

Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.
<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>

Title page

Full title: Experiences of living with motor neurone disease; a review of qualitative research

Running title: Experiences of living with MND; a review

Contributing authors:

Dikaios Sakellariou, Mr. School of Healthcare Studies, Cardiff University

Gail Boniface, Dr. School of Healthcare Studies, Cardiff University

Paul Brown, Dr. School of Healthcare Studies, Cardiff University

Corresponding author:

Dikaios Sakellariou

School of Healthcare Studies, Cardiff University

Ty Dewi Sant. Heath Park Campus

Cardiff CF14 4XN

UNITED KINGDOM

Email: sakellarioud@cardiff.ac.uk

Abstract

Purpose. This review sought to answer the question ‘what is known about people’s experiences of living with MND?’.

Methods. The review followed the guidelines of the Centre of Reviews and Dissemination. Twenty articles met the inclusion criteria and their results were analysed thematically. Data were managed and coded using the software package NVIVO and the analysis was performed in two stages, with the first stage aiming to develop descriptive themes offering an overview of the included data. During the second stage analytical themes were developed with the explicit aim to answer the review question.

Results. The themes that emerged point to the following: a) people with MND develop experiential knowledge that helps them to live with the disease and b) while people with MND believe they do not have any control over the disease, they try to have control over their lives through active choices, e.g. how and when to use adaptive equipment.

Conclusions. This review highlights the decision-making and knowledge generating processes used by people with MND. Further research is required to explore these processes and their implications for the care of people with MND.

Keywords: motor neurone disease, care preferences; patient involvement; illness experience

Experiences of living with motor neurone disease; a review of qualitative research

Introduction

Motor neurone disease is an adult onset, neurodegenerative condition that is characterised by the wasting of voluntary muscles secondary to destruction of motor neurones leading gradually to partial or complete paralysis (1). People living with MND require access to a wide range of healthcare services and their care needs change gradually but continuously requiring ongoing adaptations. Symptoms that can develop in the course of the disease include dysarthria, spasticity, sleep disturbances, pain, emotional lability, fatigue, constipation, cognitive impairment (predominantly frontotemporal dementia) and depression (2-5). As muscle groups become weaker and waste away, activities such as writing, walking, lifting, swallowing, coughing, talking and breathing become difficult and eventually cannot be carried out. Mental capacities are usually, although not always, left intact and people are aware of the unfolding condition. Eventually the muscles controlling breathing become affected and death often occurs due to respiratory failure (6).

Literature suggests that the needs of people living with MND are not always effectively met (7-9). How life is experienced, how people make sense of MND, what their priorities are and how they work towards them is not clear. Furthermore, a Cochrane review on multidisciplinary care (10) for people with MND evidenced a divergence of perspectives between healthcare professionals and people living with MND. Exploring the experience of people with MND can be challenging because the disease can progress very quickly, leading to paralysis and communication

Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.

<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>

challenges. This article presents a review of qualitative research relating to experiences of people living with MND.

Methods

Design

The review was performed in a systematic way and followed the process for review of qualitative studies developed by the Centre of Reviews and Dissemination (CRD) (11). The process of the review is presented in figure 1. The aim of this review was to provide an answer to the question of what is known about people's experience of living with MND. DS and GB developed the review protocol. DS performed the literature searches, extracted and analysed the data. GB and PB critically discussed the review process and the results. DS wrote the first draft of the article and all authors participated in the write-up of the final draft.

Inclusion criteria

Inclusion criteria were that articles reported on the lived experience of MND from the patients' perspective; used research designs that allowed in depth exploration of personal experience (qualitative designs); were published in a peer reviewed journal accessible through electronic databases; and were written in English. Articles that reported on the experience of both people with MND and their carers were included (only the data relating to people with MND were included), but not those that reported solely on the carer's experience.

Search and selection process

Articles were identified through electronic searches that were carried out between May 2011 and May, 2012. MEDLINE, CINAHL Plus and PsychInfo databases were

Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.
<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>

used for the literature search, with the keywords 'ALS' or 'amyotrophic lateral sclerosis' or 'motor neuron* disease' or 'MND' AND 'experience*' OR 'qualitative'.

All search fields (e.g. title, keywords, abstract etc.) were selected in all three databases. The basic search function was used in Medline and PsychInfo, with the 'related terms' search function activated. The advanced search function was used in CINAHL Plus, utilising the Boolean/Phrase search mode. The search and selection process is presented in figure 2.

The bibliographical details of the articles that were identified through the searches were inputted into a word document. The details included the title, the keywords, and the abstract, where available. Appropriateness for inclusion was judged by these elements. Common reasons for exclusion were: a focus on effectiveness of interventions; a focus on MND symptoms; a focus exclusively on carers' perspectives; and not being research based. Barroso et al (12) discuss the difficulties relating to identifying relevant qualitative research articles for review studies, identifying poor consistency of using the term qualitative research and inappropriate indexing as areas of concern. These issues are also recognised by the CRD and may explain the high exclusion rate observed in this review (87.9%).

Following the review of title, keywords and abstract, 27 articles were retrieved and reviewed. Consequently eleven were rejected (reasons are presented in figure 2). The reference lists of the sixteen accepted articles were examined for further appropriate studies and consequently four further articles were identified, leading to twenty studies being accepted for inclusion in the review. Inconsistent use of terminology and indexing of qualitative research may explain why these articles were not identified through the initial literature search (12).

Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.
<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>

Critical appraisal and data extraction

Articles that fulfilled all inclusion criteria were critically appraised in order to assess their quality. It was decided to assess quality in order to avoid drawing unfounded conclusions, as suggested by Thomas and Harden (13). Following the CRD (11) guidelines, quality assessment was used in order to identify the weaknesses and strengths of the articles and take these into account during the literature synthesis. No articles were rejected as a result of this process.

Quality assessment was based on the criteria developed by Long and Godfrey (14), as adapted by Riley and Boniface (15). The criteria were further adapted for the purposes of this review with the aim to assess relevance of the articles to the purposes of the review, and appraise their quality. Some questions were adapted for relevance to the context of this review (MND), while some others were not included (two questions pertaining to study setting and one question pertaining to outcomes).

Following appraisal, data were extracted in accordance with Thomas and Harden's (13) methods for developing a thematic synthesis. For the purposes of this review data were defined as the results of the studies examined. Following Thomas and Harden (13) these data were the analyses and interpretations of the researchers rather than participants' quotes, which were considered to be raw data. Segments of text under the headings of 'results' and 'findings' were treated as data and pasted onto a Microsoft Word document. The discussion section of all articles was also reviewed as sometimes it included results. All included data were entered into the qualitative analysis software NVIVO.

Data analysis

Analysis was performed using NVIVO. The first step was to read through all extracted data and code them. In total 101 unique codes were developed. Each of the codes addressed a specific issue and they were all inputted as hierarchical nodes in NVIVO with new codes being added as data were analysed. These codes were then collapsed into descriptive themes. These descriptive themes remained very close to the data and offered an overview of the findings of the reviewed studies (13). Figure 3 offers an example of the process of moving from codes to a descriptive theme.

While some of the articles reviewed explicitly addressed this question, several did not. The next stage was to synthesise the descriptive themes into analytical themes that would offer an answer the review question. Analytical themes do not merely describe but go 'beyond the findings of the primary studies and generate additional concepts, understandings or hypotheses' (13 p.7). These themes are often abstract and rather than offering a detailed or thorough description of data they provide an answer to specific review questions (13).

Results

Twenty studies met all inclusion criteria and these are presented in table 1. The included studies reported on the experience of 257 people with MND. Age range of people with MND was between 25-85 years and time since diagnosis ranged from 1 month to 13 years. The results of the review were synthesised in two broad analytical themes and several descriptive themes. These are presented in table 2. The two analytical themes are presented below.

Knowing how to live with MND; developing experiential knowledge

A diagnosis of MND, and living with the condition represents a major change in people's lives. One important element of people's experience of living with MND is that they develop knowledge that is specific to their situation. In a way, they need to learn how to live with MND as this represents a major change compared to their life prior to MND. This process of learning how to live with MND can be described as a process of developing experiential knowledge. Experiential knowledge is not evidence based but experience based, and gives people insights on what works well and what doesn't. As Pols (16 p.200), stated experiential knowledge refers to "knowledge about how to live with a chronic disease on a daily basis".

People with MND need to navigate an uncertain future; their symptoms keep progressing, new ones emerge and there is no definitive information about prognosis. Faced with too much, too little, too technical or ill-timed information people with MND often concentrate on the here and now and how they can live with MND on a day to day basis (17-22). They prefer to deal with issues as they arise since information given too far in advance tends to reveal a frightening future.

People with MND report that despite professionals' best efforts sometimes services do not meet their needs effectively, and this is mainly attributed to inadequate knowledge of MND and to the organisation of services (7). Professionals' knowledge about MND and the organisation and coordination of services have been reported as less than satisfactory (8). The early and the late stages of the disease process in

Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.
<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>

particular, are especially critical stages as they represent important changes in the life of the patients and the people around them. These stages are reflected in healthcare by diagnosis and palliative care and satisfaction levels with provided services are not always high (20, 23). On the other hand specialist multidisciplinary clinics are generally seen in a positive way, perhaps due to the specialised knowledge professionals have. However, attending these clinics can be tiring and sometimes intimidating due to the involvement of many professionals at the same time (24).

Through these experiences, people with MND develop their own understanding of how MND affects them and what services they require (25). In her exploration of experiences of care in MND, Brown (26) concluded that people with MND and service professionals operate from different standpoints, based on the value structure they adopt as being valid. The scientific standpoint adopted by professionals leads them to a focus on functional assessments, adaptations, equipment or other changes that will render a functional outcome. People living with MND however focus on what it means to live with MND; what kind of life it is, and how they can live a good life *with* it. They come from a hermeneutic standpoint that values lived experience and how people make sense of it. This standpoint is reflected in the second analytical theme.

Not having control over the disease but trying to have control over one's life

MND can represent an existential shock (26). Questions about how one should live one's life, what choices to take and how to set priorities are part of the process of

Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.
<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>

adapting to living with MND (27). People talk about and make sense of their life with MND in different ways (28, 29). Sometimes, and for some people, life appears as being over (29). Living life to the full and enjoying what is available, or modifying one's priorities, can also be part of some people's life, as is an overwhelming sense of difficulties to come, which disrupt plans, priorities and wishes. These difficulties can fracture life (28). In Brott, Hocking and Paddy's (30) study the main disruptions participants were experiencing were related to their activities and the social roles these were associated with; as their body became increasingly paralysed people found it hard to engage in meaningful activities and maintain valued social roles.

People come to accept that they have limited or no control over the disease but they try to have control over their life (18, 31). The image that people had of themselves, the idea of who they are and who they want to be is changed as a result of MND.

People living with MND modify their notion of a *desired self*, i.e. how they want to live their life and who they want to be. They engage in a continuous process of adapting to ongoing change. As King, Duke and O'Connor's (31) study highlighted, people living with MND take decisions that will enable them to keep on living and maintain a sense of self and well-being in the face of constant change and loss of physical abilities.

People with MND are faced with a body they cannot control, one they cannot rely on; a body they cannot trust (28, 29). Their abilities change all the time and things that were possible one day may not be possible the next. With changing abilities comes a moving threshold from acceptable to not acceptable levels of dependence; from loss of independence with toileting, to use of percutaneous endoscopic gastrostomy (PEG), use of non invasive positive pressure ventilation (NIPPV), loss of speech, to total loss of communication. People with MND are afraid of losing those functions

Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.
<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>

associated with dignity and communication as these are deeply connected to their identity (18, 22).

While the body cannot be trusted, or exactly because of that, people with MND try to take control over their life and they do that in many ways. From adapting activities to reflect current levels of function, to changing their diet and from becoming experts on MND, to making the best of what they have, people with MND try to effect control over their lives. This is done in a pragmatic way as people develop an awareness of what they can and what they cannot do and focussing on what is possible (28, 30, 32). Limited mobility for example can affect expression of intimacy, but modified ways can be found (33).

In seeking to take control over their life, many people use various pieces of equipment. These can include, but are not limited to, NIPPV, PEG, and alternative and augmentative communication devices (AAC). Equipment comes with certain benefits and certain drawbacks and people with MND will accept the equipment when the perceived benefits outweigh the inconvenience from the incurred changes on daily routines (21, 31). Sometimes equipment can be perceived to be too technical and complex (19, 34). People balance the advantages and disadvantages in relation to their life. The use of NIPPV for example is not always seen favourably, because it can upset other people, or because of too overt associations with disease and dependence. Also, NIPPV masks (depending on length of use and type of mask) can cause sores on the nose and around the mouth. On the other hand, NIPPV offers the benefit of restoring sleep and days with more energy (21).

Early reliance on equipment can be perceived as contributing to functional deterioration, or as a definitive sign that the threshold from independence to

Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.
<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>

dependence has been traversed (34). The decision to use equipment appears to be guided by both the perceived benefits of such equipment and the extent to which use of the equipment contributes to the person's own view of their life. It would seem that people with MND make decisions that will enable them to retain their sense of identity while at the same time addressing some of the effects of the condition on their daily life.

Discussion

This review highlights the decision making and knowledge generating processes that people with MND use in learning to live with MND. How they decide whether a PEG, NIPPV or AAC is good or bad, or how they perceive and engage with services depends upon how they view MND and their idea of a good life with the condition. This has two main implications for clinical practice.

The first implication concerns the knowledge that people with MND possess. Pols (16) discusses the importance of practical knowledge, which she refers to as the *know-now*. This knowledge is about how people make decisions on a daily basis, and as they constantly adapt their behaviour to meet the changing demands of an ongoing condition. It is about knowing what is needed to be done *now* that one's hand cannot hold the toothbrush anymore, or that the TV remote control gets too heavy to handle. This review highlights that people with MND develop knowledge through living with the disease and develop their own ways of living with it, which can range from learning how to avoid or treat sores from the NIPPV mask, to experimenting with ways to express intimacy. Tapping into the practical knowledge that enables people to

Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.
<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>

keep on with their daily lives can offer insights about the kinds of care that is most useful.

The second implication concerns the decision making processes of people with MND regarding their care and more specifically, the rationale behind what care (interventions, equipment or medications) they prefer. According to Campbell, Roland and Buetow (35) care requires an understanding of the various environments a person inhabits; medical and social. In this sense care refers to a constant negotiation in decision-making between what is good for one world and good for the other (36); what is good in a medical context is not necessarily good in a social context, e.g. a gastrostomy comes with a changed body image but also with higher calorific intake. As Good (37) and Mol (36) amongst others have illustrated, service providers and service users enter into the care encounter with different perspectives, and sometimes focusing on different realities. Hunt and Mattingly (38 p.267) referred to this multitude of perspectives as 'diverse rationalities' and 'multiple realities'. In other words, people living with a disease and healthcare professionals may use different systems of knowledge to help them make sense of what a particular disease is about, and they may reach different explanations for it.

Letiche (39) referred to care as being *in-between* or *being-two* since it cannot be defined by a single process but it is always situation-bound and depends on who delivers it and who receives it; in effect the meaning of healthcare is co-constructed by the people who are involved in it. Several strategies for managing some of the impairments of MND have been developed. However not everybody chooses to use those (40, 41). Perspectives on invasive ventilation for example differ greatly,

Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.

<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>

between and within countries (42, 43). Personal experiences of living with MND need to be listened to and respected before shared goals can be constructed.

Strengths and weaknesses of the reviewed studies

The available literature offers valuable insights into the experience of living with MND. The studies reviewed contain rich exploration of living with MND and all of them brought the perspectives of people with MND to the foreground.

The results of the studies reviewed offered similar information, to some extent. This is probably a fabricated data saturation that can be attributed to the research design that most of the studies followed, i.e. studies where participants were interviewed once. Designs based on single interviews cannot always capture the processes of ongoing change that are present in living with MND as these offer a snapshot of experience.

Furthermore, several of the studies did not make their methodology explicit. This presents the following issues that may affect the quality of the data presented:

1. The rigour of the research design could not be judged.
2. Limited transparency of research methods followed meant that several of the studies were not replicable.
3. The standpoint of the researcher was not always clear neither was the theoretical framework that guided the analysis.

In several of the studies reviewed sampling strategies were not adequately presented.

While size sample in qualitative research is guided by saturation rather than by numbers, several studies did not specify how saturation was reached. Furthermore issues of selection, response and interviewer bias were often left unaddressed.

Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.
<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>

The weaknesses observed can be addressed in future research by ensuring the methods followed are appropriate for exploring the ongoing experience of living with MND (e.g. multiple interview studies/ ethnographic studies) and are transparent. Sampling strategies and size also need to be explained in detail, and the researcher's position needs to be made explicit.

Limitations of research design

The limitations of this review pertain to the inclusion criteria, the search process, the analysis and the positioning of the reviewers. Although the review identified only 20 studies that met the inclusion criteria, this does not necessarily signify a dearth of relevant research or publications. This review only considered research articles published in peer reviewed scientific journals accessible through academic databases. There was therefore an inherent publication bias, excluding three other potential sources of evidence. The first is grey literature, including reports, and personal accounts published in the popular press and the media. The second is reflections and general discussions published in academic journals. The third is research published in books. While this review concentrated on published research based literature, inclusion of the sources above in future reviews can further the understanding of the personal experience of living with MND. Furthermore, this review only considered articles published in the English language and it was beyond the scope of the review to access material written in other languages.

All effort was taken so that the search strategy was as comprehensive as possible. For example, the reviewers searched for keywords in all fields (keywords, abstracts etc.), and used various related terms to search for MND. However inappropriate or

Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.
<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>

inconsistent use of terminology and indexing of qualitative research (11, 12) may have resulted in omission of relevant studies.

The samples of the studies selected in the review included people with different types of MND. Differences in experiences based on MND type could be explored in future studies. Furthermore, the studies reviewed were based in different cultural contexts.

While we sought to be cognisant of the context, this was not always possible as the context was often not adequately described by the authors. This may have resulted in decontextualisation of some of the data extracted from the studies. During the analysis, we followed Thomas and Harden's (13) processes for developing descriptive and analytical themes. As the latter are abstract themes, aiming to offer one answer the review question, it is acknowledged that some elements of the experience of living with MND may have not been included. However, the aim of this review was to synthesise the existing literature, going beyond merely describing it. Finally, the three reviewers are all based in the UK, and all have a background in healthcare practice. While results were critically discussed in the review team, it is possible that the analysis was influenced by the particular standpoints of the reviewers. Including people with MND in the review team could have addressed this issue. However this was not possible because of access and resource implications.

Conclusions

This review highlights the decision making and knowledge generating processes that people with MND use in learning to live with MND. Further research is required to explore these processes and their implications for the care of people with MND.

The way people make sense of their life with MND influences their needs, their choice of services and the way they negotiate their illness. Individuals, their families

Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.
<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>

and friends and healthcare professionals have their own ideas of how life is to be lived, what needs to be done, how and why. They create what Kleinman (44) called the different personal and interpersonal meanings of the disease and of life with it. These different perspectives are in constant negotiation in the context of daily life with everybody involved in a dynamic dialogue, trying to establish a common ground for understanding of what is *good* and what has to be avoided (39).

Currently there is limited knowledge on how people with MND experience life with the disease (7, 8, 26, 45). It is important that the experience of living with MND is explored so that health professionals, people with MND and their carers understand each other's standpoint when they talk about decisions that are good, about care, or about improving quality of life.

Declaration of interest

The authors report no conflicts of interest.

Acknowledgments

The authors are grateful to the two anonymous reviewers for their helpful comments and to Sarah Kantartzis and Margaret McGrath for offering feedback on earlier drafts of this article.

Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.
<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>

References

1. Eisen A. Amyotrophic lateral sclerosis: a 40 year personal perspective. *Journal of Clinical Neuroscience*. 2009;16(4):505-512.
2. Corcia P, Meininger V. Management of amyotrophic lateral sclerosis. *Drugs*. 2008;68(8):1037-1048.
3. Lou J. Fatigue in amyotrophic lateral sclerosis. *Physical Medicine and Rehabilitation Clinics of North America*. 2008;19:533-543.
4. Portet F, Cadilhac C, Touchon J, Camu W. Cognitive impairment in motor neuron disease with bulbar onset. *ALS and other motor neuron disorders*. 2001;2:23-29.
5. Tomik B, Guilloff R. Dysarthria in amyotrophic lateral sclerosis: A review. *Amyotrophic Lateral Sclerosis*.2010;11:4-15.
6. Talbot K, Marsden R. *Motor neuron disease; the facts*. Oxford: Oxford University Press; 1998.
7. Brown J, Lattimer V, Tudball T. an investigation of patients and providers' views of services for motor neurone disease. *British Journal of Neuroscience Nursing*. 2005;1(5):3.
8. Hughes RA, Sinha A, Higginson I, Down K, Leigh PN. Living with motor neurone disease: Lives, experiences of services and suggestions for change. *Health and Social Care in the Community*. 2005;13(1):64-74.
9. Van Teijlingen E, Friend E, Kamal A. Service use and needs of people with motor neurone disease and their carers in Scotland. *Health and Social Care in the Community*. 2001;9(6):397-403.

- Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.
<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>
10. Ng L, Khan F, Mathers S. Multidisciplinary care for adults with amyotrophic lateral sclerosis or motor neuron disease. *Cochrane Database of Systematic Reviews*. 2009(4):CD007425.
 11. Centre for Reviews and Dissemination. *Systematic reviews; CRD's guidance for undertaking reviews in health care*. York: Centre for Reviews and Dissemination, University of York; 2009.
 12. Barroso J, Gollop CJ, Sandelowski M, Meynell J, Pearce PF, Collins LJ. The challenges of searching for and retrieving qualitative studies. *Western Journal of Nursing Research*. 2003;25(2):153-78.
 13. Thomas J, Harden A. Methods for the thematic synthesis of qualitative research in systematic reviews. *BMC Medical Research Methodology*. 2008;8:45-.
 14. Long AF, Godfrey M. An evaluation tool to assess the quality of qualitative research studies. *International Journal of research Methodology*. 2004;7(2):15.
 15. Riley J, Boniface G. The effectiveness of occupational therapy local authority social services' interventions for older people in Great Britain; A critical literature review. Cardiff: Cardiff University, 2012.
 16. Pols J. Breathtaking practicalities: a politics of embodied patient positions. *Scandinavian Journal of Disability Research*; 2011;13(3):189-206.
 17. Cobb AK, Hamera E. Illness experience in a chronic disease - ALS. *Social Science and Medicine*. 1986;23(7):641-50.
 18. Lemoignan J, Ells C. Amyotrophic lateral sclerosis and assisted ventilation: How patients decide. *Palliative and Supportive Care*. 2010;8(2):207-13.
 19. McNaughton D, Light J, Groszyk L. "Don't give up": Employment experiences of individuals with amyotrophic lateral sclerosis who use augmentative and alternative

- Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.
<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>
- communication. *AAC: Augmentative and Alternative Communication*.
2001;17(3):179-94.
20. O'Brien MR, Whitehead B, Jack BA, Mitchell JD. From symptom onset to a diagnosis of amyotrophic lateral sclerosis/motor neuron disease (ALS/MND): Experiences of people with ALS/MND and family carers - A qualitative study. *Amyotrophic Lateral Sclerosis*. 2011;12(2):97-104.
21. Sundling IM, Ekman SL, Weinberg J, Klefbeck B. Patients' with ALS and caregivers' experiences of non-invasive home ventilation. *Advances in Physiotherapy*. 2009;11(3):114-20.
22. Vesey S, Leslie P, Exley C. A Pilot Study Exploring the Factors that Influence the Decision to have PEG Feeding in Patients with Progressive Conditions. *Dysphagia*. 2008;23(3):310-6.
23. Whitehead B, O'Brien M, Jack B, Mitchell D. Experiences of dying, death and bereavement in motor neurone disease: A qualitative study. *Journal of Palliative Medicine*. 2012;26(4):368-378.
24. O'Brien M, Whitehead B, Jack B, Mitchell JD. Multidisciplinary team working in motor neurone disease: patient and family carer views. *British Journal of Neuroscience Nursing*. 2011;7(4):580-5.
25. Cox DL. Perspectives of motor neurone disease. *Clinical Rehabilitation*. 1992;6:333-9.
26. Brown J. User, carer and professional experiences of care in motor neurone disease. *Primary Health Care Research and Development*. 2003;4:207-217.
27. Bolmsjö I. Existential issues in palliative care: Interviews of patients with amyotrophic lateral sclerosis. *Journal of Palliative Medicine*. 2001;4(4):499-505.

- Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.
<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>
28. Brown J, Addington-Hall J. How people with motor neurone disease talk about living with their illness: A narrative study. *Journal of Advanced Nursing*. 2008;62(2):200-208.
29. Locock L, Ziebland S, Dumelow C. Biographical disruption, abruption and repair in the context of motor neurone disease. *Sociology of Health and Illness*. 2009;31(7):1043-58.
30. Brott T, Hocking C, Paddy A. Occupational disruption: Living with motor neurone disease. *British Journal of Occupational Therapy*. 2007;70(1):24-31.
31. King SJ, Duke MM, O'Connor BA. Living with amyotrophic lateral sclerosis/motor neurone disease (ALS/MND): Decision-making about 'ongoing change and adaptation'. *Journal of Clinical Nursing*. 2009;18(5):745-54.
32. Foley G, O'Mahony P, Hardiman O. Perceptions of quality of life in people with ALS: Effects of coping and health care. *Amyotrophic Lateral Sclerosis*. 2007;8(3):164-9.
33. Taylor B. The impact of assistive equipment on intimacy and sexual expression. *British Journal of Occupational Therapy*. 2011;74(9):435-42.
34. Murphy J. "I prefer contact this close": perceptions of AAC by people with motor neurone disease and their communication partners. *AAC: Augmentative & Alternative Communication*. 2004;20(4):259-71.
35. Campbell SM, Roland MO, Buetow SA. Defining quality of care. *Social Science and Medicine*. 2000;51(11):1611-25.
36. Mol A. *The body multiple; ontology in medical practice*. Durham: Duke University Press; 2002.
37. Good, B. *Medicine, rationality and experience; an anthropological perspective*. Cambridge, MA: Harvard University Press; 1994.

- Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.
<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>
38. Hunt LM, Mattingly C. Diverse rationalities and multiple realities in illness and healing. *Medical Anthropology Quarterly*. 1998; 3:267-72.
39. Letiche H. Making healthcare care. Charlotte, NC: Information Age Pub; 2008.
40. Jackson CE, Lovitt S, Gowda N, Anderson F, Miller RG. Factors correlated with NPPV use in ALS. *Amyotrophic Lateral Sclerosis*. 2006;7(2):80-5.
41. Katzberg HD, Benatar M. Enteral tube feeding for amyotrophic lateral sclerosis/motor neuron disease. *Cochrane Database of Systematic Reviews*. 2011(1):CD004030.
42. Borasio GD, Gelinas DF, Yanagisawa N. Mechanical ventilation in amyotrophic lateral sclerosis: a cross-cultural perspective. *Journal of Neurology*. 1998;245 Suppl 2:S7-12; discussion S29.
43. Neudert C, Oliver D, Wasner M, Borasio GD. The course of the terminal phase in patients with amyotrophic lateral sclerosis. *Journal of Neurology*. 2001;248(7):612-6.
44. Kleinman A. *The illness narratives: Basic Books*; 1988.
45. O'Brien MR. Healthcare professionals' knowledge of motor neurone disease. *British Journal of Nursing*. 2004;13(18):1080-4.
46. Bolmsjö I, Hermerén G. Interviews with patients, family, and caregivers in amyotrophic lateral sclerosis: Comparing needs. *Journal of Palliative Care*. 2001;17(4):236-40.

Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.
<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>

Legends

Table 1

Reviewed articles, in order of publication date

Table 2

Overview of descriptive and analytical themes

Figure 1

Review process

Figure 2

Literature search and selection process

Figure 3

Example of data analysis process, from codes to one descriptive theme

Table 1

Authors/ Year	Location	N	Aim	Design	Findings/ Recommendations
Cobb and Hamera, 1986 (17)	USA	2	To explore the effect of the illness on participants’ relationships with family, friends and the healthcare system	Representative case study approach	Social relationships undergo radical change. Dissatisfaction with professional services. Importance of dialogue between lay and professional perspectives on MND is highlighted
Cox, 1992 (25)	UK	10 people with MND, 10 carers and 8	To explore the everyday needs of people with	Semi structured interviews. Exact design	Importance of physical needs and practical solutions

		occupational therapists	MND	unclear.	
Bolmsjö, 2001 (27)	Sweden	7 people with MND	To investigate existential issues in palliative care	Semi structured interviews. Exact design unclear.	Importance of the need to be respected and of the relevance of existential issues was highlighted
McNaughton, Light and Groszyk, 2001 (19)	USA	7 people living with MND (data from 5 were analysed)	To explore employment experiences of people who use AAC	Qualitative design, with use of a focus group (internet based discussion group)	Participants described several barriers to employment. The importance of identifying appropriate AAC was highlighted
Bolmsjö and Hermerén, 2001 (46)	Sweden	8 people living with MND and 8 informal	To compare needs as expressed by people living with MND and their informal	Qualitative design, with use of interviews	The two groups perceived needs in different ways and have different responses to MND

		carers	carers		
Brown, 2003 (26)	UK	6 people living with MND, 6 family carers, and 9 professionals	To explore professional and lay values of care in MND	Hermeneutic phenomenology	The three groups perceived care in different ways and focussed on different aspects of it. Listening to people's voice is important in order to construct the care they need
Murphy, 2004 (34)	UK	15 people living with MND and 13 communication partners (spouses, friends or relatives)	To explore perceptions of using AAC	Qualitative design, with use of video recordings, narratives, and field notes	Use of AAC was not as beneficial as anticipated. The main reasons for this were the complexity of equipment and the social distance that it creates (i.e. inflection is not possible)

Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.

<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>

Hughes et al, 2005 (8)	UK	9 people living with MND, 5 family carers and 15 professionals	To explore participants' experiences of living with MND, their experiences of services and suggestions for change	Qualitative design with use of semi structured interviews	Professionals' understanding of MND needs to be improved. People living with MND need more information regarding the process of the disease but also about therapies, management strategies and equipment
------------------------	----	--	---	---	---

Brott, Hocking and Paddy, 2007 (30)	New Zealand	7 people living with MND	To elucidate the experience of engaging in day to day activities from the experience of people who live with MND	In depth interviews, guided by a phenomenological design	Participants experienced MND in terms of changes in participation in daily life. As the condition progressed, changes in their levels of engagement in activities resulted to the loss of valued social roles.
Foley, O'Mahony and Hardiman, 2007 (32)	Ireland	5 people living with MND	To explore meaning of quality of life and explore the influence of healthcare on perceived wellbeing	In depth interviews, guided by a phenomenological design	Findings highlight the importance of faith, search for control, dignity, desire to maintain identity and family. Recommendation to consider how people with MND

					adapt to change as the disease progresses
Brown and Addington-Hall, 2008 (28)	UK	13 people living with MND	To explore participants' experiences of living with MND and how they talk about living and coping with the disease	Longitudinal narrative case studies	People experience MND in different ways, sharing storylines described as sustaining, preserving, enduring and fracturing. Stories help individuals, their families and healthcare professionals understand what it is like to live with MND
Vesey, Leslie and Exley,	USA	7 people living with	To explore the decision making process	Qualitative design, with use of semi structured	Participants felt that they had no control over the decision, as this

2008 (22)		MND	regarding PEG	interviews	was dictated by the medical condition. It is suggested that clear information about the process can increase the perceived control and involvement of patients in the process.
Locock et al, 2009 (29)	UK	35 people living with MND	To explore how people construct their accounts of living with MND and what they say about their life	Narrative interviews	People experience MND in different ways; as a death sentence (biographical abruption), as a major disruption to their life (disruption) or as a challenge that they try to make sense of (repair).

King et al, 2009 (31)	Australia	25 people living with MND	To develop a model explicating the dimensions of living with MND Grounded theory based on symbolic interactionism	Grounded theory based on symbolic interactionism	People living with MND make decision as their circumstances change- they need to adapt to an evolving situation and to diminishing physical abilities.
Sundling et al, 2009 (21)	Sweden	7 people living with MND and 8 family carers	To explore the experience of non invasive ventilation	Qualitative design, semi structured interviews	The use of ventilation was perceived favourably, after an initial period of adjustment, as it had a positive effect on daily life.

Lemoignan and Ellis, 2010 (18)	Canada	9 people living with MND	To explore the decision making process regarding initiation of assisted ventilation	In depth interviews, guided by a phenomenological design	The decision making process is influenced by many factors, and participants valued autonomy in the decision making process.
O'Brien et al, 2011 (20)	UK	24 people with MND and 28 (10 of them former) carers.	To explore perspectives on the process of diagnosis	Narrative interviews	Participants reported diagnostic delays and failure by healthcare professionals to recognise early symptoms. Delivery of diagnosis and support immediately afterwards was often unsatisfactory.
O'Brien et al,	UK	24 people with	To explore experiences	Narrative interviews	Overall the experiences were

2011 (24)		MND and 28 (10 of them former) carers.	of multidisciplinary care		positive with issues such as ease of access to information and specialised care being valued by the participants.
Taylor, 2011 (33)	UK	13 people with MND and 10 partners	To explore the meaning of sexuality for people with MND	In depth interviews guided by a phenomenological design	Sexuality is an important, yet overlooked aspect of people's lives. None of the informants had been given the opportunity to discuss with an occupational therapist the use of assistive equipment to enable expression of sexuality.
Whitehead et al,	UK	24 people with	To explore the	Narrative interviews	Needs during the final stage of the

Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.

<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>

2011 (23)		MND and 18 carers	experiences of people with MND and their carers during the final stages of the disease process (and during bereavement period)		disease process are not adequately met. Issues that are highlighted include: care burden, and its impact on carers and on people with MND and limited used of advance care planning tools.
--------------	--	-------------------------	---	--	---

Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.

<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>

Table 2

Analytical themes	Descriptive themes
Knowing how to live with MND; developing experiential knowledge	Navigating an uncertain future Experiences of services Managing information

Sakellariou, D., Boniface, G. and Brown, P. 2013. Experiences of living with motor neurone disease; A review of qualitative research. *Disability and Rehabilitation* 35(21), pp. 1765-1773.

<http://informahealthcare.com/doi/abs/10.3109/09638288.2012.753118>

<p>Not having control over the disease but trying to have control over one's life</p>	<p>Cannot trust the body</p> <p>Taking control</p> <p>Adaptive equipment is both good and bad</p>
---	---