with MND to clarify what is important to them (i.e. their values). Emphasize living and finding value in the here and now through mindfulness exercises (31–33). Incorporate strategies for realistic goal setting (in service of personally meaningful values) in the psychological intervention and illustrate how these can be adapted as the disease progresses.
(3a) Variability and uncertainty	Discuss how to adapt sessions to the needs of the individual with MND at the onset of the intervention and then customize treatment delivery accordingly. Compensate for difficulties using standard therapeutic procedures. Address the uncertainties a person may experience due to the variability seen in MND.
(3b) Practicalities	Offer flexibility in session delivery (e.g. videoconferencing or at home). Allow for cancellations due to hospital appointments and illness. Discuss with the person what practical steps could be taken to support them engaging in therapy (e.g. providing visual prompts or session reminders).
(3c) Being inclusive of those with communication difficulties	Be aware of a person's difficulties with speech and allow for a slower pace of the sessions where necessary.
(3d) Previous experiences	Be aware of a person's previous experiences of psychological interventions and acknowledge what they found did and didn't work for them.
(4a) Peer support	Offer opportunities for and the choice to engage in peer support in the form of an online forum or online peer-support group.
(4b) Caregivers and family	Ask the person with MND if they would like to invite a carer or family member to engage in some sessions, though being aware of the additional burden this may place on caregivers.
(4c) Caregiver burden	Incorporate supplemental exercises that caregivers could utilize in addition to pwMND (or offer caregivers individual sessions in their own right).

Note: Subthemes in Table 2 correspond with those presented in Table 3.



Loss or impairment of speech was also closely linked to perceived negative changes in pwMND's body image and sense of identity. Caregivers commonly linked embarrassment to social withdrawal which was seen to further impact on the person with MND's sense of identity and wellbeing. Other common coping strategies included denial and avoidance.

Participants across the sample felt that finding ways to reassert control into the lives of pwMND would have a positive impact on their wellbeing and quality of life. Caregivers and healthcare professionals suggested equipping pwMND and caregivers with skills and knowledge of the disease and how to best live their life with the condition, as well as encouraging and supporting autonomy and goal setting. Participants also stressed the importance of finding value in the here and now and planning for the future in order to lessen the feelings of hopelessness and regain control over the diagnosis.

#### *Variability and difficulty meeting individual needs*

A recurring theme throughout the interviews was variability. Participants stressed that pwMND have different symptoms, rates of progression, experiences, and needs. There was agreement across the sample that the delivery of any psychological intervention for pwMND would need to be flexible due to the nature of the disease. Relying on others to attend appointments or the need for special equipment (e.g. communication devices) was considered as practical barriers to participating in psychological interventions. Psychological interventions delivered at home or through video conferencing were suggested as a method to overcome these barriers. Healthcare professionals also raised the issue of memory problems that can be experienced by pwMND. It was suggested that text reminders of appointments and providing written information could be helpful for those experiencing difficulties with their memory.

Those with communication difficulties reflected on their experience of feeling forgotten about with respect to the provision of psychological interventions for pwMND. Participants explained that their communication difficulties are not always catered for, and they feel they do not have a voice and are not being heard by non-MND healthcare services.

#### *Informal support*

Various types of support were reported, including that from peers, family members, and formal caregivers. A large number of healthcare professionals emphasized the importance of peer support from other pwMND. Professionals highlighted many benefits, including sharing experiential knowledge.

Contrastingly, pwMND commented on their reluctance to engage with peers due to the fear of being confronted with the progression of MND. Nevertheless, there was agreement among the participants there was value in online peer-support groups as these would provide flexibility in attendance (as pwMND could attend online when they wanted), which would lessen the burden on caregivers.

Caregivers and healthcare professionals discussed the benefits of engaging family members or caregivers in the sessions as they would be able to help with equipment, communication and encouraging overall engagement. However, some pwMND stated that they would feel uncomfortable with family members or caregivers attending the sessions with them as they may not feel able to fully discuss their feelings and concerns for the future.

PwMND commented on having to rely on caregivers and family support throughout their diagnosis in order to attend appointments, take medication and for day-to-day personal care. This was strongly associated with fears of being a burden on loved ones and anxiety around how family members would cope after their death. Concerns about how to approach conversations regarding familial MND were also mentioned. Finally, concerns were expressed about caregivers' wellbeing and the challenges of meeting their own needs and goals, by both caregivers and healthcare professionals.

## **Discussion**

This study highlights the perceived barriers of and facilitators to uptake and engagement in psychological interventions in pwMND, as well as their needs and preferences for such interventions. A number of the identified themes and sub-themes highlighted in Table 2 are consistent with previous research, as detailed below.

With respect to the theme "Unfamiliar territory", a lack of awareness of MND in the general population and non-MND services has previously been explored (20). This has also been linked to pwMND not wanting to engage with available healthcare services (21). Training non-MND healthcare professionals, including psychological therapists, about MND may help to overcome these barriers. The sub-theme "Finding the right time" is consistent with previous studies that have reported the frustration that can arise when people with ALS are offered help when it is not needed or wanted (20) and linked denial of the diagnosis and prognosis to disengagement from MND care teams (22). This can make it challenging for healthcare professionals to approach pwMND about psychological interventions and highlights the need to revisit any

refusals to engage in psychological interventions, as this decision may change as an individual's circumstances change.

The theme "A series of losses" identified the range of emotions and physical losses pwMND experience throughout the disease course and their impact on wellbeing. The sub-theme "Feelings of loss" was strongly linked to pwMND feeling that they have no choice but to accept the losses. This is consistent with previous evidence which has shown that pwMND feel forced to accept and adapt to the losses experienced due to MND (23). Encouraging patient autonomy in psychological interventions, such as in the content and format of sessions, could allow a person with MND to reassert some level of control they feel they are losing due to the disease process (24,25). Additionally, helping pwMND to let go of futile struggles with experiences or situations that cannot be controlled in order to focus on the things that are important and matter to them may positively impact on their wellbeing.

The theme "Disease variability and difficulty meeting individual needs" highlights some of the practical barriers to psychological interventions e.g. symptoms. Psychological interventions delivered *via* videoconferencing were suggested as a way to overcome practical barriers that are associated with mobility. There is some evidence to suggest that psychological interventions delivered by videoconference or teleconference are not inferior to treatment provided in person in the general population (26). However, whether this is the same for pwMND remains to be determined.

With respect to the theme "Informal support", loss of independence requires pwMND to become increasingly dependent on their caregivers, which may increase an individual's fears of being a burden on their caregiver. Caregiver burden has been found to increase as the disease progresses (27) and it has been reported that caregivers' own expectations and priorities are shaped by the needs of pwMND (28). Adapting therapy to involve caregivers might be beneficial in addressing concerns about being a burden.

#### *Implications for psychological interventions*

Table 3 highlights a number of implications for the development and delivery of psychological interventions for pwMND that follow on from the themes and sub-themes identified in this study. These implications point toward the value of interventions that have the scope to help a person: (i) learn new skills for managing understandable difficult thoughts and emotions (e.g. in relation to progressive losses, fears about meeting others with MND, etc.); (ii) increase personally meaningful behavior through practical, achievable goal setting guided by what is important and matters to them

(i.e. their values); (iii) distinguish between what can and cannot be controlled, let go of futile struggles with experiences that cannot be controlled, and re-focus energy on personally meaningful behavior; and (iv) increase awareness of their experiences in the here and now. One such intervention is acceptance and commitment therapy (ACT), which is a novel alternative to conventional forms of psychotherapy such as cognitive behavioral therapy (CBT). It has been argued that it may be particularly beneficial for those living in objectively difficult or immutable situations such as chronic or life-limiting conditions (29,30). It uses a combination of behavioral change and motivation-based techniques, in addition to mindfulness- and acceptance-based techniques, to help people engage in personally meaningful activity in the presence of understandable difficult emotions and thoughts. Supporting this, focusing on what can be realistically achieved rather than what has been lost and practicing mindfulness awareness have been reported as successful strategies for coping with symptoms and everyday problems in pwMND (20,31–33).

#### *Research recommendations*

Further research is required in order to, first, determine whether psychological interventions are beneficial for improving psychological wellbeing in pwMND and, second, to identify whether adapting psychological interventions to meet the individual preferences of pwMND can positively impact on uptake and engagement. Studies could also assess the benefits of including caregivers in the interventions, both to pwMND and to caregivers themselves. Finally, studies could explore the impact of potential moderating factors such as cognitive and communication difficulties on engagement and treatment outcomes.

#### *Strengths and limitations*

This research identifies preferences for psychological interventions among pwMND and provides valuable insights into the barriers to and facilitators of engagement in psychological interventions, as well as the clinical implications of these. A limitation of the study is that due to communication difficulties, some participants completed the interviews by writing their responses as they were not able to engage in face-to-face interviews. Therefore, they were not able to take part in the same interview conditions. However, including these participants is also a strength of the study as 80–95% of pwMND will experience speech difficulties in the later stages of the disease (34); including data from those currently experiencing these symptoms provides a valuable insight into their experiences and needs. Another limitation is

that although purposive sampling was used to select participants with a range of characteristics, we were unable to recruit pwMND and caregivers aged less than 50, those from ethnic minority groups and those with lower levels of education, within the time constraints of the study. Therefore, it cannot be assumed that the findings of this study and hence implications for psychological interventions apply to these groups.

## Conclusions

Although a range of barriers to and facilitators of engagement in psychological interventions have been identified, it is evident that allowing for flexibility, tailoring interventions to the individual needs of pwMND, and encouraging autonomy are key attributes for psychological interventions with pwMND. It is important to note that a person's preferences and needs may change throughout the progression of the disease and, therefore, it is important for psychological interventions and therapists to be flexible in order to accommodate this.

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## References

1. Van Es MA, Hardiman O, Chio A, Al-Chalabi A, Pasterkamp RJ, Veldink JH. Amyotrophic lateral sclerosis. *Lancet*. 2017;390:2084–98.
2. McLeod JE, Clarke DM. A review of psychosocial aspects of motor neurone disease. *J Neurol Sci*. 2007;258:4–10.
3. Kurt A, Nijboer F, Matuz T, Kübler A. Depression and anxiety in individuals with amyotrophic lateral sclerosis: epidemiology and management. *CNS Drugs*. 2007;21:279–91.
4. Taylor L, Wicks P, Leigh PN, Goldstein LH. Prevalence of depression in amyotrophic lateral sclerosis and other motor disorders. *Eur J Neurol*. 2010;17:1047–53.
5. Hogg KE, Goldstein LH, Leigh PN. The psychological impact of motor neurone disease. *Psychol Med*. 1994;24:625–32.
6. Fang F, Valdimarsdóttir U, Fürst CJ, Hultman C, Fall K, Sparén P, et al. Suicide among patients with amyotrophic lateral sclerosis. *Brain*. 2008;131:2729–33.
7. Gould RL, Coulson MC, Brown RG, Goldstein LH, Al-Chalabi A, Howard RJ. Psychotherapy and pharmacotherapy interventions to reduce distress or improve well-being in people with amyotrophic lateral sclerosis: a systematic review. *Amyotroph Lateral Scler Frontotemporal Degener*. 2015;16:293–302.
8. Howlett S, Grünewald RA, Khan A, Reuber M. Engagement in psychological treatment for functional neurological symptoms—Barriers and solutions. *Psychother Theory Res Pract Training*. 2007;44:354–60.
9. Baxter S, Enderby P, Evans P, Judge S. Barriers and facilitators to the use of high-technology augmentative and alternative communication devices: a systematic review and qualitative synthesis. *Int J Lang Commun Disord*. 2012;47:115–29.
10. Massman PJ, Sims J, Cooke N, Haverkamp LJ, Appel V, Appel SH. Prevalence and correlates of neuropsychological deficits in amyotrophic lateral sclerosis. *J Neurol Neurosurg Psychiatry*. 1996;61:450–5.
11. Ringholz GM, Appel SH, Bradshaw M, Cooke NA, Mosnik DM, Schulz PE. Prevalence and patterns of cognitive impairment in sporadic ALS. *Neurology*. 2005;65:586–90.

12. Goldstein LH, Abrahams S. Changes in cognition and behaviour in amyotrophic lateral sclerosis: nature of impairment and implications for assessment. *Lancet Neurol.* 2013;12:368–80.
13. Foley G, Timonen V, Hardiman O. Understanding psycho-social processes underpinning engagement with services in motor neurone disease: a qualitative study. *Palliat Med.* 2014;28:318–25.
14. Gruis KL, Wren PA, Huggins JE. Amyotrophic lateral sclerosis patients' self-reported satisfaction with assistive technology. *Muscle Nerve.* 2011;43:643–7.
15. Foley G, Timonen V, Hardiman O. Patients' perceptions of services and preferences for care in amyotrophic lateral sclerosis: a review. *Amyotroph Lateral Scler.* 2011;12: 11–24.
16. Brooks BR, Miller RG, Swash M, Munsat TL, World FN, Res G. El Escorial revisited: revised criteria for the diagnosis of amyotrophic lateral sclerosis. *Amyotroph Lateral Scler Other Motor Neuron Dis.* 2000;1:293–9.
17. Creswell J. Qualitative inquiry and research design: choosing among five traditions. Thousand Oaks, CA: Sage Publications; 1997.
18. Braun V, Clarke V. Using thematic analysis in psychology. *Qual Res Psychol.* 2006;3:77–101.
19. Glaser BG. Theoretical sensitivity: advances in the methodology of grounded theory. Mill Valley, CA: Sociology Press; 1978.
20. Soundy A, Condon N. Patients experiences of maintaining mental well-being and hope within motor neuron disease: a thematic synthesis. *Front Psychol.* 2015;6:606.
21. Hughes RA, Sinha A, Higginson I, Down K, Leigh P. Living with motor neurone disease: lives, experiences of services and suggestions for change. *Health Soc Care Community.* 2005;13:64–74.
22. Sakellariou D, Boniface G, Brown P. Experiences of living with motor neurone disease: a review of qualitative research. *Disabil Rehabil.* 2013;35:1765–73.
23. Foley G, Timonen V, Hardiman O. Exerting control and adapting to loss in amyotrophic lateral sclerosis. *Soc Sci Med.* 2014;101:113–9.
24. King SJ, Duke MM, O'Connor BA. Living with amyotrophic lateral sclerosis/motor neurone disease (ALS/MND): decision-making about 'ongoing change and adaptation'. *J Clin Nurs.* 2009;18:745–54.
25. Nasreen K, Marziali E, Tchernikov I, Shepherd N. Comparing telehealth-based and clinic-based group cognitive behavioral therapy for adults with depression and anxiety: a pilot study. *Clin Interv Aging.* 2014;9:765–70.
26. Stubbings DR, Rees CS, Roberts LD, Kane RT. Comparing in-person to videoconference-based cognitive behavioral therapy for mood and anxiety disorders: randomized controlled trial. *J Med Internet Res.* 2013;15: 258.
27. de Wit J, Bakker LA, van Groenestijn AC, van den Berg LH, Schröder CD, Visser-Meily JM, et al. Caregiver burden in amyotrophic lateral sclerosis: a systematic review. *Palliat Med.* 2018;32:231–45.
28. Lerum SV, Solbraekke KN, Frich JC. Family caregivers' accounts of caring for a family member with motor neurone disease in Norway: a qualitative study. *BMC Palliat Care.* 2015;15:22.
29. Graham CD, Simmons Z, Stuart SR, Rose MR. Issues & Opinions: the potential of psychological interventions to improve quality of life and mood in muscle disorders. *Muscle Nerve.* 2014;52:131–6.
30. Kangas M, McDonald S. Is it time to act? The potential of acceptance and commitment therapy for psychological problems following acquired brain injury. *Neuropsychol Rehabil.* 2011;21:250–76.
31. Rabbitte M, Bates U, Keane M. Psychological and psychotherapeutic approaches for people with motor neuron disease: a qualitative study. *Amyotroph Lateral Scler Frontotemporal Degener.* 2015;16:303–8.
32. Norweg A, E. Evidence for C. Cognitive-behavioral strategies improving dyspnea and related distress in COPD. *Int J Chron Obstruct Pulmon Dis.* 2013;8: 439–51.
33. Hopwood J, Walker N, McDonagh L, Rait G, Walters K, Liffé S. Internet-based interventions aimed at supporting family caregivers of people with dementia: systematic review. *J Med Int Res.* 2018;20:e216.
34. Beukelman D, Garrett K, Yorksto K. Augmentative communications strategies for adults with acute or chronic medical conditions. Baltimore, MD: Paul H. Brookes; 2007.