# Development of the MiNDToolkit for Management of Cognitive and Behavioural Impairment in Motor Neurone Disease

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## Abstract

**Aim**: To develop structured guidance, recommendations and techniques for non-pharmacological management of cognitive and behavioural impairments in motor neurone disease (MND), called the MiNDToolkit. **Methods**: A 4-round modified-Delphi method was utilised (online and face-to-face meeting), supplemented by recent research, recommendations, expertise from allied health-professionals (AHP) clinicians, researchers and clients. **Results**: Round-1 (N=47) identified AHPs techniques. Round-2 (N=23) and -3 (N=19) used expert consensus, refining general focus, specific elements and techniques. Round-4 (N=8) applied personal, lived and occupational experience, finalising the general structure and content of specific techniques. **Conclusion**: The MiNDToolkit is composed of multiple tools to structure decision-making through flowcharts, decision-trees and checklists, provide information about impairments, assessment recommendations and techniques or strategies for non-pharmacological management cognitive or behavioural impairments in MND.

**Keywords:** non-pharmacological; management; cognitive Impairment; behavioural Impairment; amyotrophic lateral sclerosis; frontotemporal dementia; amyotrophic lateral sclerosis frontotemporal spectrum disorder

## Introduction

Cognitive and behavioural impairment is prevalent throughout all stages of amyotrophic lateral sclerosis/motor neurone disease (ALS/MND) occurring in up to 80% of patients [1]. Further, ALS/MND exists on a spectrum with frontotemporal dementia, known as the ALS- Frontotemporal Spectrum Disorder (ALS-FTLD) / ALS with Frontotemporal Dementia (ALSFTD) [2]. FTD and ALS/MND have similar and overlapping profiles of behavioural and cognitive impairment [3-5] and approximately 20% of ALS patients can develop FTD or FTD can develop ALS [2,5]. These impairments are characterised by executive or language dysfunction, social cognitive deficits or a lack of sympathy and empathy as well as changes in behaviour or pre-existing behaviour becoming more noticeable [6]. Apathy (or inertia) is one of the most common behavioural impairments observed in MND [7], however rigidity and impulsivity are also prominent [8,9].

These impairments in cognitive and behaviour functioning have been shown to impact negatively people living with MND and their families. Multiple studies have shown negative effects on wellbeing, quality of life and increased burden on caregivers or family member of people with MND [10-13]. A recent systematic review showed that behavioural impairments, specifically apathy or impulsivity, were associated with increased burden [14]. More recently, a review suggested that while cognitive and behavioural impairment can have impact on both quality of life and caregiver burden, they might impact informed decision making and advance planning [15]. Apathy has also been shown to affect prognosis, with considerable impact on survival when moderate to severe apathy is detected in comparison to mild or no apathy [16]. These behavioural impairments have also been suggested to affect treatment compliance and adherence, which may account for the impact survival and other practical outcomes. Further, presence of cognitive impairment also seems to impact on survival, particularly when executive dysfunction or frontotemporal dementia is present [17-20].

According to the ALS-FTSD Revised Diagnostic Criteria [2], these changes can be grouped as ALS cognitive impairment (ALSci), ALS behavioural impairment (ALSbi), ALS with both impairments (ALScbi) and ALSFTD; guidance is also given on assessment of such impairments [2]. As such, multiple methods of briefly assessing or screening for cognitive or behavioural impairment have been developed and applied, which include the Edinburgh Cognitive and behavioural ALS Screen (ECAS) [21], ALS Cognitive Behavioral Screen (ALS-CBS) [22], Mini-Addenbrooke's Cognitive Examination (Mini-ACE) [23] and Motor Neuron Disease Behavioural Instrument (MiND-B) [9] many of which were found to be well validated and useful for assessment for these impairments in MND [24].

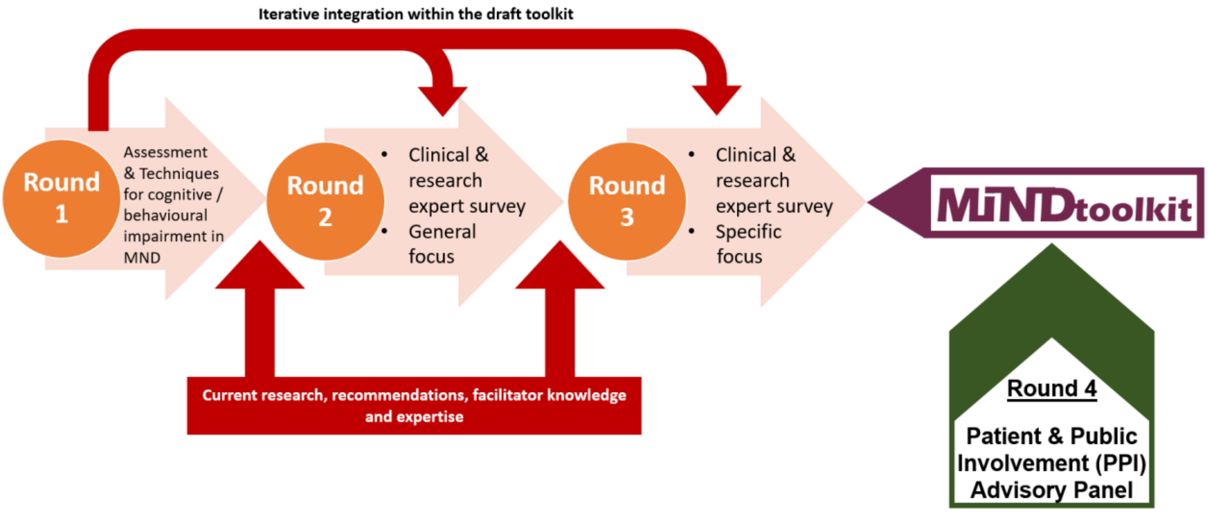
While the impact of cognitive and behaviour impairment for people with MND and their families have been well characterised, with great progress in the development and validation of assessments to detect cognitive and behavioural impairment, there has been less advancement in the development of guidance, interventions, strategies and/or techniques for the management of such issues. The aim of this paper is to describe the process of development of the MiNDToolkit, which is a set of structured tools for non-pharmacological management of cognitive and behavioural impairment for both healthcare professionals and family members or caregivers. It was developed through a compilation of current practices and research, expert opinion, current recommendations (from non-profit organisations) and patient and public consultation. As such it is important to employ a structured process to gather, synthesize and incorporate information, known as the Delphi method [25-27]. Utility of this method allows for facilitated collection of information to achieve consensus, from a variety of healthcare professionals and experts, which has been used previously to foster development of guidelines within MND [28]. Therefore, in applying this structured methodology and process would practically inform the design, content and structure of the MiNDToolkit.

## Methods

### Procedures

A modified-Delphi method [25-27] was chosen to structure and refine the information collected from all participants/respondents, with authors acting as facilitators. This Delphi method was composed of four rounds: three rounds of online consultation, and one round of face-to-face consultation. The online consultation was conducted through online surveys using SurveyMonkey and Microsoft forms, while continuously taking into account authors' expertise, current recommendations (from organisations such as the English Motor Neurone Disease Association of England, Wales and Northern Ireland, Irish Motor Neurone Disease Association) and research in the field. Round 1, Round 2 and Round 3 of the survey were used to create a proposed draft of the MiNDToolkit. Finally Round 4 was a face-to-face meeting of a Patient & Public Involvement Advisory Panel that allowed for comment and review of the proposed draft of the MiNDToolkit. Details of each Round are shown in Figure 1 and are described below.

**Figure 1. Rounds of the modified-Delphi method development of the MiNDToolkit**



## Modified Delphi Method

### *Round 1- Existing Practice, Approaches and Techniques Survey*

The questions were designed through iterative discussions by the authors, taking in to consideration previous research to understand barriers and practice in relation to cognitive and behavioural impairment assessment in MND [29-30]. Furthermore, additional questions were included to understand practical management of cognitive and behavioural impairment that healthcare professionals might be using. Allied health professionals were invited to an online survey and asked to provide information about 1) how cognitive and behavioural impairment is assessed in their everyday practice, 2) when and how frequently cognitive and behavioural impairment is assessed and 3) approaches/techniques for management of cognitive and behavioural impairment in MND that they use (and to rank which they thought were the best). This information was used to guide the structure of the MiNDToolkit and produce approaches, techniques and strategies for non-pharmacological management of cognitive and behavioural impairment in MND, as well as influence subsequent rounds of the modified Delphi method.

### *Round 2- General Clinical & Research Expert Panel Survey*

A list of international clinical and research experts working in the field of MND and FTD worldwide (America, Europe, Asia, Australia) was compiled, with the authors selecting experts taking in to account research publications within the fields of MND, FTD or MND-FTD, relevance of this to clinical management, as well as cognitive and behavioural impairment. The statements for the experts to rate for this round were formulated through iterative discussions by the authors, with ALS-FTSD diagnostic criteria [2] as a point of reference and to account the practical implications for both healthcare professionals and family caregivers or paid caregivers. Experts were invited to online surveys and asked to rate the importance of general information and other practical elements to healthcare professionals and family members or caregivers. Information was probed in relation to non-pharmacological management of cognitive and behavioural impairment in people with MND and those with MND-FTD. Statements explored expert's opinions of the importance relating to general practical aspects associated with cognitive and behavioural impairments (daily life, communication, knowledge, information, education and information). Statements were rated on a 4-point Likert scale (Unimportant, Not Very Important, Important, Very Important, ranging from 0 to 3 respectively). This was used to guide the general structure of the MiNDToolkit. Results of this round were fed back to the experts.

*Round 3- Specific Clinical & Research Expert Panel Survey*

International clinical and research experts from the list in Round 2 were then asked to provide their opinions on more specific practical aspects of cognitive and behavioural impairment. The statements for this round were once again formulated through iterative discussions by the authors, taking in to account the ALS-FTSD diagnostic criteria [2], the practical implications for both healthcare professionals and family caregivers or paid caregivers and the results from Round 2. Specifically, experts were asked their opinion on specific statements about implementation of and classification using the ALS-FTSD Revised Diagnostic Criteria [2] for health professionals or family/paid caregivers. Also, experts were asked about what specific information (Mental Capacity, Advanced Care Planning) is important to consider, as well as the importance of previously produced techniques (those from Round 1) for healthcare professionals and also caregivers in terms of non-pharmacological management of cognitive and behavioural impairment in people with MND and those with MND-FTD. Statements were rated on a 4-point Likert scale (Unimportant, Not Very Important, Important, Very Important, ranging from 0 to 3 respectively). This was used to guide and further refine the content of the MiNDToolkit.

*Round 4- Patient & Public Involvement Advisory Panel*

A half-day, interactive, in-person meeting held at the University of East Anglia, facilitated by authors was set to show the MiNDToolkit and to discuss it. The structure of the meeting was to: a) briefly introduce cognitive and behavioural impairment in MND and the challenges associated with these; b) describe the purpose and the development of the MiNDToolkit; c) review and discussion MiNDToolkit itself. In particular, attendees were asked to consider the general format, contents and strategies/techniques of the proposed draft of the MiNDToolkit. Attendees were encouraged to reflect on their own experiences in relation to cognitive and behavioural impairment in MND. Facilitators took notes throughout the meeting, which were synthesised, critically discussed and incorporated in the MiNDToolkit after the meeting.

This study received ethical approval from University of East Anglia Faculty of Medicine and Health Sciences (FMH) Research Ethics Committee (REC)/ Institutional Review Board (IRB). Participant provided informed consent.

## Participants

*Round 1, 2 and 3 – online consultation respondents*

Recruitment of allied health professionals occurred through the Motor Neurone Disease Association of England, Wales and Northern Ireland, conferences/symposia and various social media outlets. For Round 1, 47 professionals completed the whole survey. Another 83 completed the survey partially, with a total of 99 individuals interacting with the survey worldwide. All the respondents were practicing allied healthcare professionals working with people with MND. 61 international experts were selected by authors based on publication within fields of MND, FTD and MND-FTD. 23 experts (Round 2) and 19 experts (Round 3) in MND, FTD and MND-FTD responded and participated. Table 1 shows the descriptive demographics of respondents who completed the surveys for Rounds 1, 2 and 3 fully.

**Table 1. Demographics and descriptors of respondents from Round 1, 2 and 3**

| | Round 1 Respondents (N=47) | Round 2 Respondents (N=23) | Round 3 Respondents (N=19) |
|---|---|---|---|
| Age of respondents, years (Mean, SD) | 45.7 (8.5) | | |
| Gender (% F) | 87.2 | | |
| Education % | | | |
| Postgraduate | 55.3 | | |
| Undergraduate | 44.7 | | |
| Time working with patient group, years (Mean, SD) | 11.0 (8.4) | 19.3 (8.6) | 11.0 (8.4) |
| Part of Multidisciplinary Team % | 93.6 | | |
| Profession % | | | |
| Allied Health Professionals (e.g. Occupational Therapists, Speech & Language Therapists, Physiotherapists) | 44.7 | | |
| Consultant (e.g. Neurologist, Psychiatrist, Neuropsychologists, Psychologists, Palliative Care) | 29.8 | 87.0† | 68.4‡ |
| Specialist Nurses/Nurses | 19.1 | | |
| Other (e.g. Care Coordinators, Social Workers, Researcher) | 6.4 | 13.0 | 31.6 |

SD = standard deviation. † Neurologist = 47.8%, Psychiatrist = 8.7%, Clinical Neuropsychologist = 21.7%. ‡ Neurologist = 47.4%, Clinical Neuropsychologist = 21.1%. Not applicable fields in grey.

### *Round 4- Patient & Public Involvement Advisory Panel members*

Eight attendees (or member) were invited through the Norfolk MND Care and Research Network for participation in the advisory panel meeting. The consultation was conducted with 2 current and 1 former caregiver of people with MND with FTD, 1 person living with MND and their family member (both retired healthcare professionals), an MND Specialist Nurse Practitioner (HC), an MND Respiratory Nurse Practitioner, an MND Association Volunteer (also a retired healthcare professional).

## Results

### *Round 1- Existing Practice, Approaches and Techniques Survey*

In terms of assessment, 85.1% of health professional respondents assessed for cognitive or behavioural impairment in MND. Of those, 57.5% used a combination of clinical judgement with objective measurement, 27.5% used objective measurement only and 12.5% used clinical judgement only. Further exploration of objective measurements showed that the ECAS was the most commonly used assessment, however the Addenbrooke's Cognitive Examination-Revised (ACE-R), and its different versions, including the Mini-ACE), ALS-CBS and MiND-B were also cited as being used, although less commonly. In terms of frequency of assessment, 34.8% of health professional respondents said that assessments occur variably (when needed or observed or if something changes), and the remainder gave a range of 1 to 4 assessments per year (most commonly 2). Based on this, it was decided that the MiNDToolkit should include a flowchart and checklist for healthcare professionals to use as a step-by-step guide for their consultations relating to information about and assessment of (type and frequency) cognitive and behavioural impairment in MND. Additionally, educational materials in relation to the ALS-FTSD Revised Diagnostic Criteria and common cognitive and behavioural impairments were also to be included, so as to improve clinical judgement of healthcare professionals. Furthermore, recommendations for assessment and timescale were made, to emphasise the importance in objective measurement of cognitive and behavioural impairment in MND.

Of the respondents surveyed, 46.8% were aware of or used approaches for management of cognitive and behaviour impairment in MND. From these 46.8% of respondents, Table 2 shows a summary of approaches or techniques ranked as the best by these respondents (healthcare professionals), classified by thematic category.

## Table 2. Best approaches and techniques (N=35) for management of cognitive and behavioural impairment in MND from Round 1

| No. of approaches (%) | Thematic category | Summative details of approaches/techniques |
|---|---|---|
| 17 (48%) | Structured Support Approaches | - Strategies for managing the deficits or issues identified<br>- Strategies for managing the deficits or issues identified<br>- Adapting the environment (e.g. more/increasing structure, less distraction, safety measures)<br>- Break down tasks into manageable small steps<br>- Verbal interaction<br>- Distraction<br>- Routine<br>- Physical focus<br>- Behavioural approach<br>- Person Centred<br>- Family work/systemic approach<br>- Acceptance and Commitment Therapy |
| 7 (20%) | Personal Support Approaches | - Compassionate communication<br>- Increased time<br>- Make suggestions and let person come to own conclusions |
| 7 (20%) | Information and Education | - Information about cognitive change, Support<br>- Information/Education for carers/families and staff about impairment and impact<br>- Inform carers on changes and need for their involvement in patient care |
| 2 (6%) | Referral to support services | - Referral Occupational Therapist/Clinical Psychologist<br>- Refer on to psychological support services |
| 2 (6%) | Medication | - Medicate to reduce problem |

Table 2 shows that Structured Support Approaches were most often endorsed. Best approaches noted by allied health professionals was working with families or applying systemic approaches and providing caregivers or families with relevant strategies. Additionally, for patients adapting the environment (e.g. increasing structure, less distraction, safety measures) as well as breaking down tasks in to manageable small steps, verbal interaction and routine were noted as best approaches. In terms of Information and Education Approaches and Personalised Support Approaches, these were both second most endorsed. For the latter, best approaches were centred on themes of information and education for caregivers, family members and staff about the impairments and impacts. Additionally, information for caregivers or family members about what impairments to expect and the need for their involvement in patient care were noted. Personalised Support Approaches that were noted as best by allied health professionals was employing compassionate communication, increasing time and giving suggestions so that the person can come to their own conclusions. These four groupings of approaches had strong themes of providing support to the family member and caregivers, as well as patients. Finally, least endorsed were approaches which involved referral to support services (such as Clinical Psychologists or Occupational Therapists) and medication.

Current recommendations, research and authors expertise in the field, in concordance with the Round 1 results, produced an initial 7 techniques for healthcare professionals and caregivers or family members. These were:

1. Simplify decision making (e.g. limit choices and reduce open ended questions)
2. Support problem-solving (e.g. specify the topic being discussed, prompt if needed)
3. Clarify Complex Information (e.g. break down information, explain terminology that is unfamiliar)
4. Encourage & prompt engagement (e.g. encourage non-verbal responses, use visual and verbal cues, engage interest)
5. Motivate everyone involved (e.g. act as an external motivator to help the person initiate)
6. Facilitate shared decision making (e.g. encourage decisions to be made with involvement of the family/caregiver)
7. Support the family/caregiver (e.g. support for the family/caregiver in understanding cognitive and behavioural symptoms, expressing themselves to the patient, taking respite)

*Round 2- General Clinical & Research Expert Panel Survey*

According to experts, all statements were comparatively scored as ‘Very Important’ for healthcare professionals in terms of specific techniques, adaptations of daily practice and communication style when cognitive and behaviour impairment as well as FTD might be present. Further, according to experts, all statements were comparatively scored as ‘Very Important’ for family caregivers and paid caregivers in terms of information about and examples of cognitive impairment, behaviour impairment and FTD, as well as helpful contacts in relation to these problems. Similar to healthcare professionals, experts responded that specific techniques and communication styles were ‘Very Important’ when cognitive and behavioural impairment, as well as FTD, might be present. See Supplementary Table 1 for details of Round 2.

*Round 3- Specific Clinical & Research Expert Panel Survey*

According to experts, techniques relating to all individual levels of the ALS-FTSD Revised Diagnostic Criteria (ALSci, ALSbi, ALScbi and ALSFTD) were equally important (i.e. ‘Very Important’) for the health professionals, whereas techniques for the ALSci, ALSbi and ALScbi levels were rated as ‘Important’, with the exception of techniques for ALS-FTD which was rated as ‘Very Important‘ for family caregivers or paid caregivers. According to experts, the 7 techniques were rated as having varying importance for both the family health professionals and family members or paid caregivers. All techniques were rated as at least ‘Important’ with none being rated as ‘Unimportant’. See Supplementary Table 2 for details of Round 2 results including those for specific techniques.

Additionally, according to experts, for the all ALS-FTSD diagnosis (ALSci, ALSbi, ALScbi, ALS-FTD), mental capacity was rated as ‘Very Important’ to consider by healthcare professionals. For ALSci and ALSbi, earlier advance care planning was rated as ‘Important’ to consider, whereas for ALScbi and

ALS-FTD, earlier advance care planning was rated as ‘Very Important’ to consider by healthcare professionals. See Supplementary Table 3 for details of Round 2 Mental Capacity and earlier Advanced Care Planning results.

### Patient & Public Involvement Advisory Panel

Overall the panel agreed that the general format of the proposed draft of the MiNDToolkit was well-structured, pertinent and might be appropriate for use by healthcare professionals and family members. The panel advised that further information and educational material on common cognitive and behavioural impairments (and how they might be observed) would be beneficial for inclusion in the toolkit for both the healthcare professional and the family members or caregivers. Additionally, it was noted that further clarification and information on the ALS-FTSD Revised Diagnostic criteria terms (ALSci, ALSbi, ALScbi and ALS-FTD) would be useful for reference to healthcare professionals.

Specifically, the flowchart and checklist received positive feedback, were understood well and were perceived as helpful by all panel members. The panel noted that the toolkit should include multiple recommendations for cognitive and/or behavioural assessment, therefore providing options rather than favouring one specific assessment tool. In terms of the management techniques, these were all noted as valuable for both the healthcare professionals and family members. However, the panel members emphasised that fewer techniques with more examples would be more appropriate. Panel members with experience with people living with MND-FTD provided additional techniques not originally specified by healthcare professionals and experts earlier Rounds. Further, the panel members requested that space was provided for healthcare professionals to specify their own personal techniques for management which they have found effective in their clinical practice.

The panel members extensively discussed mental capacity in relation to cognitive and behavioural impairment in MND. While there is a lack of research in this area, panel members emphasised the importance of mental capacity and advanced care planning, further supported by panel members who are or were carers of people with MND-FTD. It was deduced that healthcare professionals should be mindful of facilitating involvement of family members in advanced care planning while considering mental capacity of the person with MND who? may have cognitive or behavioural impairment.

**MiNDToolkit**

The resulting MiNDToolkit was composed of 8 tools. Tool 1 is the Flowchart that ensures that steps are followed and documented in terms of detection and assessment of cognitive and behavioural impairment. Tool 2 is the Checklist to further document ALS-FTSD categorisation, as well as specific cognitive and behavioural impairments, guide and document the use of the subsequent tools. Tool 3a is the Criteria for Categorisation which is for the healthcare professional only and summarises the ALS-FTSD diagnostic criteria and categorisation of ALSci, ALSci, ALScbi and ALS-FTD. Tool 3b is the Impairments Mapped Against Domains which outlines common cognitive and behavioural impairments and their characteristics (executive dysfunction, language problems, issues with social cognition/lack of sympathy or empathy, apathy/lack of motivation, disinhibition/impulsivity, rigidity/inflexibility). This tool is for both the healthcare professional and the family members. Tool 4 is Behavioural and Cognitive Assessment which recommends and signposts tools for assessment. Tool 5 is for Managing Cognitive Impairments and Tool 6 is for Managing Behavioural impairment. These tools provide strategies and techniques that can help both the healthcare professional and family member with common cognitive or behavioural impairments. Tool 7 is Mental Capacity and Advanced Care Planning and is aimed alert the healthcare professional of the importance of these areas in relation to cognitive and behavioural impairment. Tool 8 is Clinical Reflective Notes for Health Professionals and Follow up: evaluation of strategies used by family and carers which allows the healthcare professional to document the effectiveness of strategies and techniques for each of the cognitive impairments upon follow-up. Additionally, the MiNDToolkit includes a completion inventory to ensure that relevant steps and Tools have been utilised. Please contact the corresponding author for further information about the MiNDToolkit.

## Discussion

Here we present the development of the MiNDToolkit for non-pharmacological management of cognitive and behavioural change or impairment in MND and MND-FTD. This was produced using modified-Delphi method, over several Rounds, and utilising various sources, spanning across current research, non-profit recommendations, service user, expert and health professional experience which has also supported the creation of a set of techniques and strategies. To further facilitate this, a structured delivery method, inclusive of assessment recommendations and forms for documentation of decision making has been included so as to supplement the implementation of the toolkit and the techniques or strategies within it. The MiNDToolkit is aimed at a range of healthcare professionals (e.g. occupational therapists, nurses, speech and language therapists etc.) and family members or caregivers so as to help mediate the impact commonly observed cognitive and behaviour impairment, inclusive of FTD, while following to the most up to date diagnostic criteria [2].

The importance of cognitive functioning and behaviour have been recognised through the National Institute for Health and Care Excellence Pathway and Guidance management of MND [31]. Cognitive and behavioural status has been isolated as significant throughout the care pathway, specifically within areas relating recognition and referral, information and support at diagnosis, organisation of care, managing other motor/non-motor symptoms and planning for end of life. NICE [31] guidelines further focus on a patient's needs and a tailored approach towards management of cognitive and behavioural impairment, with further signposting towards dementia specific guidance [32]. In dementia, such problems are addressed by referring to established codes of practice for the management of cognitive or behavioural impairment in specialist clinics and community teams [32]. However, these dementia guidelines may be difficult to apply to MND, due to this condition's fast progression, the physical disability and communication difficulties [6]. The MiNDToolkit utilises adapted techniques that can be used to account for the physical, psychological and communicative complexities of MND.

The approaches, techniques and strategies determined from Round 1, showcased that there is a multiplicity of methods being employed by healthcare professionals to manage cognitive and behavioural impairment, many of which are non-pharmacological. The most commonly used approaches, those termed 'Structured Support Approaches', thematically clustered around elements of already utilised therapeutic, evidence-based techniques, such as Cognitive Stimulation Therapy and Behavioural Management Techniques, as well as explicit mention of Acceptance and Commitment Therapy. These therapies have been found to be effective in dementia e.g. [33-35]. Other approaches were more personalised in terms of providing more general support, education and information for family members or caregivers about cognitive and behavioural impairment, which are also often observed in management of dementia e.g. [36]. However, the application of these approaches occur at variable points in the MND care pathway, due to lack of standardised practice or guidance in relation to interventions. There are various barriers that could effect implementation of routinely assessing cognitive functioning and behaviour in MND [29,30]. These barriers mostly due to provision of resources (fiscal and staff related), training and information for healthcare professionals, patients and family members, but also a lack of advice in terms of intervention and management for cognitive and behavioural impairment. Additionally, while the survey in Round 1 showed do healthcare professionals utilise their own or adaptive approaches for managing cognitive and behavioural impairment, the majority (just over 50%) of healthcare professionals did not have any approaches. As such this provides justification for the gap and therefore need for structured management approaches for these impairments for a range of healthcare professionals. Therefore, development and future use of the MiNDToolkit will aim to reduce the advice for management and intervention barrier, through providing a structured method to provide this.

The benefits of using a modified-Delphi methodology allowed for continual synthesis and updating of information and structure of the MiNDToolkit. Utilizing online surveys for initial rounds of the Delphi process, known as e-Delphi, has several advantages [37]. Firstly, it is a cost and time efficient method of gathering opinion and information, which also ensures a more diverse range of respondents and experts to be involved in the process (due to the global reach of internet-based research). The computerised nature of this process allows for quicker and more automated processing to arrive to a consensus opinion. A consensus was reached at an early stage within this modified-Delphi process (i.e. by Round 3), which can be attributed to the preparatory Round 1 that determined existing practice, approaches and techniques. This enabled for the facilitators (RR and EM) to create subsequent rounds based on the information gathered in Round 1, while also pooling on existing recommendations and current research, and iteratively synthesizing information throughout subsequent rounds. A further addition of a Patient & Public Involvement Advisory Panel (Round 4) face-to-face meeting ensured that a diverse pool of service users (both family members, people living with MND and healthcare professionals) can critically appraise the structure, practicalities and content of the MiNDToolkit. Of note, for Round 4 of the MiNDToolkit development, most of the attendees had extensive experience with people affected by cognitive and behavioural impairment and MND-FTD. Significantly the Patient & Public Involvement Advisory Panel meeting for the MiNDToolkit yielded meaningful changes, as well as additions, improving ecological validity of its content for use in the community. Patient & Public Involvement should continue to be a gold standard when designing interventions, and recent research has affirmed the positive impact of this e.g. [38]. As such, the current lack of evidence-based therapies for managing cognitive and behavioural impairment in MND, warrants the application of novel methodology to advance the field, such as the modified Delphi method used to design the MiNDToolkit. As assessment and awareness of cognitive and behavioural impairment increases, so will the provision for development of management techniques that can be incorporated in to the toolkit. Further consideration should be given to cultural perceptions of cognitive and behavioural impairment manifestations. This is an emerging area within neurodegenerative disease research, with projects looking to investigate dementia in developing countries and includes cultural experiences [39], the findings of which could potentially be applied to MND and MND-FTD. For example, sensation seeking (akin to disinhibition/impulsivity) may be variable dependent on gender and cultural norms e.g. [40], and as such may require adaptation to implementation and practice relating to the MiNDToolkit. However, further research is needed to further understand perception of culturally behavioural and cognitive norms.

In conclusion, the structured research-, experientially- and practically-driven modified-Delphi process has produced the MiNDToolkit for non-pharmacological management of cognitive and behavioural change or impairment for MND and MND-FTD. The toolkit’s 8-tool configuration provides a step-by-step guide for administration and documentation as well as providing educational material and recommendations of techniques and strategies for management of these impairments, in order to help people living with MND, their families and also healthcare professionals. Future research will look to determine the feasibility of the MiNDToolkit as an intervention within clinical practice, with an aim to determine implantation fidelity, explore potential outcomes prior to piloting and larger trial research. As such, this will build an evidence for managing cognitive and behavioural impairment in MND, in an effort to change practice.

## Future Perspective

As awareness and facilitation for assessment of cognitive and behavioural impairment in MND increases, it will create opportunity for development or adaptation of techniques, strategies or therapies for management. As the evidence-base accumulates, it will require more structured methods of incorporating and delivering these management methods for cognitive and behavioural impairment, across different aspects of care, for which the MiNDToolkit is equipped.

With different cultural experiences potentially influencing cognitive and behavioural impairment, as well as dementia, these will be of paramount importance to take in to account for assessment and management. The techniques, strategies and approaches for management of these impairment might therefore need cultural adaptation to facilitate better delivery as well as effective management. As such, the MiNDToolkit will be adapted for implementation to different cultures.

Future studies will look to expand provision for management of cognitive and behavioural impairment in MND using non-pharmacological strategies or techniques. This will improve the focus on person- and family-centred care and build more evidence toward the utility and applicability of implementing interventions such as the MiNDToolkit so as to make real impact in people’s lives.

**Summary points**

- Cognitive and behavioural impairment can occur frequently in amyotrophic lateral sclerosis (ALS)/ motor neurone disease (MND), which exists on a spectrum with frontotemporal dementia (FTD)
- There are multiple methods of detection of these impairments however, there are currently no structured methods or techniques for management of these impairments for healthcare professionals or family members/caregivers
- An iterative, 4 round modified Delphi method was used to create the MiNDToolkit for management of cognitive and behavioural impairment in MND, supplemented with, current expertise, research and recommendations
- Rounds 1 to 3 (online surveys) included allied health professionals worldwide and clinical and research experts to determine the content and structure of the MiNDToolkit. Round 4 (Patient & Public Involvement advisory panel meeting) utilised experience of specialist allied health professionals, people living with MND and family member of people living with MND and/or FTD to review and finalise the MiNDToolkit
- Round 1 collated allied health professionals' approaches, as well as frequency of assessment and various strategies or techniques for managing these impairments. These were thematically focused on adaptation, support, education, personalised and evidence-based approaches.
- Round 2 and 3 collectively reinforced the importance of structured education, information and understanding of diagnostic criteria for cognitive and behavioural impairment, for both healthcare professionals and family members/caregivers. All of these were ~~all~~ incorporated in the MiNDToolkit

- Round 4 further refined the arrangement of tools, content of recommendations and examples provided, techniques and practical aspects of the MiNDToolkit based on lived and specialist experience
- The resulting MiNDToolkit is a structured approach to managing cognitive and behavioural impairment in MND for healthcare professionals and family members/caregivers. A feasibility study is currently underway in the UK.

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