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ARTICLE

Receiving the news of a diagnosis of motor neuron disease: What does it take to make it better?

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Abstract

Our objectives were to identify the experiences of people with MND in receiving the diagnosis and to determine which aspects of breaking this bad news were associated with greater satisfaction with the way the diagnosis was delivered to them. An anonymous postal survey was facilitated by all MND associations in Australia, in 2014, and centred on the SPIKES protocol for communicating bad news. Of the patients ($n = 248$, response rate 29%), 36% were dissatisfied with the delivery of the diagnosis and gave low ratings on the ability/skills of their neurologists to deliver the diagnosis. It was evident that the longer the patients spent with their neurologists during breaking such bad news, the more they were satisfied and the higher they rated the neurologists' abilities/skills. The largest significant differences between neurologists rated as having high or low skills in delivering the diagnosis were in four domains: 1) responding empathically to the feelings of patient/family; 2) sharing the information and suggesting realistic goals; 3) exploring what patient/family are expecting or hoping for; and 4) making a plan and following through. In conclusion, with over one-third of patients dissatisfied with their experience, there is room for improvement in the practice of neurologists in specified areas that could form the basis for changing practice, and the development of standards and protocols likely to have implications at the international level.

Key words: MND/ALS, breaking bad news, empathy, SPIKES protocol, MND diagnosis

Introduction

Motor neuron disease (MND), also known as amyotrophic lateral sclerosis (ALS) or Lou Gehrig's disease, is a progressive neurodegenerative disease. Causes are unknown for approximately 90% of people with MND, and all cases are fatal with no effective treatment affecting the underlying disease mechanism (1). Presenting symptoms vary but typically include weakness in the hands or feet, trips and falls, swallowing difficulties, and slurred speech and nearly half of all people with MND may have impaired cognitive function, although overt frontotemporal dementia is less commonly observed (2,3). The time between diagnosis and death averages two to three years and most people with MND die from respiratory failure (1,4). In population

studies, approximately 10–20% of patients have a prolonged survival (5). The psychosocial impact of MND is intensified by the rapid speed of deterioration and relentless losses experienced by people with MND and their families (6–8).

Receiving a diagnosis of MND is challenging for patients and their families and is akin to an existential shock (7,9). The manner in which patients learn of a serious diagnosis is central to good practice guidelines in healthcare and underpins protocols developed for communicating bad news (10,11). Practice guidelines for neurologists acknowledge the challenges they face in communicating diagnoses of MND and emphasise communicating the diagnosis face-to-face in a private room without distractions; providing adequate time, at least 45–60 min, for conveying the diagnosis and

its discussion; complementing the face-to-face discussion with printed materials about the disease and relevant support services; and establishing a follow-up appointment that occurs within two to four weeks of diagnosis (12,13). Thus, the diagnosis is envisaged to be made in a way that acknowledges the individual's emotional and spiritual needs as well as addressing the medical and practical needs (14).

The communication of an MND diagnosis has implications for the way that patients and families move from the news of their diagnosis to the actions required for support throughout the illness trajectory; however, few studies have investigated communication of the diagnosis from the perspectives of people receiving the news (15). A survey of people attending a specialist MND centre in Italy demonstrated that most respondents were satisfied with the communication of the diagnosis (16). However, other studies demonstrate less satisfactory experiences. A survey of 64 people with MND in the United States showed that 27% reported at least one misdiagnosis before the MND diagnosis, with 8% undergoing costly and unnecessary surgeries (17). A more recent American survey of 144 people with MND demonstrated that fewer than half were satisfied with the way the diagnosis was communicated and 16% characterized it as poor (18). These issues with communication of the diagnosis feature prominently in qualitative studies exploring experiences of people with MND and their family caregivers. Interviews with 24 people with MND, 18 family caregivers, and 10 bereaved caregivers concluded that the delivery of the diagnoses ranged from being communicated in an informative and sensitive manner to being communicated abruptly, without empathy, and in public spaces (19). In an Australian study involving interviews with 16 bereaved family caregivers of people with MND, the caregivers described an absence of compassion during delivery of the diagnosis, which had long-standing effects (6). To date there are no studies with large sample sizes that have been conducted to ascertain the nature and extent of these challenging experiences.

We aimed to identify the experiences of people with MND in receiving the diagnosis, determine their overall satisfaction with the way they were given the news, and assess which aspects of the process of receiving the news were associated with greater satisfaction.

Methods

The study was approved by Curtin University Human Research Ethics Committee. The methods consisted of a cross-sectional design using an anonymous postal survey. The development of the questionnaire was undertaken after a comprehensive review of the international literature in this field and

with extensive consultation with clinicians and the executive officers of the MND associations in Australia.

Data collection

Australian MND associations provided the number of patients on their lists who were diagnosed in the last three years and were still alive. Envelopes were mailed to each association containing an invitation letter bearing the letterhead of the association, a patient survey with an information sheet, and a reply paid envelope. MND associations attached names and address labels and posted the envelopes in their state. No further contact was made to encourage response. Data collection spanned a period from April 2014 to January 2015.

Survey instrument

The patient survey consisted of 51 questions: demographic information (age, gender, marital status, education and postcode), date symptoms first started, date the diagnosis was first made, time spent by the neurologists giving the diagnosis. The perceived ability/skills of neurologists in delivering the diagnosis were assessed using a 5-point scale from excellent to poor. Attributes of effective communication of bad news were measured by the SPIKES protocol, a well-accepted system for communicating bad news developed by Baile et al. (10) and used by McCluskey et al. (18). The six domains are: 1) Setting – establishing the appropriate setting; 2) Perception – determining the needs and the perception of the patient; 3) Invitation – requesting an invitation to give the news; 4) Knowledge – providing knowledge and information to the patient; 5) Emotion – exploring the patient's feelings; 6) Strategy – forming a strategy with the patient to go forward. Each domain of the SPIKES protocol (setting, perception, invitation, knowledge, emotion and strategy) was assessed using direct questions requiring a 'yes', 'no' or 'do not recall' response, and directed statements requiring a response along a 5-point scale from 'strongly agree' to 'strongly disagree'. Open ended questions were included to capture more details from respondents.

Analysis

Frequencies and proportions were calculated and reported for categorical variables, and mean, standard deviation, median and range were calculated and reported for continuous and discrete variables. Normal distributions were tested using parametric means tests, and non-normal distributions were tested using non-parametric means tests.

The SPIKES domains were analysed by calculating a summary score for each domain. There were three questions in each of the setting and emotion domains, and two questions in each of the

perception, emotion, knowledge, invitation, and strategy and support domains. Responses of 'yes' and 'no' were coded 1 and 0, respectively. The sum of the questions in each domain was divided by the number of questions in that domain to give an average score. These scores were reported as per a continuous/discrete variable with mean, median, standard deviation and range. Responses of 'do not recall' were not included in the analysis but these were few cases. The internal consistency (Cronbach's alpha) was particularly good for three SPIKES domains: emotion ($\alpha=0.897$), knowledge ($\alpha=0.731$) and invitation ($\alpha=0.636$).

Further analysis was also undertaken with people with MND split into two groups based upon responses to question about how they rated the ability and skills of the neurologist giving them their diagnosis: those that were rated 'poor, below average or average' were assigned to one group (average or below = low rating), while those that were rated 'good or excellent' were assigned to a second group (above average = high rating). Further comparisons using mean/median tests or χ^2 tests were then made within these two groups according to the ability and skills rating of the neurologist to determine any differences in their experiences. Indicative responses to the open ended questions were selected to illustrate the above and below average experiences within each domain (20).

Results

MND associations posted 864 questionnaires, with nine returned as no forwarding address was available. Responses were received from 248 patients, yielding an overall response rate of 29% (ranging from 35% to 26% between five Australian states). Comparative analyses were conducted on 243 patients as five had missed completing a few sections of the questionnaire.

Respondents' profile

The mean age of respondents was 66.4 years (SD = 11.0, range 30–91 years), 59% were male, 78% were married, and 75% were retirees. The median period from diagnosis was 15 months (1–87), period from first symptoms to diagnosis was 10 months for the median (range 1–119). Over two-thirds (69%) reported having cervical/lumbar symptoms at onset, 19% had bulbar symptoms and the rest a combination of symptoms. About one-third of people with MND had seen another neurologist prior to their diagnosis, 15% had seen an ENT specialist, 11% an orthopaedic surgeon, 9% a speech pathologist, and 8% a chiropractor. The majority were given the diagnosis by a neurologist (95%) through several visits: 33% had two visits, 17% had three visits and 19% had more than three visits. The median length of the consultation was 30

min (range 1–300 min). Seventy percent of patients reported that they had a relative present with them during the consultation.

Ratings of neurologists' abilities/skills and satisfaction with delivery of diagnosis

About two-thirds of patients (64%, $n=156$) rated the abilities and skills of their neurologists at delivering the diagnosis as 'above average' (high rating) and 36% ($n=87$) rated the ability as 'average or below' (low rating). When asked to rate their satisfaction with the delivery of the diagnosis, 65% of people with MND were satisfied (very satisfied/satisfied) and 35% were not satisfied (very dissatisfied/dissatisfied). Patients' satisfaction with the delivery of diagnosis was strongly associated with the patients' ratings of the neurologists' abilities/skills ($\chi^2(1)=88.7$, $p<0.001$).

Duration of consultation

Patients who rated highly the abilities of their neurologists had a shorter period between first symptoms and diagnosis (mean 20 and 26 months, respectively, $p=0.021$), had significantly longer consultation times (median 40 vs. 30 min, $p<0.001$), felt they had sufficient time taken to receive diagnosis (just enough 84% vs. 48%, and not enough 4% vs. 34%, $p<0.001$), and were males (64% vs. 49%, $p=0.042$). Figure 1 demonstrates that the patients' ratings of the neurologists' abilities/skills increased as the duration of consultation increased. Similarly, Figure 2 presents the patients' satisfaction with the delivery of diagnosis increasing as the duration of consultation increased.

Comparisons within each SPIKE domain

Table I presents the differences in each SPIKES domain between the neurologists with high and low ratings in delivering the diagnosis.

Setting: creating the right setting. The two groups of neurologists (with high and low ratings of ability) significantly differed in two out of the three variables of the setting domain; the diagnosis was given in a completely private space and there were no interruptions.

Perception: determining what the patient/family knows. There were no significant differences in this domain between the two groups of neurologists, in terms of the neurologist's perception of the patient's extent of knowledge of their condition and how much detailed information they wanted to have from the neurologist.

Invitation: exploring what patient/family are expecting or hoping for. Patients who rated highly the abilities of their neurologists were significantly more likely to be asked how much they knew about MND and how much detail they wanted to know.

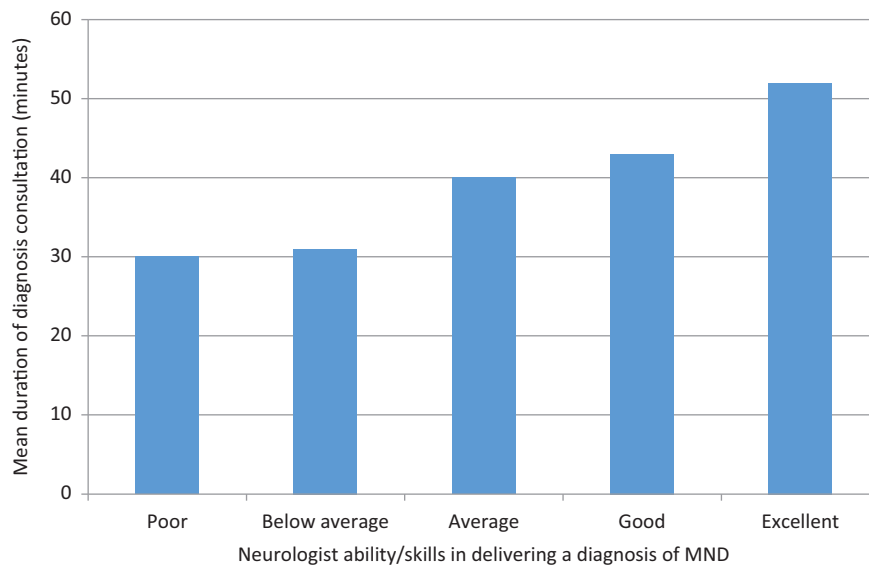


Figure 1. People with MND ratings of the neurologists' abilities/skills and consultation duration.

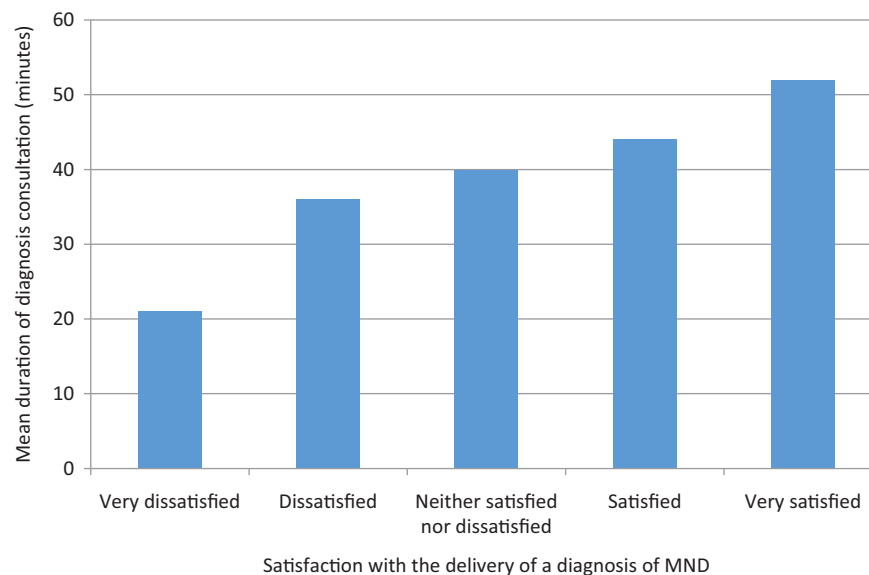


Figure 2. People with MND ratings of satisfaction with delivery of diagnosis and consultation duration.

Knowledge: sharing the information and suggesting realistic goals. Patients who rated highly the abilities of their neurologists were significantly more likely to be satisfied with how much detail they were provided, and were significantly more likely to be satisfied with the type of information they received. The highly rated neurologists were more likely to discuss how the diagnosis was reached, the degree of certainty, the current state of knowledge, current research and therapeutic trials, and the Australian MND Registry. Those patients who rated highly the abilities of their neurologists were more likely to receive the diagnosis in writing, further information on aspects of MND, information about MND associations, MND association publications and fact sheets, relevant MND internet sites, a copy of

the consultation letter and more likely to receive an estimate of their life expectancy.

Emotion: responding empathically to the feelings of patient/family. Patients who highly rated the abilities of their neurologists were more likely to agree that their neurologist gave them the diagnosis with warmth, care and empathy, that they were allowed more time to express their emotions, and they were allowed enough time to have these emotions responded to.

Strategy: making a plan and follow-through. The following referral and support aspects were more likely to be discussed with patients who rated highly their neurologists' abilities: a referral to an MND multidisciplinary clinic, a referral to the MND association, a follow-up plan for immediate and

Table I. Comparisons (%) within each SPIKES domain between the neurologists with high and low ratings in delivering the diagnosis.

| SPIKES domain | | High rating <i>n</i> = 156 | Low rating <i>n</i> = 87 | <i>p</i> -value |
|--|---|-------------------------------|-----------------------------|-----------------|
| Setting: creating the right setting | Completely private space | 98 | 91 | 0.019 |
| | No interruptions | 95 | 86 | 0.027 |
| | Relative/friend present | 72 | 70 | 0.653 |
| | Seen alone | 19 | 24 | 0.412 |
| Perception: determining what the patient/ family knows | Knew some/much information about MND | 34 | 28 | 0.389 |
| | Wanted a lot/just enough detail about MND | 86 | 79 | 0.147 |
| | | | | |
| Invitation: exploring what patient/family are expecting or hoping for | Asked by neurologist how much they knew about MND | 54 | 24 | <0.001 |
| | Asked by neurologist how much detail they wanted to know about MND | 40 | 13 | <0.001 |
| | | | | |
| Knowledge: sharing the information and suggesting realistic goals | Satisfied with detail provided | 82 | 45 | <0.001 |
| | Satisfied with type of information received | 76 | 31 | <0.001 |
| | How the diagnosis was reached | 71 | 56 | 0.024 |
| | The degree of certainty of diagnosis | 76 | 61 | 0.013 |
| | The current state of knowledge | 42 | 22 | 0.002 |
| | Current research and therapeutic trials | 21 | 7 | 0.003 |
| | The Australian MND Registry | 39 | 17 | <0.001 |
| | Receive the diagnosis in writing | 21 | 8 | 0.011 |
| | Receive further information on aspects of MND | 31 | 14 | 0.003 |
| | Information about MND Association | 54 | 21 | <0.001 |
| | MND Association publications and fact sheets | 32 | 12 | <0.001 |
| | Relevant MND internet sites | 18 | 3 | 0.001 |
| | Copy of consultation letter | 24 | 13 | 0.031 |
| | Estimate of life expectancy | 64 | 48 | 0.020 |
| | Diagnosis given with warmth, care and empathy | 88 | 29 | <0.001 |
| Emotion: responding empathetically to the feelings of patient/family | Allowed time to express emotions | 84 | 28 | <0.001 |
| | Allowed time to have these emotions responded to by neurologist | 83 | 26 | <0.001 |
| | | | | |
| Strategy: making a plan and follow- through | Referral to an MND multidisciplinary clinic | 44 | 22 | <0.001 |
| | Referral to the MND Association | 51 | 23 | <0.001 |
| | Role of community support | 29 | 8 | <0.001 |
| | Role of community palliative care | 21 | 8 | 0.011 |
| | Support from neurologist | 50 | 28 | 0.001 |
| | Support from MND specialist nurse | 32 | 16 | 0.007 |

long-term support, the role of community support and the role of community palliative care. Furthermore, the neurologist was rated highly by patients if the support was received by a neurologist or the MND specialist nurse compared to other health professionals. There was no statistical difference between the two groups in the timing of the next follow-up visit to the neurologist (about seven weeks) and the median interval between subsequent follow-ups (12 weeks).

Summary comparing all SPIKES domains

Table II and Figure 3 summarize the difference between the patients' ratings of neurologists' abilities (high and low ratings) across the six SPIKES domains. There were statistically significant differences in the performance ratings in delivering the diagnosis mainly across four domains, and the

largest significant differences between the two groups of neurologists were in the following domains: invitation, knowledge, emotion and strategy.

Table III presents reported experiences of people with MND within each SPIKE domain, with respondents' quotes depicting positive and negative experiences categorized by ratings of neurologists' skills in delivering the diagnosis.

Discussion and conclusion

This is the first national Australian study to provide a comprehensive insight into the process of receiving an MND diagnosis from the patients' perspective. Previous studies in Australia and elsewhere were qualitative, relied on small samples and portrayed mainly disaffected patients due to the self-selection bias (6,19,21).

Table II. Ratings of the neurologists' abilities and skills to deliver the diagnosis, grouped under the six SPIKES domains.

| SPIKES domains | Rating of abilities of neurologists by people with MND | | | | | |
|-------------------|--|------------|---------------|------------|-----------------|---------------------|
| | High rating | | Low rating | | <i>p</i> -value | Cronbach's α |
| | <i>n</i> = 156 | | <i>n</i> = 87 | | | |
| Setting | | | | | 0.031 | 0.470 |
| Mean \pm SD | 0.90 | ± 0.16 | 0.83 | ± 0.22 | | |
| Median (Min, Max) | 1.0 | (0.3, 1) | 1.0 | (0.3, 1) | | |
| Perception | | | | | 0.127 | 0.290 |
| Mean \pm SD | 0.60 | ± 0.29 | 0.54 | ± 0.30 | | |
| Median (Min, Max) | 0.5 | (0, 1) | 0.5 | (0, 1) | | |
| Invitation | | | | | <0.001 | 0.636 |
| Mean \pm SD | 0.47 | ± 0.43 | 0.19 | ± 0.31 | | |
| Median (Min, Max) | 0.5 | (0, 1) | 0.0 | (0, 1) | | |
| Knowledge | | | | | <0.001 | 0.731 |
| Mean \pm SD | 0.79 | ± 0.34 | 0.38 | ± 0.43 | | |
| Median (Min, Max) | 1.0 | (0, 1) | 0.0 | (0, 1) | | |
| Emotion | | | | | <0.001 | 0.897 |
| Mean \pm SD | 0.85 | ± 0.31 | 0.26 | ± 0.38 | | |
| Median (Min, Max) | 1.0 | (0, 1) | 0.0 | (0, 1) | | |
| Strategy | | | | | <0.001 | 0.473 |
| Mean \pm SD | 0.32 | ± 0.17 | 0.20 | ± 0.15 | | |
| Median (Min, Max) | 0.3 | (0, 0.9) | 0.1 | (0, 0.7) | | |

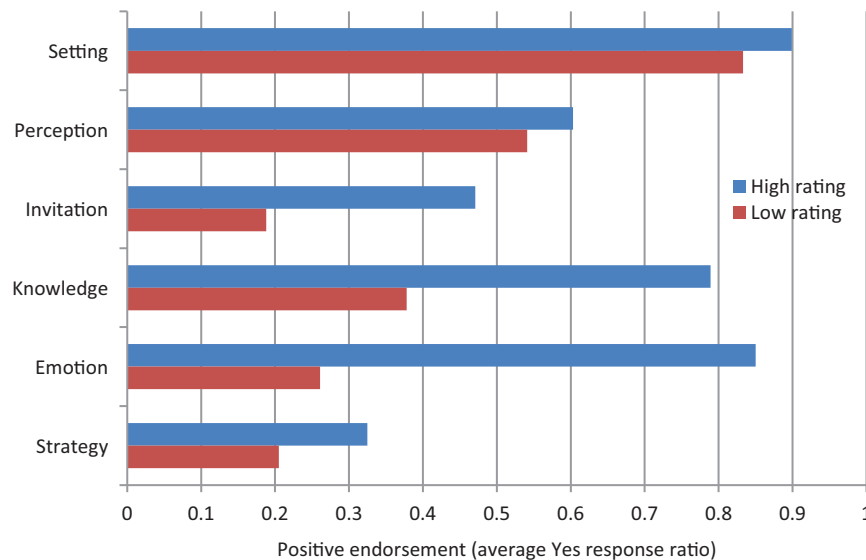


Figure 3. People with MND ratings of the neurologists' abilities/skills grouped under the six SPIKES domains.

Respondents to this national survey seemed to fall into two distinct groups: 64% were satisfied with the delivery of the diagnosis and rated highly the abilities/skills of their neurologists, and 36% were dissatisfied with the delivery of the diagnosis and gave low ratings on the abilities/skills of their neurologists. The qualitative comments of people with MND in Table III significantly reinforce the positive and negative experiences, which were closely related to the perceived ability of their neurologist and the reported satisfaction with the diagnosis delivery process. These comments are not dissimilar to those obtained from the smaller qualitative studies and particularly the comments about the need for empathy (6,19,21).

Additionally, the overall rating of skills was closely associated with performance within the SPIKES domains. In particular, the largest significant differences in ability/skills in delivering the diagnosis between the two groups of neurologists according to the patients were in four domains: 1) Emotion (responding empathically to the feelings of patient/family), where empathy was an important attribute of highly rated neurologists and how they dealt with the emotions of the patient and family; 2) Knowledge (sharing the information and suggesting realistic goals) where highly rated neurologists gave information about all aspects related to the disease, certainty, research, estimation of life expectancy and information on the MND association; 3) Invitation

Table III. Respondents' feedback depicting their experiences with the two groups of neurologists in each SPIKE domain.

| SPIKE domains | Rating | Quotes illustrating positive and negative experiences categorized by ratings of neurologists' skills and satisfaction with delivery of diagnosis (high and low ratings). |
|--|--------|--|
| SETTING (Creating the right setting) A completely private space | Low | "Info given to me only referred to negatives which upset me - at time of info given no private area to remain in and went out to public area that was full and I was upset in front of them." (P190) |
| Presence of interruptions | Low | "I was not impressed by the number of interruptions during my neurologist consultations i.e. Phone calls, missing reports, and copies of reports being brought in ..." (P340) |
| Presence of a relative or friend at the diagnosis | High | "Neurologist was very professional but also empathetic and answer(ed) question from both patient and carer plus two adult children" (P225) |
| | Low | "I should have been told to bring my husband or a support person when he knew he was giving me the diagnosis." (P230) |
| PERCEPTION (determining what the patient/family knows) Perception for just enough knowledge needed | High | "At initial diagnosis I did not want to know long-term details. About three months later I wanted to know long-term details." (P370) |
| | Low | "Explanation too fast and too technical despite my reasonable biological knowledge." (P201) |
| INVITATION (exploring what patient/family are expecting or hoping for) Prior knowledge about MND | High | "I had been told by my first neurologist that MND was on the table so I had done some research in the months between him and seeing neurologist ... It was all pretty grim so I did not delve too deeply". (P09) |
| | Low | "I think they should start with 'What do you know about MND?' As an icebreaker." (P229) |
| Details wanted | High | "The Dr. was very kind, sympathetic. He asked how much I wanted to know." (P73) |
| | Low | "At the first interview I should have been given the opportunity to be asked if I would like more information or not. The internet is a daunting place and that is where I had to source information." (P205) |
| KNOWLEDGE (sharing the information and suggesting realistic goals) Satisfaction with how much detail was provided | High | "I was not bombarded with the whole story in one hit and allowed time to deal with it." (P296) |
| | Low | "I was told very bluntly about my situation and how it would basically progress. He said if I had any plans do them now." (P176) |
| Satisfaction with type of information received | High | "Information given also by way of DVD - excellent!" (P94) "The neurologist spoke clearly, calmly and answered my questions in 'layman's' language." (P162) |
| | Low | "Would gladly have driven into a pole on the way home because I was given no hope." (P64) |
| | High | "Not enough information given, certainly at the first two appointments. His presentation could best be described as robotic." (P205) |
| Discussion of how diagnosis was reached | High | "... I asked whether it could be MND and she replied that that was what she was thinking. She then explained the reasons for her diagnosis to my wife and me, and explained the associated implications very clearly and with empathy." (P152) |
| | Low | "1st visit to neurologist said he would like to do nerve conduction tests. He didn't say why. I asked could I have an ageing disease like MS. He was surprised that I asked and said 'No, I suspect motor neuron disease.'" (P234) |
| The degree of certainty | High | "Clear, matter of fact, fairly unequivocal, reasonable estimate of life span, follow-up with counsellor over following months." (P430) |
| | Low | "Not decisive enough - which can lead to false hopes of a mistaken diagnosis." (P305) |
| The current state of knowledge | High | "The support and professional advice I am receiving are as good as it gets. Because the disease develops in different parts of the body and at different rates of progression, getting accurate answers on the future is virtually impossible." (P158) |
| | Low | "Diagnosis and explanation were quite perfunctory with no explanation to follow-up care. Terminology used was outdated and wrong as to specifics. Expected lifespan estimate was optimistic (7 years)." (P341) |

(continued)

Table III. Continued

| SPIKE domains | Rating | Quotes illustrating positive and negative experiences categorized by ratings of neurologists' skills and satisfaction with delivery of diagnosis (high and low ratings). |
|---|--------|---|
| Current research and therapeutic trials | High | "I was invited to be part of their research and had research MRIs and check-ups at regular intervals, maybe 2 or 3 months." (P94) |
| The Australian MND Register | Low | "Somewhat confused as appointment with neurologist not explained as an MND Research clinic." (P136) |
| Diagnosis in writing | High | "After the diagnosis I had contact with a representative from the MND registry and she has been in contact ever since. . . Since being in contact with the MND Registry I have had adequate information sent to me." (P258) |
| Further information on aspects of MND | High | "Thorough delivery of how diagnosis was reached. Hand written copy of doctor's notes given to me on departure which I found handy to refer back on." (P232) |
| | Low | "I was not given a diagnosis in writing until I insisted on it. I was not advised to seek a second opinion." (P343) |
| | High | "After initial diagnosis it was 10 days before neurological tests and confirmation of MND. Neurologist saw me same day at (Name) to discuss all aspects" (P180) |
| | Low | "I was used as a learning subject for a student. Lack of info about types of MND. I wasn't told what type of MND I had. Here's what he said: 'There's no known cause - no cure - no treatment - no remission - the disease is terminal and can progress rapidly'." (P343) |
| Information about MND association | High | "Neurologist said many times to contact MND Association who will help and provide information." (P296) |
| Relevant MND internet sites | Low | "We wanted more details about MND help and the Assoc - received no guidance." (P30) |
| | High | "One good thing is that the clinic nurse provided a referral to the MND Association website." (P299) |
| | Low | "My knowledge came from internet. Therefore sought advice of 2nd neurologist. The literature given was inappropriate and horrifying - even 3 years down the line." (P66) |
| Estimate of life expectancy | High | "I felt that it was very important to receive an honest assessment of my prognosis and life expectancy." (P18) |
| | Low | "I was totally shocked by my initial diagnosis and the estimate of my possible life expectancy." (P273) |
| EMOTION (Responding empathically to the feelings of patient/family) | High | "He detected it very quickly and was so gentle and caring in the way he told me, I was not upset. I was glad to find a name for all my medical problems." (P264) |
| Diagnosis given with warmth, care and empathy | Low | "No positive aspects at all (no help nor care). Had the bedside manner of a brick." (P335) |
| | | "Neurologist was clinically thorough but uncaring when providing diagnosis of MND. Gave brochure for information. Told me I had 3 years left to live. Was left alone in city 2 hrs from home after diagnosis." (P85) |
| More time allowed to express emotions | High | "The neurologist was lovely. . . the appointment was in the afternoon and she spent quite a long time with us we did not feel rushed." (P09) |
| | | "I received diagnosis by neurologist and then [she] left room so I could maintain my composure in private. Then [she] came back and gave me time to question her before seeing me again next day." (P54) |
| | Low | "I felt the neurologist was very insensitive. Especially saying it is 'terminal'. I didn't have enough time to comprehend what I had just been told." (P234) |
| | | "Despite knowing a lot, expecting the bad mutation, it was a shock. We didn't have time (or empathy?) to discuss feelings and consequences." (P430) |
| Enough time to have these emotions responded to | High | "The diagnosis was given very professionally with appropriate empathy and a good understanding of my reactions to it - which were calm." (P152) |
| | Low | "More information and more time would have allowed opportunity for me to be questioned about my feelings and for me to respond accordingly." (P205) |

(continued)

| SPIKE domains | Rating | Quotes illustrating positive and negative experiences categorized by ratings of neurologists' skills and satisfaction with delivery of diagnosis (high and low ratings). |
|--|--------|---|
| STRATEGY (Making a plan and follow-through) A referral to an MND multidisciplinary clinic | High | "After making contact with specialist neurologist, support and care have been all one could hope for. Local allied health, as well as staff at MND clinic, have provided all the information I have needed. Also kept one step ahead with any equipment required. Local rep of MND Association has also been very supportive. After a shaky start in which I felt a bit bereft of knowledge of where to turn, my care has been first class." (P284) |
| | Low | "Was not offered follow-up from initial neurologist. By internet research we made contact with Professor [Name] and had initial consultation... where diagnosis of ALS was given and support through medication and clinic... contact also with MND Association initiated at this stage." (P341) |
| A referral to the MND association | Low | "Yes - in hindsight while original neurologist diagnosed and was compassionate - no f/up plan or support was given - we had to source this ourselves - no idea there was an MND association etc. poorly supported until I contacted MND Association - have been well supported since then." (P30) |
| | High | "Needed more [support] at beginning - monthly for first 4-6 months, then longer intervals. Need time to ask all Q's, go through anger/grief, then settle into monitoring and adapting to progress of disease." (P52) |
| A follow-up plan for immediate and long-term support | Low | "It was just a diagnosis to the neurologist and no emotional support or ongoing guidance was given." (P341) |
| | High | "... put into hospital and the palliative care Dr took over from the neurologist." (P389) |
| | Low | "After diagnosis, the neurologist providing 2nd opinion became regular neurologist at MND clinic. Now regularly see palliative specialist Dr - appointments every 3 months." (P201) |

(exploring what patient/family are expecting or hoping for) where highly rated neurologists made the effort to ask how much the patient already knew about their condition and how much detail they wished to have; and 4) Strategy (making a plan and follow-through) where satisfied patients appreciated having discussions about referrals to the MND association, to the multidisciplinary clinic, the role of palliative care, the role of community support and a follow-up plan for immediate and long-term support.

It is evident that the longer the patients spent with their neurologists during breaking this bad news, the more they were satisfied with the delivery process and the higher they rated the neurologist's abilities/skills. The median length of the consultation reported in this study was 30 min. However, delivering the diagnosis is a process that requires 45–55 min according to patients who rated the ability of the neurologist 'good to excellent' (Figure 1), or those who were 'satisfied to very satisfied' (Figure 2). This is very similar to the standard outlined in the European guidelines on the clinical management of ALS: Breaking the news – communicating the diagnosis, which states that enough time needs to be available on the part of the physician (at least 45–60 min) (12). Regarding follow-up support, respondents reported a median of seven weeks for the first follow-up visit after diagnosis, while the recommended practice should be within two to four weeks or sooner. Only about 40% of patients reported being referred to an MND association (when all should be referred) and 16% received the diagnosis in writing. Some improvements are clearly needed in these areas for the patients to feel more supported. Comments in Table III highlight the evident relief in accessing better support once patients were connected to the MND associations. Having the diagnosis in writing would help the patient and family communicate the diagnosis to such support organizations.

Other current practices reported in this study that align with the European guidelines include that for 95% of patients the diagnosis was communicated by a consultant neurologist and for 70% of patients the diagnosis was communicated in a stepwise fashion over two or three visits. Also, the majority of patients (96%) reported that the diagnosis was given in a completely private space and had no interruptions while given the diagnosis (91%), and that 70% had a relative present with them when given the diagnosis.

With 29% response rate to the survey, we cannot be certain of the representativeness of this group of the general population of people with MND in Australia, nor does Australia have a comprehensive register that captures all cases in order to make comparisons. However, the profile of respondents seemed to be similar to another comparable study

that used a survey in terms of age, gender and marital status (18). We cannot also be certain if more satisfied or dissatisfied people made the effort to respond. In the McCluskey et al.'s survey (2004) in Philadelphia, USA, where 65% responded ($n=144$), a larger proportion (56%) gave low ratings of their neurologists (18). However this survey was conducted in just one region of the country compared to our national coverage.

Recall of details may not be a considerable issue in this study, as the median time from diagnosis was about 1.5 years where recollections of receiving the diagnosis are possibly still unaffected, in contrast to McCluskey et al., where on average patients were trying to remember details of what happened six years earlier (18).

Because of the anonymous nature of the survey, we cannot tell the number of neurologists involved in these patients' assessments. However, there is a nearly equal and good representation from all five Australian states to give confidence that an adequate number of neurologists are likely to have been involved in these assessments. Nevertheless, it would be valuable to have neurologists also report themselves on their practices (forthcoming article).

This survey is based on one protocol (SPIKES) and we may have obtained different results had we used other protocols. However, the European guidelines support both the positive and negative findings based on the SPIKES protocol (12). Finally, although our survey instrument has not been tested through a formal validation analysis, given that it has stemmed from the SPIKES protocol, it does possess face validity and our findings suggest that most domains within our questionnaire have good internal consistency (Table II).

In conclusion, this study, in its quantitative and qualitative components, has highlighted 'what it takes to make it better' for patients who were receiving the diagnosis, in terms of the neurologist showing more empathy, having longer consultation times and shorter follow-up periods, more referrals to MND associations, and the neurologist sharing more information. These are issues also encountered in other countries and for other life-limiting illnesses and therefore this study is likely to have implications at the international level (22,23). With over one-third of patients dissatisfied with their experience, there is room for improvement in the practice of neurologists. The following comment captures a number of areas needing improvement: "All neurologists need to be sensitive that the way they give the diagnosis will have ongoing impacts for life. 'There is no going back' applies not only to the words themselves but the way in which they are delivered. I appreciate honesty as long as it is sensitively delivered." (P205). Such improvements may be attainable through educational programmes

and the development of best practice protocols with applicability at the international level. Improving the delivery of the MND diagnosis is central to quality care and the benefits would be for both patients/families and the neurologists, as breaking such daunting news is challenging for both groups, the givers and the receivers.

Acknowledgements

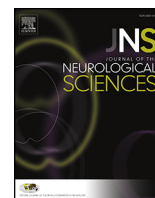
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Breaking the news of a diagnosis of motor neurone disease: A national survey of neurologists' perspectives



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ABSTRACT

Communication of the diagnosis of MND is daunting for patients and neurologists. This study aimed to establish a knowledge base of current Australian practice of breaking the news of an MND diagnosis, to assess the neurologists' educational and training needs and to compare the feedback obtained from neurologists and patients to international practice guidelines. An anonymous survey of neurologists was undertaken in Australia (2014). 73 neurologists responded to this national survey (50.4% response rate). Nearly 70% of neurologists reported finding it "somewhat to very difficult" communicating the MND diagnosis, and 65% reported feeling moderate to high stress and anxiety at the delivery of diagnosis. Compared to international guidelines, areas for improvement include length of consultation, period of follow up and referral to MND associations. Two-thirds of neurologists were interested in further training to respond to patient's emotions and development of best practice guidelines.

This is the first national study to provide a comprehensive insight into the process of delivering the MND diagnosis from the neurologists' perspective and to make comparisons with those of patients and the international guidelines. This study forms the basis for developing protocols to improve communication skills and alleviate the emotional burden associated with breaking bad news.

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1. Introduction

Receiving a diagnosis of MND is a challenge and a source of dissatisfaction for patients and their families [1–4]. The manner in which the diagnosis is given has implications for the way patients and families move from the news of their diagnosis to the actions required for support throughout the illness trajectory [5,6] and bereavement response [7].

Despite these findings, few studies have focussed on neurologists breaking the MND diagnosis to patients. A recent investigation of 25 neurology residents in the US showed that 68% thought that breaking

the diagnosis was stressful, 45% were not confident they were communicating the diagnosis effectively, and only 9% agreed they had received adequate training [8]. Additionally, analyses of video recordings of the same residents breaking the diagnosis to a patient indicated room for improvement, especially in demonstrating empathy and communication skills. The neurology residents made several suggestions for further training (e.g., practice guidelines), practice opportunities, and skills development relevant to communicating a diagnosis of MND. These findings correspond with studies showing that physicians report the delivery of bad news is a stressful experience [9,10] yet overlooked in their training [11]. These findings point to the importance of appropriate training and supervision opportunities, as well as the development and use of guidelines and protocols for the delivery of bad news.

The SPIKES protocol is a well-accepted system for communicating bad news [12]. The protocol recommends the health professional establish an appropriate Setting, determines the needs and Perceptions of the patient, requests an Invitation to give the news, provides Knowledge

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and information to the patient, explores the patient's feelings and responds Empathetically, and forms a Strategy with the patient to go forward. A similar set of guidelines for communicating with adult patients with advanced life-limiting illnesses and their caregivers was developed in Australia [13].

Given the absence of controlled trials of communicating the MND diagnosis, the American Academy of Neurology [14] concluded there was insufficient evidence supporting any particular method and noted that useful strategies have been developed for communicating a diagnosis of cancer. Best practice guidelines for MND in the United Kingdom [15] stress the need for support for patients and family carers from diagnosis so that the individual's spiritual and emotional needs may be addressed alongside medical and practical needs. Further, their standards of care emphasise that people living with MND require sensitive communication of the diagnosis combined with appropriate information about MND and support services, ensuring the provision of emotional/psychological support, a follow-up appointment within two weeks of diagnosis, and direct referral to the relevant MND association. Similarly, clinical practice guidelines developed in Europe [16,17] promote the communication of the diagnosis by a consultant neurologist with a good knowledge of the patient. The neurologist should begin by asking what the patient already knows or suspects. The diagnosis would be given in person with time available for discussion (at least 45–60 min) and complemented with printed material about the disease, relevant support and advocacy groups, and a summary of the discussion. Patients should be reassured that they will have regular follow-up visits to a neurologist within 2–4 weeks of diagnosis and supported by a specialist MND care team, where available. The neurologist should avoid withholding the diagnosis, providing insufficient or unwanted information, and communicating in a manner that is callous or removes hope.

Multidisciplinary MND clinics are considered to provide best practice coordinated care from the time of diagnosis, where specialist MND care teams deliver integrated services dealing with neurology, rehabilitation and palliative care. They have well established links to MND associations [14,18] and care is supported by regular decisions about symptom management and quality of life as the patient's condition deteriorates [19].

Reducing the physician's emotional burden associated with breaking bad news and the development of skills in communicating bad news are increasingly recognized as priorities in medical education [8]. To date, there are no Australian guidelines for communicating a diagnosis of MND and there is limited information about how neurologists actually communicate the diagnosis in Australia and elsewhere and the nature and extent of the challenges they face.

1.1. Objectives

This study aims to inform best practice in breaking bad news in the MND field. The specific objectives of this article were to:

- 1) Determine the current practice of neurologists in breaking the news of an MND diagnosis in Australia
- 2) Assess the neurologists' educational and training needs related to breaking bad news and responding to patients' emotions
- 3) Compare the neurologists' experience to that of patients undertaken in a separate survey in the same year (2014)
- 4) Assess the current practice of neurologists in breaking news in comparison to international best practice standards and highlight differences and similarities

2. Methods

The study was approved by Curtin University Human Research Ethics Committee (HR 188/2014).

2.1. Neurologists' survey

The development of the questionnaire was undertaken after a comprehensive review of the international literature in this field and with extensive consultation with clinicians and the MND associations in Australia. Moreover, the questions included in the sections on consultations and follow up were constructed to align with the international guidelines, where possible, to allow for comparisons. The neurologists' survey was advertised in The Australian and New Zealand Association of Neurologists (ANZAN) e-bulletin and website (by liaising with ANZAN secretariat). It was also advertised in the ANZAN scientific meeting in May 2014. The survey was made available to be completed online by clicking a link in the e-bulletin that goes to ANZAN members. The three neurologists on the research team trialled this version and gave comments before it was made generally available. However, in order to improve the response rate, it was also necessary to do mail-outs based on lists provided by the MND associations in every Australian state. The neurologists' survey comprised 45 questions grouped in five sections: Demographics; how patients' consultations were conducted; the communication plan and support for patients; the personal experiences in giving the diagnosis; and the neurologists' education and training needs and interest in developing best practice guidelines.

2.2. Patients' survey

The methods for the patients' parallel survey were reported in Aoun et al. [4]. The reported practice of Australian neurologists was compared to those reported by patients and addressed in the results section of this article. For every guideline, reports from both groups were compared where similar information was available between the two surveys and the international guidelines.

2.3. Analysis

Frequencies and proportions were calculated for categorical variables, and mean, standard deviation, median and range were computed for continuous and discrete variables. Neurologists' practices were analysed as a comparison according to whether they practice in an MND multidisciplinary clinic (MDC) or not. This comparison could only be undertaken in a few areas due to the sample size of those in MDCs ($n = 11$). The analysis was largely descriptive and the inferential comparisons included were those that can be compared with the international guidelines. Non-parametric tests were used for the comparisons due to the small sample sizes. Fisher's Exact Test was used for categorical variables and Mann-Whitney U for continuous variables. The statistical correction for multiple tests was Bonferroni's multiple test correction (α/n) for the comparisons between the different groups of neurologists.

3. Results

Seventy-three neurologists responded to the survey corresponding to a 50.4% overall response rate, and corresponding to 80% of neurologists who work in dedicated MDCs. The response rate was based on the lists provided by the MND associations in Australia, as the link in the e-bulletin was not favoured. Four respondents were not included in the analysis as they did not diagnose or currently care for MND patients, therefore 69 surveys were subsequently analysed.

3.1. Profile of neurologists

The mean age of respondents was 52.7 years ($SD = 10.3$), 78% were male, median length of practice was 20 years (range 1–44), 90% trained in Australia, 16% ($n = 11$) worked in a MDC and saw a median number of 10 new patients per year (range 5 to 80). Non-MDC neurologists saw

a median of 5 patients per year (range 1 to 15). The median number of current patients was 45 for MDC neurologists (range 4 to 100) and 3 for non-MDC neurologists (range 1 to 30).

3.2. Patients' consultations

The median period between first clinical consultation and diagnosis was four weeks (range 1–26), and 68% of neurologists reported requiring two consultations to convey the diagnosis. The difference in consultation times was significant between the two groups ($p < 0.001$): a mean of 23 min for non-MDC and twice as long for those in MDC (45 min) (Table 1). 78% of neurologists were always able to give the diagnosis in a private space and 41% always able to avoid interruptions. 75% tended to refer for a second opinion. While 98% of neurologists reported having a relative present during the diagnosis, 23% of them reported that on some occasions they have seen patients alone, with a difference between the two settings, although not significant (MDC = 9%, non-MDC = 26%) (Table 1). 80% of neurologists did not have any particular day of the week and timing they would refrain from giving the diagnosis (such as Friday afternoon).

At the time of giving the diagnosis, the most discussed clinical aspects pertained to: the degree of certainty of the diagnosis (94%), the course/prognosis of the disease (93%) and how the diagnosis was reached (91%).

3.3. Follow-up support

Follow-up support was always initiated by 68% of respondents within 4 weeks from diagnosis (range 1–12) with subsequent follow ups of 12-weeks interval (range 4–26); 73% reported referring to an MND association for information and ongoing support. However, referral to an MND association was significantly higher for those in MDC (100%) compared to non-MDC (67%) ($p = 0.028$) (Table 1).

The follow-up support was mainly provided by the neurologist (MDC = 91% and non-MDC = 72%); followed in non-MDC settings by the GP (40%), the MND association (31%) and then a specialist nurse (17%). However, in MDC settings, the GP took a lesser role (18%) and more of the support was provided by the MND association (55%) and the specialist nurse (36%) (Table 1).

3.4. Personal experiences in giving the diagnosis

About 70% of neurologists found communicating the diagnosis “very to somewhat difficult”, 43% found it “very to somewhat difficult” responding to patients' and/or their family members' reactions and 65% experienced “high to moderate” stress and anxiety at the diagnosis delivery (Figs. 1–3).

Table 1
Summary of comparisons in practice of neurologists (MDC and non-MDC).

| | Neurologists | | p-Value ^a |
|---|---------------|-------------------|----------------------|
| | MDC n = 11 | Non-MDC n = 58 | |
| Delivery of diagnosis (2 or more consults) | 55% | 83% | 0.052 |
| Length of consultation (minutes) | 45 | 23 | 0.001 |
| Private space (yes - always/frequently) | 100% | 96.6% | 1.000 |
| Avoid interruptions (yes - always/frequently) | 91% | 91% | 0.579 |
| Patient seen alone | 9% | 26% | 0.436 |
| Referral to MND Association for information and ongoing support | 100% | 67% | 0.028 |
| MND Association publications given | 64% | 40% | 0.190 |
| Diagnosis in writing | 27% | 21% | 0.694 |
| Follow-up support: Neurologist | 91% | 72% | 0.270 |
| Follow-up support: MND Association | 55% | 31% | 0.172 |

^a Comparison between MDC and non-MDC neurologists completed with Fisher's Exact Test for categorical data and Mann-Whitney U Test for continuous data.

The most difficult parts of discussing the news of an MND diagnosis related to: Being honest but not taking away hope (80%); dealing with the patient's emotion (38%) and spending the right amount of time (28%). The reasons for experiencing these difficulties included the lack of an effective treatment (77%), fear of causing distress (36%) and fear of not having all the answers (20%). One neurologist expressed such feelings: “Having had a migraine after each MND clinic, feeling stressed and anxious about having so little to offer, I have gradually accepted the limitations of my skills, and some confidence that assisting the patients honestly and empathetically, and not ‘abandoning’ them is of value to most patients.” (P47).

3.5. Education and training needs

When asked whether neurologists had training in “techniques of responding to patient's emotions”, 44% had no training, and 28% had learnt from sitting in with clinicians. However, 25% were very interested and 49% somewhat interested to undertake further training in such techniques. There were some differences between those interested/somewhat interested ($n = 50$) and those not interested ($n = 18$) in further training in techniques of responding to patient's emotions:

- interested neurologists in further training had been practicing for a shorter period of time (median 17 years) compared to those not interested (median 26 years), although the difference was not significant;
- more of the interested neurologists had found it difficult/somewhat difficult communicating the MND diagnosis compared to those not interested (76% vs 47%, $p = 0.049$) and more of the interested neurologists had found it difficult/somewhat difficult responding to patients' reactions compared to those not interested (55% vs 11%, $p < 0.001$);

However, one neurologist was dubious about the value of education: “Diagnosing MND is always unpleasant for both patient and physician... I think many physicians don't deliver the news well but I don't believe it is something that can be taught... Perhaps we should be selecting junior doctors to enter physician training on the basis of their communication skills? The science is easily learned. Trying to teach empathy, sincerity, and understanding is impossible. Selecting for those qualities is easy.” (P52).

3.6. Development of best practice guidelines

When asked whether neurologists had “specific training in giving an MND diagnosis”, 54% had received no specific training and 23% had sat in with clinicians. Respondents were very interested (38%) to somewhat interested (44%) in having best practice guidelines developed. There were some differences between those interested/somewhat interested ($n = 56$) and those not interested ($n = 12$) in the development of best practice guidelines:

- interested neurologists in development of best practice guidelines had been practicing for a shorter period of time (median 19 years) compared to those not interested (median 27 years), although the difference was not significant.
- interested neurologists tended to have a longer consultation time to deliver the diagnosis compared to those not interested (30 vs 18 min, $p = 0.053$).
- More of the interested neurologists had found it difficult/somewhat difficult responding to patients' reactions compared to those not interested (46% vs 33%, $p = 0.019$).

3.7. Comparison with patient feedback

The key comparisons between the two parallel surveys of patients and neurologists are summarised in Table 2.

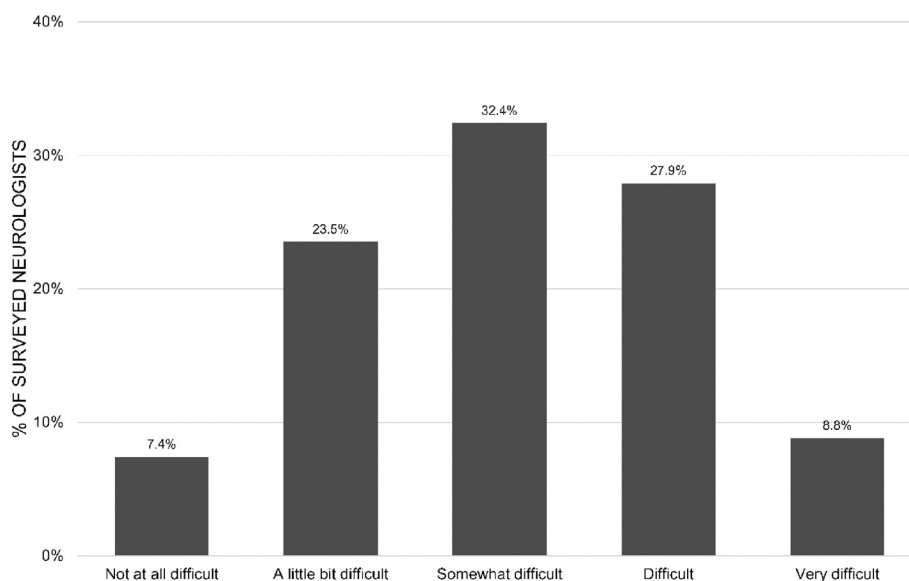


Fig. 1. Level of difficulty in communicating an MND diagnosis.

Seventy-eight percent of neurologists reported that they deliver the diagnosis in a stepwise fashion in two or more consultations, and 22% in one consultation. This is nearly comparable to 70% of patients reporting that they had at least two visits to receive the diagnosis [4]. The stepwise process of delivering the news is described as follows by one neurologist: “My practice was to let the patient know that their condition was of concern as soon as MND appeared likely (e.g., at 1st consultation) and continue to prepare patient in subsequent 1–2 consults before giving the news without taking away hope.” (P16).

Neurologists reported that in most cases a relative was present when they communicated the diagnosis, but on some occasions, 23% of neurologists saw the patient alone. This is comparable to 70% of patients reporting having a relative present with them with 21% reporting that they were alone during the delivery of the diagnosis [4].

Neurologists reported that the time spent with the patient to deliver the diagnosis was 20 min, however the length of consultation was twice as long in multidisciplinary MND clinics (Table 1). Seventy percent of neurologists reported having some level of difficulty finding enough time to discuss the diagnosis. Patients reported a median consultation

time of 30 min and 70% of them felt they had sufficient time to receive the diagnosis. Patients who rated highly the ability of their neurologists had significantly longer consultation times (40 vs 30 min, $p < 0.001$), and felt they had enough time to receive the diagnosis (84% vs 48%, $p < 0.001$) [4].

Regarding information, support and advocacy, 73% of neurologists reported providing information about and referred to MND associations, 44% gave MND associations' publications and 22% the diagnosis in writing. However only 42% of patients reported receiving information on MND associations, 24% MND association publications, 40% referral to an MND association and 16% the diagnosis in writing [4].

Concerning plans for follow-up, half of the neurologists ‘always’ encouraged patients to contact them if they have additional concerns. After receiving the diagnosis, 56% of patients reported that they were supported by an MND association, 42% by their GP, 41% by the neurologist, 28% by the MND clinic and 27% by the MND nurse [4]. Only 2.4% reported not being supported. Follow up support was always initiated by 68% of neurologists within a median 4 weeks from diagnosis (range 1–12 weeks). However, patients reported a median of 7 weeks for the first

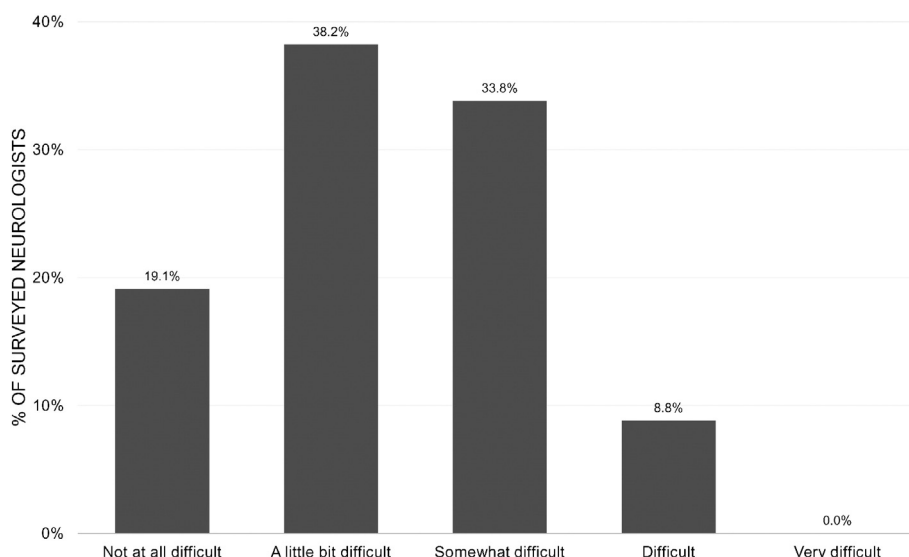


Fig. 2. Level of difficulty in responding to patient's and/or their family member's reactions (e.g., crying, anger, disbelief).

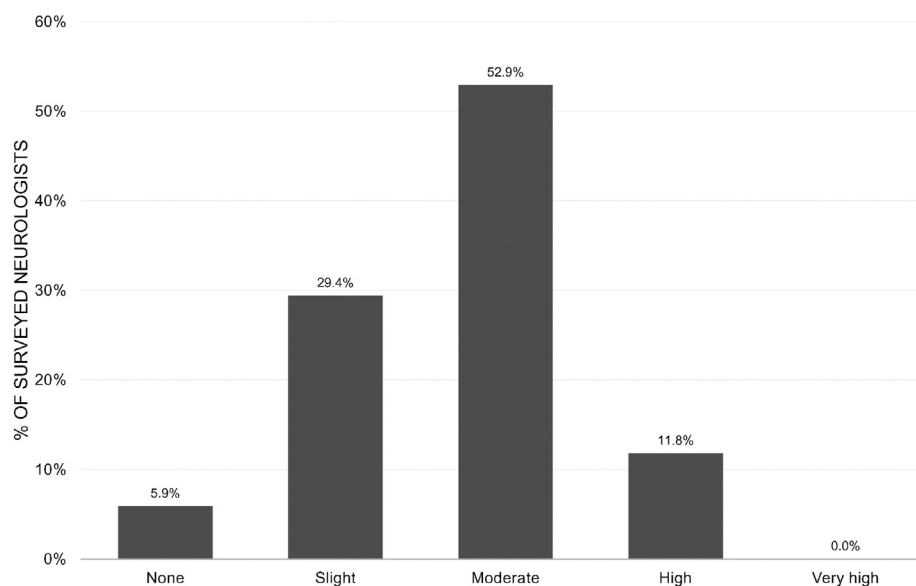


Fig. 3. Level of stress and anxiety experienced during the delivery of the diagnosis.

follow up visit after the diagnosis (range 1–52 weeks). Both neurologists and patients reported that the median interval between follow-ups was 12 weeks. In addition, 80% of neurologists did not report that they have a best day or time to deliver the diagnosis.

When it came to the setting, 78% of neurologists always gave the diagnosis in a private space, 41% always avoided interruptions. The majority of patients (96%) reported that the diagnosis was given in a completely private space/environment; and the majority (91%) had no interruptions while given the diagnosis.

4. Discussion

This is the first national Australian study to provide a comprehensive insight into the process of breaking the MND diagnosis from the neurologists' perspective, considering responses came from half of the

neurologists, who deal with MND, in the country. This sample size is comparable to that of a European study conducted in 2001, where 73 neurologists from MDCs responded (66% response rate) to a wider survey on ALS clinical management and terminal care [20]. However, our study had 80% response rate from neurologists in MDCs.

Several aspects of good practice are met in Australia from the evidence reported by the two groups (neurologists and patients) in comparison to international guidelines. Table 2 summarises the key comparisons that align to the international guidelines. As recommended, the majority (95%) of people with MND reported receiving the diagnosis from a neurologist in Australia, and the majority of neurologists reported delivering the diagnosis in a stepwise fashion. However, there is room for improvement in first ascertaining what the patient and family understand as only 43% of people with MND reported being asked how much they knew about their condition and 43% were given just enough information [4]. This would clarify the patients' understanding of their current situation and the context in which decisions about goals of care are to be made.

There is also scope for improvement in responding empathically to the feelings of patient/family when 30–40% of patients highlighted a gap in this skill, and a considerable proportion of neurologists reported difficulties in this domain. In particular, the largest significant difference between the two groups of neurologists (high and low ratings of skills) was in empathy as it was seen as an important attribute of highly skilled neurologists [4].

As recommended, the diagnosis is nearly always given in person. The Australian practice of communicating the diagnosis in 20 to 30 min falls short of the recommended guideline of 45 to 60 min. There needs to be a commitment to take more time to deliver the diagnosis and have 45–60 min available. It is evident that the longer the patients spent with their neurologists during breaking the diagnosis, the more they were satisfied with the delivery process and the higher they rated the neurologists' ability/skills [4]. There is room for improvement in making sure patients are not seen alone and are advised to have a support person with them when receiving the diagnosis.

There needs to be a routine practice for all neurologists to refer to MND associations (as per the European guidelines), as 27% of neurologists did not refer and 60% of patients reported not being referred. In addition, it would be useful if all neurologists gave the diagnosis in writing as this would help the patient and family communicate the diagnosis to support organisations (such as the MND associations) and health professionals involved in their care.

Table 2
Neurologists' reported practice in delivering the diagnosis compared to the experience of people with MND from the parallel survey and the EFNS Guidelines.

| | People with MND [4] N = 245 | Neurologists n = 69 | EFNS guidelines [16] |
|---|--------------------------------|------------------------|----------------------|
| Diagnosis by neurologist | 95% | – | Always |
| Delivery of diagnosis (2 or more consultations) | 70% | 78% | Step-wise |
| Length of consultation (minutes) | 30 | 20 | 45–60 min |
| Private space (yes – always/frequently) | 96% | 97.1% | Always |
| Avoid interruptions (yes – always/frequently) | 91% | 91% | Always |
| Patient seen alone | 21% | 23% | Never |
| Referral to MND Association for information and ongoing support | 40% | 73% | Always |
| MND Association publications given | 24% | 44% | Always |
| Diagnosis in writing | 16% | 22% | Recommended |
| Enough information given | 43% | – | Always |
| Asked of any previous MND knowledge | 43% | – | Always |
| Follow-up support: Neurologist | 41% | 75% | Always |
| Follow-up support: MND Association | 56% | 35% | Always |
| Diagnosis to follow-up (median, weeks) | 7 | 4 | 2–4 weeks |
| Diagnosis given with warmth, care & empathy | 67% | – | Always |
| Sufficient time to express emotions | 63% | – | Always |
| Sufficient time to have emotions responded to | 62% | – | Always |

As recommended, patients were supported by a range of health professionals. The Australian practice of a close follow up visit of 4–7 weeks from diagnosis falls short of the recommended guideline within 2–4 weeks or sooner. In addition, it was interesting to find out that the vast majority of neurologists did not mention that, for example, Friday afternoons were not appropriate to deliver the diagnosis where there was little opportunity for patient support at the weekend.

Although there were a few respondents from MDCs to make meaningful comparisons, findings point to the practice of neurologists involved in MDCs being more aligned with the international guidelines in terms of length of consultation and other indicators listed in Table 1. Borasio et al. [20] highlighted that standards are usually higher in areas with specialized ALS centres in Europe. In a recent Australian study [21,22], specialized multidisciplinary MND care was found to facilitate patient engagement with clinicians in decision-making by providing an optimal environment for information provision, support, and planning, stability and care continuity. One neurologist in this study commented that: *“The major issue is lack of access to multidisciplinary MND clinics for further management esp. in rural areas. It is very difficult as a neurologist in private practice to coordinate and access the multidisciplinary care required esp. for patients who live at a distance”*. (P45).

The feelings of stress and anxiety associated with delivering the diagnosis are comparable to those reported in the literature [8,11]. However, it is worth noting that the impact of delivering a diagnosis may be milder or more severe depending on the types of MND, the prognostic differences between them and the genetic implications where applicable, and this is worthy of further research. While the neurologists' survey in this study did not collect such information, the findings from the patients' survey indicate that 69% reported having cervical/lumbar symptoms at onset, 19% had bulbar symptoms and the rest a combination of symptoms [4]. Although the two anonymous surveys were undertaken in the same year, we cannot ascertain how many of the responding neurologists were involved with the patients in the parallel survey.

The needs for education and training in communicating the diagnosis are comparable to those reported in the literature [8,11]. Essentially, two thirds of responding neurologists (46/68) were interested in both further training responding to patients' emotions and development of best practice standards. It is worth noting, and perhaps expected, that those interested had reported more difficulty in communicating the diagnosis. Therefore, it is recommended that the peak bodies (MND Australia and ANZAN) consider education and training programs aimed at improving the skills of neurologists and neurology trainees in responding to patients' emotions, based on the evidence from this study. It is also recommended that these two peak bodies develop MND specific best practice standards in communicating the MND diagnosis, based on the evidence from this study and existing international protocols.

4.1. Limitations

Limitations include that the questionnaire was not validated other than by consensus from experts that included three neurologists, a palliative care physician, two nurses, a psychologist and representatives from MND associations. However, the questions included in the sections on consultations and follow up were constructed to align with the international guidelines where possible. Due to the anonymous nature of both patient and neurologist surveys, we could not ascertain how many of the responding neurologists were involved with patients in the parallel survey. Therefore, the comparative analysis between the two groups was descriptive, and those indicators aligned with the international guidelines were mainly included.

There may have been a selection bias and those neurologists who see only a few MND patients may have had a less stressful experience and therefore may have opted not to participate. However, it was

intentional to include neurologists with a frequent experience with the disease, as the introductory part of the questionnaire had a statement that said: “If the number of patients in questions 7 (number you diagnose per year) and 8 (number you currently care for) is zero, then there is no need to proceed with the rest of the questionnaire and we thank you for your time”. In addition, the cohort of neurologists who responded consisted of older and experienced neurologists (median length of practice of 20 years), with possibly the findings not reflecting the experience faced by “young” neurologists.

As this article reported on many aspects of delivering the diagnosis, it was deemed impractical to incorporate a theoretical framework for every aspect. Future qualitative articles from this study will focus on single aspects with corresponding theoretical frameworks, where appropriate.

5. Conclusions

To our knowledge, this is the first study to take into account the perspectives of both neurologists and patients and to address an important knowledge gap in the clinical care of individuals with MND. This study added to our understanding of the process of delivering a diagnosis of MND and the pressures it placed on neurologists in terms of stress and anxiety. The comparison between neurologists' experiences and those of patients provided a novel view of the topic. Interpreting the findings in relation to accepted international guidelines for care provided a sound benchmark against which to judge the extent to which neurologists in Australia are achieving recognized standards and pointed to areas in need of improvement. We believe that this study has filled a gap in the literature as one respondent expressed the following remark which was representative of many: *“I am glad that you are doing this questionnaire. I think that more research needs to be done in this area - so well done! Happy to support in any way I can”* (P17). This study could form the basis for improving practice to alleviate the emotional burden associated with breaking bad news as poor communication increases the risk of burnout and fatigue [5]. This is achievable by the two peak bodies in MND and Neurology, MNDA and ANZAN, instigating educational programs and developing standards and protocols with applicability at the international level.

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ARTICLE

"Until there is a cure, there is care": A person-centered approach to supporting the wellbeing of people with Motor Neurone Disease and their family carers

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Abstract

Background: There is a lack of interventional studies to support the psychosocial wellbeing of people with MND (PwMND) and their family carers. Moreover, their experiences with the models of care already provided by ALS/MND voluntary organisations have not been well investigated.

Objectives: To report on a person-centered model of care, the MND Advisory Service, designed to support the needs of PwMND and their family carers, to explore their experiences with the service they received and to identify which aspects of this service were most needed and valued by the service users.

Methods: Participants were recruited from the MND Association in Western Australia. PwMND and carers were invited to separately complete anonymous postal surveys (2015-17), using a mixed method design comprising quantitative and open-ended questions.

Results: The average response rate across the 3 years was 38% (138 patients and 117 carers); 84-89% of both groups felt more supported and cared for as a result of the service and 79-82% felt they were able to make more informed decisions to manage their health and wellbeing. Ninety percent of both groups found the MND Advisory Service of high value practically and emotionally, especially the personal contact and time dedicated to the visit, with 86-88% stating that the service had met their expectations.

Conclusions: Two unique features of this service, not provided by other services to the same extent, are the emotional benefit to both groups and the particular focus on the family carers' needs. Until such person-centered models of care are properly investigated through the experiences of their users, erratic changes in care funding will pose a threat to their effective operation and even viability. The drive to find a cure should not detract from the fact that PwMND and their families still need to be supported physically and psychologically until then.

Keywords

Case management, emotional burden, family carers, MND/ALS, MND Advisory Service, MND Associations, models of care, person-centered healthcare, physical burden, psychological burden, psychosocial care, satisfaction, support groups, support needs

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Introduction

Although Motor Neurone Disease (MND) is relatively uncommon, with a prevalence of 7 in 100,000, the associated direct (e.g., healthcare, expensive equipment) and indirect (lost productivity and income) costs and intangible losses (independence, quality of life) contribute to the high cost of this illness to Society. According to the Deloitte Access Economics report [1], the total cost of

MND is estimated to be \$1.13 million per person with MND in 2015, the total cost in Australia estimated to be \$2.37 billion with productivity costs comprising 38% of the costs. The per-person costs of MND are substantially higher than a number of other diseases. About 2,000 people live with MND each year in Australia.

Notwithstanding the physical, psychological and emotional burden of the disease on MND family carers, the Deloitte report has quantified the economic disadvantage

on families supporting people with MND (PwMND), who provide an estimate of 7.5 hours of informal care per day to PwMND. The productivity loss due to such informal care in Australia was estimated to be \$68.5 million in 2015, or \$32,728 per person, with individuals shouldering most of these costs (\$44.0 million) and with government bearing the rest (\$24.5 million) [1].

With this background of adverse social and economic impact of the disease, a recent literature review lamented the lack of interventional studies, particularly for the psychosocial wellbeing of patients, their family carers and their social networks [2]; this was also reported in an earlier review [3]. Nevertheless, in many developed countries, voluntary support services, such as the ALS/MND Associations, play a central role in the delivery of case management services to people with MND and their families, coordinating and supplementing the care that patients and their families receive from health services [4-6]. Even so, such interventional models of care have not been well researched.

In Australia, state-based MND advisory services provide a range of services to PwMND and family carers, including information and education for PwMND, family carers and health professionals, equipment provision, fundraising, support and advocacy. MND Advisors visit PwMND in their homes to assess their current needs and to assist their connection with healthcare services. In metropolitan areas, MND Advisors form part of MND multidisciplinary clinic teams [7], coordinate support activities with other team members and support PwMNDs' access to external services. In regional and rural areas, MND Advisors act as a link between patients, families and local health services, to connect patients to services and to educate health professionals in the unique needs and priorities of PwMND. Advisors frequently travel long distances to provide this home-based support in rural areas. Australian MND Associations' model of service delivery is comparable to some other international ALS/MND associations that offer direct services to patients and carers, but also differs from many associations that act purely as self-help or support groups [8].

Experiences of people with PwMND and their families accessing case management-based care from MND advisory services, have not, to date, been well investigated. A few studies in fields other than MND reported the positive aspects of such a service, being accessibility, time and personal contact [9,10]. Additionally, studies have shown that PwMND and their family carers feel their needs for emotional support are not always met by their interactions with healthcare professionals [11,12]. This is despite many studies reporting that emotional support is one of the primary expectations of service users in MND care [12-16]. Family carers participating in a person-centered intervention that systematically identified and addressed their support needs from the MND Advisory Service reported that it provided them with an opportunity to share their difficult experiences, to gain increased insight into emotional concerns and enhanced awareness of supports, acknowledging their role as caregivers [17]. In addition, participants described the benefits related to more timely access to support and improved links to resources [17].

Findings from a recent Dutch study exploring the value and need of case management in MND care shed more light on the appreciated aspects of such a service [18]. These included the combination of house calls and time dedicated to these visits providing a more person-centered care, the proactive approach anticipating future needs and preparing for what to expect next and also emotional support which enhanced the feeling of safety and coping with their situation. Therefore, investigating experiences and measuring satisfaction with such community-based models of case management is crucial if services are to continually improve their responsiveness to the needs of those they serve.

The objectives of this study are to report on a person-centered model of care designed to support the needs of PwMND and their family carers in the community and to explore their experiences with the services they receive from the MND Advisory Service in Western Australia. In particular, we aimed to identify which aspects of this service were in most need and of value to the service users, to better inform and tailor services.

Methods

Ethical approval was granted from Curtin University Research Ethics Committee (HRE2017-0133). Anonymous surveys were undertaken in the years 2015, 2016 and 2017.

The MND Advisory service of the MND Association of Western Australia (MNDWA) is partly funded by the state department of health, but mostly from fundraising to be able to provide the following services through home visits and telephone and e-mail contacts:

- Provision of information regarding the disease
- Provision of information regarding services and supports
- Emotional support
- Equipment provision and coordination
- Care coordination including assisting with accessing respite/long term residential care
- Education for both client and carer throughout progression of their disease
- Access to counselling
- End of life support

This study adopted a mixed method design consisting of quantitative questions and open-ended questions, for respondents to give their qualitative feedback on their experiences. The two versions of the survey were:

- Satisfaction with MND CARE-People with MND
- Satisfaction with MND CARE-Family Carers

These surveys were modified from the FAMCARE-2

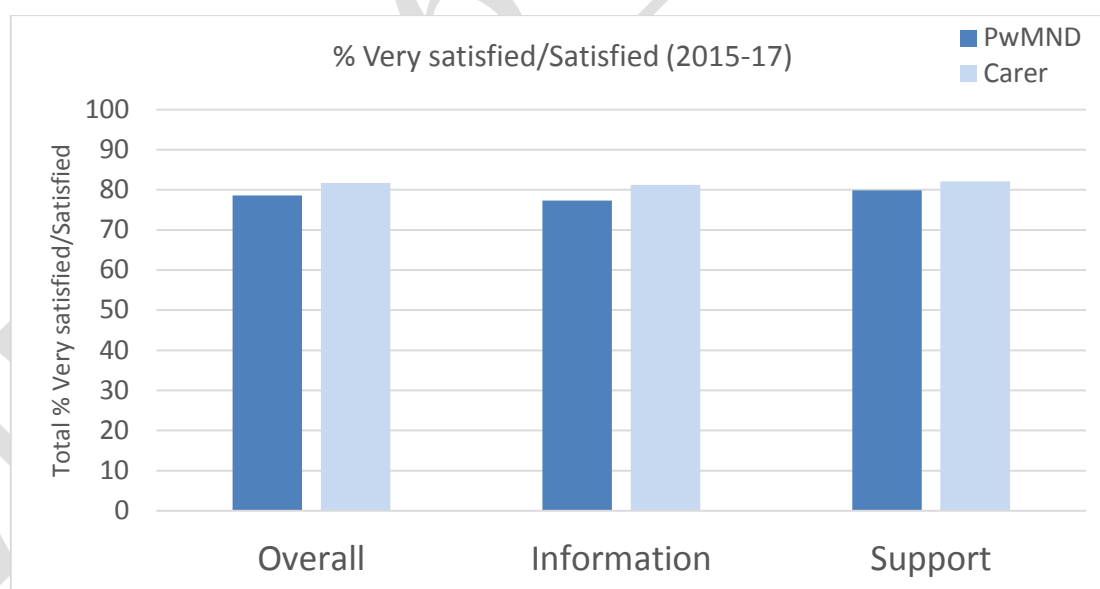
Table 1 Satisfaction-Information subscale 2015-17

| | PwMND | | Family Carers | |
|--|---------------------------|------|---------------------------|------|
| | Very satisfied/ Satisfied | | Very satisfied/ Satisfied | |
| | n | % | n | % |
| The information given regarding the Association's supports and services | 131 | 94.1 | 114 | 98.3 |
| The introduction by the MND Advisors explaining their role and how they can assist | 123 | 90.9 | 108 | 92.1 |
| The information provided by the MND Advisors regarding the disease, its progression and what to expect | 115 | 77.4 | 97 | 84.7 |
| The information provided as to how to access counselling | 109 | 80.5 | 83 | 80.4 |
| The information provided as to how to access respite | 89 | 70.9 | 63 | 71.5 |
| The information provided on how to access services relating to symptom management | 108 | 79.9 | 83 | 76.4 |
| The information by the MND Advisor with regard to accessing long term residential care | 100 | 56 | 84 | 67.1 |
| The introduction to Advance Health Directive by your MND Advisor | 84 | 68.9 | -- | -- |
| The education programs provided to the family carer | -- | -- | 78 | 79.6 |

Table 2 Satisfaction Support subscale 2015-17

| | PwMND | | Family Carers | |
|---|---------------------------|------|---------------------------|------|
| | Very Satisfied/ Satisfied | | Very satisfied/ Satisfied | |
| | n | % | n | % |
| Relationship with your MND Advisor | 122 | 89.9 | 102 | 88.8 |
| Meetings with the MND Advisor regarding the plan of care | 108 | 79.9 | 82 | 81.3 |
| The level of information and education provided as needs change | 101 | 72.1 | 86 | 79.5 |
| The emotional support provided to family carer and other family members | 100 | 75.9 | 82 | 79.8 |
| The emotional support provided to PwMND | 102 | 74.2 | 82 | 78.3 |
| The provision and coordination of equipment | 109 | 86.3 | 91 | 87.7 |
| The extent to which the family is included in discussions | 107 | 80.8 | 83 | 82 |
| The MND Advisors' response to changes in the care needs | 65 | 79.9 | 51 | 79.3 |

Figure 1 Summary of satisfaction for PwMND and family carers



Scale questionnaire, which is widely used in Palliative Care [19]. Responses range from 5 (being very satisfied) to 1 (being very dissatisfied), with a response option available if the item is not relevant to the respondent's situation. In consultation with MNDWA, the 2 surveys were adapted to cover the key services provided by the Association, which fell into 2 groups: (a) provision of information (8 items) and (b) provision of support (8 items) in several

aspects of needs. Reliability analyses were undertaken using Cronbach's α for the information subscale and the support subscale.

Additionally, the survey incorporated 2 impact questions: to what extent PwMND and their family carers agreed that, as a result of the service, they felt:

- cared for and supported in the community to

maintain the best possible quality of life throughout the progression of the disease

- supported to make informed and better decisions to manage their health and wellbeing throughout the progression of the disease through accessible, understandable and timely information.

PwMND and their family carers were also invited in the 2017 survey to rate the value of the MND Advisory Service to them, both practically and emotionally, by considering the following supportive aspects and adding other aspects according to their experience: (1) being visited at home; (2) personal contact; (3) the time dedicated to the visit; (4) a proactive approach anticipating patient needs; (5) practical support and (6) emotional support. Response categories were: 'a little', 'quite a bit' or 'a lot'.

All patients registered with the MND AWA were invited to participate in this annual survey. Approximately 145 patients are registered with the MND AWA each year, but PwMND who are pre-terminal at the time of the annual survey are excluded, as per advice from the MND Advisors. Questionnaire packages were posted by MND AWA containing an invitation letter explaining the study and bearing the letterhead of the Association, one patient survey and one carer survey with a reply-paid envelope addressed to the research team. PwMND and family carers were encouraged to complete the surveys independently and those requiring assistance from their family were advised to express their own opinions.

Descriptive statistics for variables were calculated: frequencies and proportions for categorical variables; means, standard deviations, medians, minimums and maximums for continuous variables. All statistical analysis was conducted using SPSS Version 24. The open-ended responses were categorized to 2 support domains: practical or emotional.

Results

The results are based on a total of 138 patients and 117 family carers who responded to the survey across the 3 years (Tables 1 and 2). Satisfaction rates were computed as the average of the 3 years. The average response rate was 38% for PwMND and no response rate could be computed for family carers as not all PwMND had a family carer. Across the 3 years, the Cronbach's α for the information subscale ranged from 0.963 to 0.856 for patients and from 0.96 to 0.861 for carers. The Cronbach's α for the support subscale ranged from 0.969 to 0.896 for patients and 0.96 to 0.856 for carers. The subscales achieved a high level of internal consistency implying that all items in the subscales essentially measured the same construct which is the satisfaction with the service.

Experiences of PwMND

PwMND had an overall satisfaction rate of 78.6% regarding the services they received from MND AWA:

77.3% were satisfied with the overall information they received (Table 1) and 79.9% were satisfied with the overall support they received (Table 2). Items below these average satisfaction levels were in the information subscale and were about accessing long-term residential care and the introduction of an advance health directive. As a result of the service, 84.2% of PwMND agreed/strongly agreed that they felt more supported and cared for and 78.7% of PwMND agreed/strongly agreed that they were able to make more informed decisions to manage their health and wellbeing.

Experiences of Family Carers

Family carers had an overall satisfaction rate of 81.7% regarding the services they received from MND AWA: 81.3% were satisfied with the overall information they received (Table 1) and 82.1% were satisfied with the overall support they received (Table 2). As a result of the service, 88.6% of family carers agreed/strongly agreed that they felt more supported and cared for (as a family carer) and 81.9% of family carers agreed/strongly agreed that they were able to make more informed decisions to manage the health and wellbeing of their care recipient. Figure 1 summarises the 2 subscales of satisfaction for the 2 groups.

Profile of respondents in 2017

The profile of respondents (who responded to the survey in 2017) did not differ significantly from the profile of the population of PwMND who received the survey in terms of age (mean age for population = 64.1yrs and sample = 68.4yrs), gender (percent male for population = 62.2% and for sample = 63.5%) and location (percent regional and rural for the population = 19.6% and for sample = 15.4%). The majority of PwMND were married, with 13.5% divorced or widowed. The median time from diagnosis to survey completion was 26 months and the median period from symptom onset to diagnosis was 10 months (Table 3). The proportion of patients over 65 years who responded was 64.7%. Most family carers were female (74.4%), married (93%), spouses (79.1%), with a mean age of 62.6 years. Responses came from 43 dyads (patients/carers in same family) and 9 patients did not have corresponding carers (Table 3).

Value of the MND Advisory Service

Most of the respondents in the 2017 survey (80-95% of PwMND and family carers) found all aspects of the MND Advisory Service to be of high value to them practically and emotionally, with the highest proportions for PwMND (90%) and carers (95%) valuing the aspects of personal contact and the time dedicated to the visit (Table 4). About 87% of patients and 88% of carers reported that the service

Table 3 Demographics of People with MND and their Family Carers (Year 2017)

| | | PwMND | | Family Carers | |
|---|----------------------------|-------|---------|---------------|---------|
| | | n=52 | | n=43 | |
| | | n | % | n | % |
| Gender | Male | 33 | 63.5 | 11 | 25.6 |
| | Female | 19 | 36.5 | 32 | 74.4 |
| Age (Years) | Mean (SD) | 68.4 | 10.26 | 62.6 | 11.38 |
| | Median (Min., Max.) | 68 | 36, 85 | 64 | 35, 8 |
| Marital status | Never married or single | 2 | 3.8 | 0 | -- |
| | Married or <i>de facto</i> | 40 | 76.9 | 40 | 93 |
| | Separated or divorced | 7 | 13.5 | 3 | 7 |
| | Widowed | 3 | 5.8 | 0 | -- |
| Patient - carer dyad | Yes | 43 | 82.7 | 43 | 100 |
| | No | 9 | 17.3 | 0 | -- |
| Postcode (Region) | Metropolitan | 43 | 82.7 | 37 | 86 |
| | Regional/Rural | 8 | 15.4 | 6 | 14 |
| | Unknown | 1 | 1.9 | 0 | -- |
| What is your relationship to your relative/friend with MND? (family carers) | Wife/husband or partner | - | - | 34 | 79.1 |
| | Girlfriend/boyfriend | - | - | 1 | 2.3 |
| | Sister/brother | - | - | 1 | 2.3 |
| | Daughter/son | - | - | 3 | 7 |
| | Other relative | - | - | 3 | 7 |
| | Friend | - | - | 1 | 2.3 |
| Time between symptom onset and diagnosis (months) | Mean (SD) | 21 | 27.5 | 22.4 | 22 |
| | Median (Min., Max.) | 10.1 | <1, 137 | 15 | <1, 113 |
| Time between diagnosis and survey (months) | Mean (SD) | 48 | 76.14 | 38.1 | 54.66 |
| | Median (Min., Max.) | 26 | 2, 455 | 18.4 | 2, 298 |

Table 4 Value of MND Advisory Service (percentage quite a bit/a lot)

| | PwMND (%) N=52 | Family Carer (%) N=43 |
|--|----------------|-----------------------|
| Being visited at home | 89.3 | 90 |
| The personal contact | 90.2 | 95.1 |
| The time dedicated to the visit | 91.7 | 95.1 |
| The proactive approach anticipating your needs | 85.7 | 92.3 |
| The practical support | 88.2 | 92.7 |
| The emotional support | 79.6 | 84.6 |

Box 1 Practical Support

People with MND

The practical support of patients was manifested in areas such as financial support; provision of bed and walker; advice about saliva management; setting up a disabled bathroom and access to disabled equipment; provision of afternoon respite; supply of equipment; payment for C-PAP machine; provision of a portable cough assist machine to allow travel; provision of a sling; woollen pillowcases to prevent sore ears; loan of an i-pad; sorting a doctor's appointment; coordinated the volunteer transport service - bus service; physiotherapy advice for neck support; response to queries chasing a [government funded support program]; coordinated with hospital to fix a blocked tube in half an hour while previously it took an admission to hospital.

Family carers

The family carers reiterated all the practical support already mentioned by the patients. In addition, the carers mentioned in particular the carers' courses:

- "By running carers' courses to educate and meet other carers has been a great help"

Others mentioned a series of practical supports such as:

- "fantastic file of information; very quick equipment loan; social and info sessions for patient and carer family which we intend to attend for peer support; regular clinic"
- "(1). Set up regular respite three afternoons per week through a Federal grant program I don't know about. (2). Advocated strongly for my husband to be accepted by the [government funded support program] - which we didn't know existed. (3). Organised a hospital bed which made it possible for [government funded support] services to commence. (4). Organised a carer course which was very helpful. (5). Did some research and found us a nursing service that was prepared to support us with a non-routine bowel management technique"

Box 2 Emotional support

| People with MND | Family carers |
|---|--|
| <ul style="list-style-type: none"> - "They always listen and are very supportive" - "Keeps in touch regularly" - "Being able to ring them always and my carer now is great" - "Keeping me on a level footing" - "Visits are a great feeling"; "being available for visits and calls" - "By giving me and my family the care that we needed, with the service that was needed straight away". - "Just knowing they are there for us". - "Emotionally - you do need a backbone to construct a stable foundation". - "I feel emotionally supported in all ways" - "The information and support they provide make all the difference" - "Prior to their contact I was "in the dark" as to what do; who to contact etc." - "Professional, supportive, always there, knowledgeable" | <ul style="list-style-type: none"> - "Emotionally - legitimising Dad's illness/demystifying MND. Emotionally - the genuine level of caring and interest". - "Being on the end of the phone" - "Always there when needed" - "General understanding/caring attitude". - "Very simply they are great". - "Referral to counsellor - invaluable for ongoing support regarding coping mechanisms". - "They helped me with all the support and how to cope with a tragic situation". - "Just knowing that you can contact someone and speak to them concerning anything makes it easier". - "Always available for phone discussions". - "General go to person with queries on most MND matters". - "Knowing they're there for any help, support or otherwise. The MND nurse is a valuable asset and is always ready to assist when required" - "Being able to talk on the phone and emailing for advice, or just to update on condition of health". |

met their expectations 'quite a bit/a lot' and 80% did not think that anyone else could have provided the same

services they received from the MND Advisory service of the Association. The high value put on the service was

reflected in patients and carers' comments such as:

"Professional, supportive, always there, knowledgeable"; "A valuable asset"; "Prior to their contact I was in the dark" (Boxes 1 and 2).

Discussion

This study assessed the perceived benefits of the MND Advisory Service from the perspective of PwMND and their family carers, to continuously tailor the service to the need of these service users and better inform allocation of resources. Satisfaction rates were high at 80%, with PwMND and their carers reporting making more informed decisions to manage their health and wellbeing as a result of the service. To our knowledge, this is the first study to combine quantitative and qualitative analyses to assess the impact of a community-based model of care for MND. The quantitative component derives from an evidence-based palliative care tool that has been adapted to MND care and the subscales achieved a high level of internal consistency. This implies that all the items in the sub-scales essentially measured the same construct, that is, satisfaction with the service.

It is remarkable that 90% of patients and family carers find the MND Advisory Service of high value to them practically and emotionally; in particular, valuing the aspects of the personal contact and the time dedicated to the visit. It is worth noting two unique features to this service that are not provided to the same extent by other services: the emotional benefit to PwMND (in addition to the practical benefit) and the focus on the needs of the family carers, two aspects called for in the literature [2,3,17]. The needs of carers in this study mirror those articulated in another study, where PwMND highlighted their need for psychological support to manage their fears and those of their family members during the course of their illness [20]:

"[having] ability to discuss fears and especially a lengthy dying process with psychological support. The devastating impact on my husband and how he would cope. I would like to discuss my longevity as my fear of life is greater than death."

This person-centered model of care has been a regular feature of the MND Associations in Australia, yet it is used in only a surprisingly small number of countries, or as a one-off trial in The Netherlands [18]. From the findings of this study, MND Associations are providing an exemplary and relatively unique service to PwMND and their families.

However, the viability of this model of care may be at risk. Changes to care funding could pose threats to this effective service delivery that improves patient care and quality of life of patients and their families. For example, the introduction of the National Disability Insurance Scheme (NDIS) in Australia has the potential to complicate the provision of specialised care and support services to neurodegenerative conditions such as MND [21]. There are already numerous barriers to patients and carers accessing well-timed, specialised services in

regional and rural areas including isolated health professionals who lack experience in MND [7,22]. This is reflected in our survey where rural patients were less satisfied mainly due to the lack of access to such supportive services in rural areas. Anecdotally, the introduction of the NDIS for PwMND experiencing rapid disease progression has given cause for concern and needlessly distracted from multidisciplinary clinics and MND Associations working together to deliver seamless and coordinated specialised care. The development of NDIS care plans with care planners who lack understanding of the rapidly changing needs of PwMND means that plans based on a patient's initial presentation at the time of diagnosis become rapidly out of date. MND Advisors have indicated that much of the time previously devoted to providing direct support to PwMND and family carers is now taken up with assisting them to negotiate and update their plans.

Additionally, there are inequities in funding provision for PwMND. While those under the age of 65 are entitled to NDIS funding, PwMND over 65 years can only access the much lower support from aged care services. As approximately 40% of MND patients are diagnosed after 65 years of age, many older PwMND are likely to experience inequitable access to care services. Moreover, changes to MND Associations business models may result in inequity between services state by state in Australia. As service providers with the NDIS, rather than registered charities, MND Associations must now attract health funding to be an NDIS provider. Not obtaining this funding limits MND Associations' capacity to participate in the funded care market and deliver an equitable, coordinated service to all PwMND and their families.

A further risk to this model of care lies in services' capacity to obtain charity funding as competition for funding increases. Case management-based programs such as the MND Advisory services rely on donations to provide care to PwMND and family members, regardless of their eligibility for government funding packages. Research funding for MND is also dependent upon public donations. As with other life-limiting conditions, there is a strong emphasis on finding a cure, or developing treatments that can effectively slow the disease and extend survival. While this is very important, it should not however come at the expense of funding research or programs that improve the quality of life of PwMND and their family members. *"Until there is a cure, there is care"* (the motto of MND Australia and other Associations), people living with MND should not have to compromise on services and research that improve their quality of care and support because of funding shortfalls. One such person-centered carer intervention (the Carer Support Needs Assessment Tool or CSNAT) has significantly reduced caregiver distress and strain in the cancer field [23] and when piloted in the MND field [17] it demonstrated that it gave carers a sense of validation, reassurance and empowerment. This focus on carers is important, with Miles and Asbridge [24] emphasizing that carers are "vital partners in increasing the person-centeredness of health and social care systems". Furthermore, the CSNAT provided MND Advisors with better structure, focus and improvement to their standard practice, thus

recommending that such, person-centered, evidence-based research needs to be integrated in routine practice on a larger scale. Therefore, further research into successful interventions for the psychological wellbeing of PwMND and family carers and how these could be extended to meet their needs, as more becomes known about the disease, is needed.

This study has its limitations in that it examined satisfaction with an MND Advisory Service in one state of Australia. Although small scale, it indicates that person-centered models of care addressing practical support and psychological wellbeing are valued by PwMND, aligning with international findings [18]. The response rate of 38% is considered better than the average rate of postal surveys (about 20%) and higher than the response rate to a recent MND national survey (29%) [13]. Furthermore, it is three years' worth of data and there is a little overlap between the three cohorts, as mostly different clients have responded every year, with the same 17 responding in 2016 and 2017 and five of those in the three years.

Conclusion

Until such person-centered models of care are consistently investigated through the experiences of their users, erratic changes in care funding will pose a threat to their effective operation and even viability. The drive to find a cure should not take away from the fact that PwMND and their families still need to be supported and cared for physically and psychologically until then. Larger scale studies are needed to build on these findings, towards developing international guidelines for quality of life, in tandem with those for symptom management. These are also needed to alleviate the physical, psychological and economic burden of the disease on family carers as PwMND are mostly cared for at home. The National Voices report on Person-Centred Care [25] recommended that patients should be "treated in a way that recognizes and respects the outcomes that matter most to them" and to "make person-centred and community-based approaches part of normal business". This person-centered approach fits with the current international emphasis on the subjective experience of the illness, encompassing the practical, social, emotional and existential concerns [26], which have been the remit of MND Associations.

Acknowledgements and Conflicts of Interest

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