A qualitative meta-synthesis of evidence (meta-ethnography) exploring the personal experiences of gastrostomy tube in neurodegenerative diseases: a case of motor neurone disease

Dominika Lisiecka, Aine Kearns, Fiona Bourke, Ian Lawson and Cath Muir

ABSTRACT

Purpose: The lived experiences of a gastrostomy tube (GT) in adults with neurodegenerative diseases (NDDs) are not well understood. The aim of this qualitative meta-synthesis was to review and synthesise the available evidence to inform clinical practice and identify research gaps.

Methods: Meta-ethnographic synthesis of qualitative studies was conducted with systematic searching of eight databases from inception to March 2021. Qualitative studies reporting personal experiences of GTs in adults with NDDs were identified. New theories were developed during translation of concepts from each study and combined as a “line-of-argument” synthesis. Patient and public involvement was incorporated as two of the authors are living with an NDD and a GT.

Results and conclusions: Of 2863 unique records identified, only nine fulfilled the review criteria. All studies recruited participants with motor neurone disease (MND); no other NDDs were represented. Two main themes emerged: decision making and living with GT. Decision making was the predominant theme and data regarding living with GT were sparse. There is limited research on the lived experience of a GT in adults with NDDs. The lived experience of GT in MND is complex and individualised. Future research is indicated to inform clinical practice.

IMPLICATIONS FOR REHABILITATION

- Evidence related to the lived experiences of gastrostomy tube (GT) in neurodegenerative diseases (NDDs) is lacking especially in relation to the time after GT insertion.
- Decision making in relation to GT is a complex and individualised psychological process for some people, while others perceive no decisional conflict.
- Support from healthcare professionals is crucial during the decision-making time and should not cease after GT insertion.
- Support from healthcare professionals can help resolve any clinical complications and also incorporate GT into everyday routines.
- Healthcare professionals should be aware that their views on the benefits and problems related to GT may differ to those of individuals with a NDD.

Introduc...
is associated with shorter survival than spinal onset [4]. In bulbar onset, swallowing difficulties (dysphagia) are present early in the disease [11,12,14]. As MND progresses, dysphagia affects the majority of people irrespective of the clinical phenotype and the site of onset [15–17]. Over the last few years, major progress has been made in regard to understanding of MND, resulting in improved quality of life for people living with this disease [12], but the cure for MND is still to be found [11]. Riluzole was the only disease-modifying medication from 1995 [18] to 2017 when edaravone was approved in the USA [19]. Both treatments are only temporarily effective and have restricted acceptance [11,20,21].

Non-oral feeding may be appropriate for some people with NDD [22–24]. A GT is a narrow plastic tube inserted into the stomach, most commonly using radiological or endoscopic placement method, to provide hydration, nutrition, or medication directly to the stomach [25]. GT is often recommended due to swallowing problems (dysphagia) [23,26]. Complications related to insertion of a GT are common in NDD especially within the first month; however, they do not appear to have a negative impact on survival [24]. The clinical evidence suggests multiple benefits of non-oral feeding, such as reduced fear of choking, but the research evidence regarding the efficacy and the perceived personal impact of non-oral feeding in neurological diseases is limited [23]. The suitability for non-oral feeding will be influenced by the type of NDD. For example, this intervention is generally discouraged in dementia [23] except for milder cases and for a limited time [27] but is often recommended in MND [28].

It is well recognised that nutrition is an important prognostic factor in MND and weight loss is associated with decreased survival [29–32]. Taking adequate nutrition and hydration orally can be challenging in MND due to dysphagia, arm weakness, respiratory problems, depression, or low mood [14]. Respiratory status is taken into consideration when deciding about GT insertion, as the forced vital capacity should be >50% for the procedure to be safe [33]. Many factors can influence personal decision making to accept or decline a GT in MND [34] and this decision is reported as complex [35–37]. Some people with MND find the discussion regarding GT difficult and they delay their decision; sometimes this delay is lost and the insertion procedure may no longer be safe [14]. Patient-reported benefits of GT include weight stabilisation, reduced anxiety when eating and a perceived survival benefit [38]. Studies of clinical practice however, report mixed evidence regarding the effectiveness of GT on survival in MND [39]. To date, no review of evidence has been undertaken examining the personal experiences of GT for people living with a NDD, such as MND. This knowledge is imperative to identify the positive and negative experiences related to GT and to ensure provision of comprehensive support for people living with NDD.

The aim of this study was to synthesise the current body of knowledge regarding the personal experience of a GT in a population of people with NDD and develop recommendations for research and clinical practice. As there has been no previous evidence synthesis on this topic, a search strategy was developed to systematically identify all relevant studies during the initial research stage. The search included selected types of NDD (MND, dementias, Huntington’s disease, multiple system atrophy, progressive supranuclear palsy, multiple sclerosis, and Parkinson’s disease). The research question was: what are the personal experiences of GT in a population of people with NDD? However, despite searching the evidence for a variety of NDD, only studies including people with MND were identified; therefore, this article will present the lived experiences of a GT in MND.

**Methods**

It is acknowledged that systematic synthesis of qualitative studies may generate more holistic knowledge and improve the depth of existing theories [40]. Meta-ethnography is considered the most frequently used qualitative evidence synthesis methodology particularly within the healthcare professions [41–43]. We chose this approach as the most suitable to fulfil our aim. Meta-ethnography is not simply the sum of parts of single studies [43]. During meta-ethnography conceptual data from primary studies are systematically extracted and compared to create new concepts, explanations, interpretations, and theories while preserving the original meanings of included studies [43,44]. Therefore, a philosophical underpinning of meta-ethnography is in line with subtle realism, which acknowledges that all research involves subjective perceptions and absolute certainty of findings cannot ever be claimed [45]. Subtle realism accepts the possibility of multiple valid interpretations of a phenomenon under study [43]. As our interest was to investigate personal experiences, we rejected quantitative approaches and chose meta-ethnography as the best framework to identify existing and develop new concepts pertaining to the experiences of GT in NDD [43,46].

This qualitative evidence synthesis was conducted in line with the seven phases of meta-ethnography [44] which allow for rigorous synthesis of qualitative data [42]. To ensure the highest reporting standards, this article presents the process of meta-ethnography by fulfilling recently published eMERGe meta-ethnography reporting guidance [46]. All 19 criteria (Table 1) are presented throughout the manuscript to provide in depth evidence of how eMERGe guidelines were followed. Phase 1 with all corresponding criteria (1–4, as per Table 1) has been fully addressed in “Introduction” section. This review was registered on PROSPERO (CRD42020180700). In order to allow for multi-perspective interpretation of findings, patient and public involvement (PPI) was incorporated during the evidence synthesis and discussion (Table 1: criteria 15–19), as two authors have personal experience of GT and MND (IL and CM). Although PPI involvement is considered to be an important part of empirical health research and is recognised by Cochrane Consumer Network as advantageous in order to produce relevant and accessible reviews, it is still reported as infrequent within published meta-ethnographic studies [47].

**Searches**

A search strategy was developed by DL and FB in consultation with AK. Final searches were completed in March 2021 by DL and AK. The following electronic databases were searched: APA PsycArticles, APA Psychinfo, Academic Search Complete, Cinahl, Medline, Embase, Pubmed, and Cochrane Boolean Operator. Keywords were identified from the research question (Table 2) and combined using the “OR” and “AND” Boolean terms. Truncation was used to incorporate similar words, for example, the search term “Parkinson” encompassed “Parkinson’s,” “Parkinsonian,” etc. Further synonyms or alternative terms were combined to ensure systematic searches of the databases, following a consultation with the university librarian. In anticipation of finding a small number of articles there was no time limit applied during the search.
In addition to the database searches, Google Scholar and the university library were hand searched to identify any further relevant articles. Articles were scanned by title and abstract to establish their pertinence to the research question. For the first stage of screening, duplicates and clearly irrelevant studies (e.g., studies not related to NDD) were removed by one reviewer (DL). In the next stage, three independent reviewers screened each study abstract (DL, FB, AK). Any conflicts regarding the eligibility of a study were resolved through discussion and the final decision regarding inclusion was made through consensus. Papers were then read thoroughly by all authors and were included if they reported the lived experience of non-oral feeding from the perspective of a person with a neurodegenerative disease and utilised qualitative methods within research designs that allowed in-depth exploration of personal experience. Peer-reviewed studies reporting on decision making in relation to a GT as well as studies examining lived experiences post GT insertion were considered eligible for inclusion. Studies which were limited to quantitative results were excluded, but mixed methods studies were eligible if the qualitative data were presented in sufficient detail. Studies recruiting both people with NDD and their caregivers, or where other conditions (outside NDD) were also investigated, were excluded if it was not possible to extract the data from participants with NDD (where findings were reported jointly). Taking into account the limited number of articles meeting the criteria, experiences recorded in alternative publications such as a letter to an editor should also be considered for inclusion as all patient experience will greatly educate the research question and future practice [48]. Therefore, a letter to the editor was also included in this review. This letter contains a comprehensive research description including all standard reporting details (study aim, type, methods, population, results, discussion) and following contact with the author it was established that no future article will be published on these data.

The critical appraisal skills programme tool CASP (Supplementary material 1) was used to evaluate the quality and rigour of each of the selected articles [49,50] prior to conducting the synthesis including the letter to the editor [51]. Although the assessment of quality in qualitative study methodology has long been debated, the CASP tool is one of the most commonly used tools in qualitative evidence synthesis [52]. Noyes et al. note that the CASP tool maps on to domains that provide an assessment of methodology strengths and limitations and it can be used with any qualitative research methodology. Two authors (AK and FB) independently assessed each study and any differences were discussed. The studies were then reviewed collaboratively and a consensus was reached. All papers had clear aims for which qualitative methodologies were appropriate. Two studies did not report their recruitment strategy and in line with their aims they presented raw unanalysed qualitative data [53,54]. The CASP tool

### Table 1. A summary of eMERGe reporting guidance [46].

| Phase | Article headings | Reporting criteria |
|-------|------------------|--------------------|
| Phase 1: selecting meta-ethnography and getting started | Introduction | 1. Rationale and context |
| | | 2. Aims |
| | | 3. Focus |
| | | 4. Rationale |
| | | 5. Search strategy |
| | | 6. Search processes |
| | | 7. Selecting primary studies |
| Phase 2: deciding what is relevant | Methods | 15. Synthesis process |
| Phase 3: reading included studies | Findings | 16. Outcome of synthesis process |
| Phase 4: determining how studies are related | Findings | 17. Summary of findings |
| Phase 5: translating studies into one another | Findings | 18. Strengths, limitation, and reflexivity |
| Phase 6: synthesising translations | Methods | 19. Recommendations and conclusions |
| Phase 7: expressing the synthesis | Discussion | |

### Table 2. Boolean operator terms.

| Boolean operator terms | Population (people with a neurodegenerative disease) | Exposure (gastostomy tube) | Outcome (experience) |
|-----------------------|-----------------------------------------------|-----------------------------|----------------------|
| S1                    | Amyotrophic Lateral Sclerosis OR ALS OR Alzheimer OR Dementia OR Huntington OR Motor Neuron Disease OR MND OR Multiple System Atrophy OR MSA OR Progressive Supranuclear Palsy OR PSP OR Multiple Sclerosis OR MS OR Neurodegenerative OR Neurological OR Parkinso | Artificial feeding OR Artificial nutrition OR Enteral feeding OR Enteral nutrition OR Feeding tube OR Parenteral feeding OR PEG OR PIG OR PEJ OR Tube feeding OR non-oral feeding OR Gastrostomy | Patient understanding OR Patient perspective OR Patient views OR Patient feelings OR Perspective OR Views OR Feelings OR World experience OR Lived experience OR qualitative OR phenomenology |
| S2                    | Language | Exclusion |
|                      | Time limits | Qualitative studies, either standalone or as part of mixed methods studies, where the qualitative element is reported in detail. Quantitative studies, large-scale surveys which include some qualitative data through open questions. Participants ≤18 years old. Studies with carers as well as people with neurodegenerative diseases where it was not possible to extract the findings from the person with a neurodegenerative disease. | English |}


Table 3. Characteristic of included studies.

| Author            | Study aim                                                                 | Type of study         | Theoretical approach/paradigm | Data collection method | Adaptations to interviews | Data analysis method | Sample size | Patient population | MND phenotype | Patient age | Patient gender | Socio economic Status | Country | Region |
|-------------------|---------------------------------------------------------------------------|-----------------------|--------------------------------|------------------------|---------------------------|----------------------|-------------|-------------------|--------------|-------------|----------------|----------------------|---------|--------|
| Leslie [53]       | To explore patients’ thoughts on information, values, outside pressure, support, and their reflections back on the process. | Qualitative report    | Not reported                   | Semi-structured interviews                      | Carers present            | Thematic analysis     | 2           | MND               | ALS/MND      | 67 and 75    | 2 female    | Not reported            | Texas    |         |
| Sterling et al. [54] | To share a patient with ALS’s perspective of her experiences of oral and non-oral feeding | Patient report        | Not reported                   | Written extract                |                           | Thematic analysis     | 1           | ALS               | ALS          | 61           | 1 female    | Not reported            | USA      |         |
| Stavroulakis et al. [37] | To explore the decision-making process in relation to timing of gastrostomy insertion from the perspective of the patients and their informal carers. | Retrospective qualitative study | Qualitative approach           | Semi-structured interviews | (1) Participants with significant communication difficulties used communication aids; (2) Interview questions were set out beforehand in order to prepare written responses if wished. (3) Carer/participant interviews were facilitated | Thematic analysis     | 10          | PwMND (8 carers were also interviewed) | ALS/MND      | Mean 67.1 (range 42–91) | *3 male, *7 female | Not reported            |         |
| Greenaway et al. [56] | To identify factors that influence decision making in accepting or declining NV and/or gastrostomy from the perspective of pwALS | Prospective qualitative study | Not reported                   | Not reported                     |                           | Descriptive qualitative research | 21          | MND               | MND          | Range 41 years 10 months to 76 years 2 months | *10 PwMND (8 carers were also interviewed) | Not reported            |         |
| Stavroulakis et al. [38] | To explore the effect of gastrostomy on the lives of patients with MND and their carers. | Retrospective qualitative study | Qualitative approach           | Semi-structured interviews | (1) Participants with significant communication difficulties used communication aids; (2) Interview questions were set out beforehand in order to prepare written responses if wished. (3) Carer/participant interviews were facilitated | Thematic analysis     | 21          | MND               | MND          | Mean 67.1 (range 42–91) | *3 male, *7 female | Not reported            |         |
| Pols and Limburg [57] | To analyse what quality of life comes to mean by studying it in the daily lives of patients | Theoretically informed qualitative study | Empirical ethics ethnographical study | Interviews and observations |                           | Stepwise inductive analysis | 33          | MND               | MND          | Not reported | Not reported | Not reported            | Australia |         |
| Chhetri et al. [52] | To explore patients’ experiences with non-oral feeding and its impact on their QoL | Prospective qualitative study | Not reported                   | Questionnaire                   |                           | Interpretive description approach | 19          | PwMND (35 carers were also interviewed) | ALS/familial ALS = 12 | Mean 67.3 years (range 40–79 years) | 10 male, 9 female | Not reported            |         |
| Labra et al. [35]  | To explore: (1) the factors that may impact on gastrostomy uptake and (2) the reasons why people with MND accept or decline gastrostomy decisions for pwMND and their carers. | Cross-sectional mixed-methods study design | Interpretive description methodology | Semi-structured interviews |                           | Interpretive description with inductive analysis | 12          | MND               | ALS          | Mean 67.3 years (range 40–79 years) | 10 male, 9 female | Not reported            |         |
| Paynter et al. [58] | To describe the lived-experience of making healthcare decisions for pwMND and their carers. | Qualitative study      | Qualitative approach           | Semi-structured interviews |                           |                          | 3          | MND               | MND          | Mean 67.3 years (range 40–79 years) | 10 male, 9 female | Not reported            |         |

(continued)
does not provide results that are easily translated into "low," "medium," and "high" quality classifications and so a "deciding criteria" can be used to establish relative study quality and its importance for the review [55, p.39]. In this qualitative synthesis, the presentation of qualitative data from participants with NDD living with, or in the process of deciding whether to live with, a GT was considered the deciding criteria for inclusion in the review.

**Data extraction approach and determining how the studies are related**

Following the completion of phases 1 and 2, each article which was deemed relevant to the research question was meticulously read by all authors individually to understand the overall argument of the article. Study characteristics and key contextual information were extracted from each article (Table 3). Data related to the experience of GT were extracted from across all articles. Both participants’ quotes and authors’ concepts were extracted (first- and second-order constructs). At the initial stage, the emerging themes were coded in NVivo 12 Pro by two authors (DL and AK) independently. As a theme or concept was discovered, a quote was selected to reference each point alongside researcher’s notes related to the theme [42]. A summary table documenting the list of themes was also created for better clarity. Subsequently, three consensus meetings were held between two authors (AK and DL) to agree on the dominant themes and the hierarchy of themes. If agreement could not be reached, the 3rd author (FB) was available to resolve any disagreements and facilitate consensus.

**Translating and synthesising studies**

Reciprocal and refutational translation of studies were conducted systematically. Disconfirming cases were included in the translation and further informed the synthesis. First, studies were organised in a chronological order and where possible second-order constructs within themes were compared and translated from one study to another to develop third-order constructs or explanatory theories. Two papers included predominantly first-order constructs [53, 54].

DL led this process initially before a consensus was reached between all authors. The line of argument synthesis which aims to provide a new interpretation and explanation of the phenomenon [46] was created by all authors.

**Findings**

**Selected studies**

Following the systematic search, 2863 articles were identified through electronic databases and a further four through screening of reference lists. After a systematic elimination process (see Figure 1, PRISMA), nine articles were selected for inclusion in this meta-ethnography (Table 3).

All studies gathered qualitative data either as the sole source of data or as part of a mixed methods study [35]. Two studies did not employ methods of qualitative data analysis and presented the unanalysed data in the paper [53, 54]. Four of the studies were conducted in the UK [37, 38, 51, 56], one in the Netherlands [57], one in the USA [54], and two in Australia [35, 58]. One study does not report where the research took place but the author’s affiliations were based in the USA [53].

Seven of the studies gathered data using interviews and one study also integrated data from observations [57]. One study
used a questionnaire with both Likert scale and open questions [51] and one shared a patient’s written account of personal experience [54]. Four of the studies outlined procedures undertaken to support participants with communication difficulties during interviews [37,38,53,56]. These procedures included involving carers in the interviews if the patient wished, use of communication aids (e.g., Litewriters), and sending out the interview questions beforehand to allow participants to prepare written responses in advance [37,38]. Four studies looked at factors that influence decision making for GT insertion [35,37,53,56], one looked at healthcare decision making in general which included GT [58]. Four studies explored the patient experience of, and quality of life associated with, a GT [38,51,54,57]. In two of these studies, the participants also reflected on their decision making prior to GT insertion [38,54]. Greenaway et al. investigated decision making in relation to both GT and non-invasive ventilation; however, data related to GT only were extracted for first-order concepts and where possible for second-order concepts [56].

A total of 146 participants with MND were included in the meta-synthesis. All were either living with a GT or considering GT insertion. Four studies reported the phenotypes of MND with a dominance of the limb onset \( (n = 37) \) in comparison with \( n = 32 \) bulbar onset and \( n = 3 \) primary lateral sclerosis [35,37,38,58]. Two studies investigated people with MND and their caregivers [37,58]. Only the data from the participants with MND were included in the first-order concepts. It was generally not possible to separate out the experience of carers and participants with MND in the second-order concepts, as they were presented based on the whole study population rather than being divided into the two groups (patients and caregivers). A study by Stavroulakis et al. interviewed 10 people with MND and eight carers, these participants were followed up and their data are reported in a subsequent study included in the meta-synthesis [38].

One study does not describe the age or gender of participants and reports findings on data that the authors gathered during interviews for their study in conjunction with interview and observation data gathered by a colleague [57]. The experiences of 54 males and 53 females were explored in the other eight studies and two case report studies presented the experience of female participants only [53,54]. Where described, the participants ages ranged from 40 to 91 years. There was no reference to socioeconomic status, ethnicity, or urban versus rural living in any of the studies. Seven of the studies reported funding was awarded and five were funded by charities that support research into MND [37,38,51,56,58].

The outcome of translation
Following the process of reciprocal and refutational translation initially within each study and subsequently across all nine studies, all authors developed new theories relevant to each theme. These theories are presented as third-order interpretations (Tables 4 and 5).

The outcome of synthesis
The hierarchy of themes which emerged across the studies was established. Subordinate themes identified were clustered into two superordinate themes: (1) decision making (Table 4) and (2) everyday living with a GT (Table 5). The first superordinate theme was noticeably richer in relation to the quantity of data and involved a much deeper level of investigation in comparison with the second theme. Data presented below is a synthesis of first and second order constructs from all included studies. An effort was made to preserve the original meaning of all studies when
Table 4. Theory development: decision making (superordinate theme 1).

| Superordinate theme | Influence of other people | Choice and control | Influence of dysphagia |
|---------------------|---------------------------|-------------------|-----------------------|
| I: decision making  | HCP and families are increasingly involved in decision-making regarding GT. HCP may provide the right level of information. | Participants felt involved in the decision-making regarding GT, especially those who were advised to opt for GT early. Wishing to continue oral diet was the main factor for delaying GT insertion. | Patient who chose to have GT: "I was frightened of choking. (…) I find I won't do that again!" Patient who chose not to have GT reported feeling: "I was scared. Because you were choking." |
|                     | Sterling et al. [54]      | Participants accepted GT when they were facing physical symptoms and when they considered GT as the only alternative. | Patients' perceptions of dysphagia and nutritional status contribute to GT acceptance, but are not primary or sole reasons. Perceptions of own dysphagia are not always in line with clinical measures. This (continued)
Table 4. Continued.

| Superordinate theme | Leslie et al. [53] | Sterling et al. [54] | Stavroulakis et al. [37] | Greenaway et al. [56] | Stavroulakis et al. [38] | Pols and Limburg [57] | Chhetri et al. [52] | Labra et al. [53] | Paynter et al. [58] | Third order interpretation |
|---------------------|--------------------|---------------------|-------------------------|----------------------|-------------------------|---------------------|-------------------|-----------------|-----------------|--------------------------|
| 1. decision making |                    |                     |                         |                      |                         |                     |                   |                 |                 | delay GT insertion for as long as they perceive themselves able to manage oral diet or until the fear of choking becomes a real concern. Addressing future/potential problems are not equally important for everyone. Although own perceptions of dysphagia appear to play an important role in the decision making, the final decision depends on many other personal factors. Professionals should spend time investigating personal perceptions of dysphagia and not focus on the clinical measures alone. |
| GT in the wider context of MND | Patient who chose to have GT: “I won’t be getting the right amount of minerals and nutrients that the body needs while I’ve still got the use of my arms and legs and I’m so mobile.” | Patient who chose not to have GT: “ Didn’t think it (GT) would help with all the other things going on.” | The onset of bulbar symptoms or experiencing them for a short time has not triggered the decision to accept GT. Uncertainty over trajectory of MND delayed the decision. | Witnessing physical deterioration, acceptance of the disease, and willingness to sustain life impacted decisions regarding GT. | Onset of MND was an important factor: in bulbar onset people perceived GT as a solution to their current swallowing problems, in the limb onset they approach GT as a solution potentially required in the future. | The rate of disease progression potentially contributes to GT uptake: when “steady” rate of decline was predicted participants accepted GT; when “slow” or “rapid” rate of decline was predicted participants were more likely to refuse GT. | The disease trajectory and access to information in advance impact on opportunity for choice for GT. | Current and expected trajectory of MND (symptoms location and progression rate) impact decision-making regarding GT. The decision regarding GT is deeply rooted in the trajectory of MND. |
| Personal values | Patient who chose to have GT: “I should gain some quality of life back that I’ve lost.” | Patient who chose not to have GT: “Just didn’t want a tube” | “I could either starve to death or consent to having a PEG tube inserted.” | Participants who accepted MND and who wished to prolong their survival were more likely to accept GT. | Personal values (such as the aesthetic aspect of body image) influence the perceptions on GT. | Personal values, such as aiming to prolong own life or the importance of own body image, play a role in decision-making process regarding GT. | | | | |

(continued)
Table 4. Continued.

| Superordinate theme | Leslie et al. [53] | Sterling et al. [54] | Stavroulakis et al. [37] | Greenaway et al. [56] | Stavroulakis et al. [38] | Pols and Limburg [57] | Chhetri et al. [52] | Labra et al. [53] | Paynter et al. [58] | Third order interpretation |
|---------------------|--------------------|---------------------|--------------------------|-----------------------|--------------------------|------------------------|----------------|----------------|----------------|--------------------------|
| Timing              | The timing of the discussion regarding GT placement must be carefully considered. | When I first became aware of the possibility that I might need a PEG tube, I said no way would I do that. | Best timing of GT insertion was not clear for participants. None of them opted for an early insertion despite advice from HCP. | Participants waited for the right time to accept GT. Their focus was not on the future, but the present. The concept of time was influential for the decision-making process and consisted of “living in the moment”; “right thing, right time” and “predicting the future.” GT recommended too early was viewed as not necessary. | Timing of GT is hard to be established as the perceptions of GT change (from “a frightening symbol of deterioration” to “becoming an eraser of complaints and concerns”). The medical perspective is often not in line with personal perspective of the GT’s identity. Timing when the identity of GT changes – people see the value of it in the way it can enable them to do nice things. Participants approached the progression of MND one step at the time. | GT discussions should take place early, be person-centred, exploratory, regularly reviewed, and should factor lengthly delays in patient’s decision-making. Some participants consented to GT to address future issues such as nutrition. | There is a fine line between preparing and traumatising people when providing early information. | Some people are willing to address potential/expected future problems more than others. There are two main points when the timing should be carefully considered by HCP: 1. The optimal timing of recommending GT 2. The optimal timing of insertion of GT. The above timings are influenced by personal perceptions of GT (personal readiness) and are likely to vary for different people. HCP have role in guiding people with MND to ensure their perceptions of GT evolve in line with the timing of medical recommendations. |
| Emotions and feelings | Emotive words used in SLTs may be “uncomfortable” and “helpless” discussing progression of MND or “unsure” how to respond. Patient who chose to have GT: “Frightened.” (…) “I’m worried about the procedure.” Patient who chose not to have GT: “frightened” and “shocked” | Uncertainty regarding the progression of MND and/or the benefits of GT. GT may be perceived as stigma (“not normal”). | Fear of the future, fear of the procedure, fear related to previous health experiences emerged as influencing decision-making. Also, both emotional support and emotional pressure from family members. | People have to recover from the shock of the diagnosis before they can start reorganising their life and make decisions including GT. People may avoid thinking about their own decline. | Wanting to be comfortable, hoping to reduce burden or increase energy levels, stress related to meals influenced decision-making. | The decision-making process can evoke multiple emotions and feeling in people with MND (and also HCP). These should be recognised and support should be offered. | |

*HCP: healthcare professionals.*
Table 5. Theory development: living with GT (superordinate theme 2).

| Superordinate theme 2: living with GT | Leslie et al. [53] | Sterling et al. [54] | Stavroulakis et al. [37] | Greenaway et al. [56] | Pols and Limburg [57] | Stavroulakis et al. [38] | Chhetri et al. [52] | Labra et al. [35] | Paynter et al. [58] | Third order interpretation |
|--------------------------------------|--------------------|---------------------|-------------------------|---------------------|--------------------------|-------------------------|---------------------|---------------------|---------------------|-----------------------------|
| **Problems**                         |                    |                     |                         |                     |                          |                         |                     |                     |                     |                             |
| **Insertion**                        | GT insertion was referred as "terrible" by all participants. | The meaning and the perceptions of GT change over time. For people who highly value their body appearance, GT was seen as a deformation of their body and increased their suffering. The negative views of GT preceded more positive experiences. Site of onset important as people with bulbar onset and preserved physical function benefit from GT tube more. | GT complications, feed administration, handling the equipment, supplies and storage, tube discomfort, time burden, and loss of ability to share a meal were listed as challenges related to GT. Loss of ability to share a meal was reported as a key adjustment and this can lead to significant psycho-social consequences. | Other consequences of MND were perceived as more severe than GT for some people. | The majority of participants reported little or no problems with GT. Problems were most common during the first 3 months post insertion. The majority developed some clinical complications (leakage, pain, redness/irritation, bleeding, and infection). GT was also associated with social isolation, loss of eating related pleasure, changes in body image and in attire. Problems with handling of GT, dependence on others, and family conflicts were reported. Some participants did not regret their decision to accept GT, some wanted to have it removed at 6 months, but none of them wished for a GT removal at 12 months. | People often feel unprepared for any complications related to insertion of GT, this needs to be addressed by HCP before consent for GT insertion is sought. The problems associated with GT are more common during the first few months, so more support should be provided during this time. The perceived problems depend on personal values, but also on the onset of MND (the more physical ability is maintained the less problems associated with GT). MDT support is required (e.g., OT to assist with dressing, handling of equipment, planning daily activities; psychologist to offer help with altered body image and social impact of GT, medics and nurses to educate and offer pre /post GT insertion support; SLT to facilitate oral intake if desired, dietitian to manage the feeding regime). |
| **Feeding regime**                   | "During the process of regulating and balancing my feedings, I was overfed, which caused me to throw up and bleed." | "Some days I feel like my body has been altered with the tube hanging and winding around my stomach. I don't feel whole anymore (…)." | "I ... the surgeon put the tube on my waistline. Try finding jeans to fit around a feeding port and tube." | "My friends and family have a hard time eating in front of me. (… ) No one asks me to go out to eat anymore." | Benefits "I admit the PEG tube has taken away the negative time and ..." | Benefits of GT outweigh other issues such as altered body image. | Reduced anxiety, stabilisation of weight, and prolonged survival | GT typically improved perceived QOL. Advantages from GT were usually noted |
| **Body image**                       |                    |                     |                         |                     |                          |                         |                     |                     |                     |                             |
| **Clothes**                          | "During the process of regulating and balancing my feedings, I was overfed, which caused me to throw up and bleed." | "Some days I feel like my body has been altered with the tube hanging and winding around my stomach. I don't feel whole anymore (…)." | "I ... the surgeon put the tube on my waistline. Try finding jeans to fit around a feeding port and tube." | "My friends and family have a hard time eating in front of me. (… ) No one asks me to go out to eat anymore." | Benefits "I admit the PEG tube has taken away the negative time and ..." | Benefits of GT outweigh other issues such as altered body image. | Reduced anxiety, stabilisation of weight, and prolonged survival | GT typically improved perceived QOL. Advantages from GT were usually noted |
| **Benefits**                         | "I admit the PEG tube has taken away the negative time and ..." | "Some days I feel like my body has been altered with the tube hanging and winding around my stomach. I don't feel whole anymore (…)." | "I ... the surgeon put the tube on my waistline. Try finding jeans to fit around a feeding port and tube." | "My friends and family have a hard time eating in front of me. (… ) No one asks me to go out to eat anymore." | Benefits "I admit the PEG tube has taken away the negative time and ..." | Benefits of GT outweigh other issues such as altered body image. | Reduced anxiety, stabilisation of weight, and prolonged survival | GT typically improved perceived QOL. Advantages from GT were usually noted |

(continued)
| Superordinate theme 2: living with GT | Leslie et al. [53] | Sterling et al. [54] | Stavroulakis et al. [37] | Greenaway et al. [56] | Pols and Limburg [57] | Stavroulakis et al. [38] | Chhetri et al. [52] | Labra et al. [51] | Paynter et al. [58] | Third order interpretation |
|-------------------------------------|-------------------|--------------------|---------------------|-----------------|-----------------|-----------------|-----------------|-----------------|-----------------|-----------------------------|
| Support from family                 |                   |                    |                     |                 |                 |                 |                 |                 |                 | energy I spent trying to eat. |
|                                     |                   |                    |                     |                 |                 | Satisfaction with GT may develop over time when GT facilitates/enables positive events. GT allows for enjoying food without pressure. Personal values, and therefore perceived benefits from GT, may change over time as people adjust to their disabilities. People with bulbar onset perceived most benefits from GT as it addressed their swallowing problems and weight loss while they were still able to use their limbs well. The availability of informal carers impacts on the effect of GT. | were the main benefits of GT. The majority of participants perceived the benefits to outweigh the problems. a few months after insertion and generally outweighed the problems. GT offered a sense of relief and security, was perceived as essential for survival, facilitated travelling, taking medications, improved nutrition, weight gain, and saved time and energy. GT was also reported to reduce anxiety. | from GT and that the perception can change over time. This would require an ongoing support. Personal values (such as love for food or the importance of body image) impact on the perceived benefits from GT. This should be evaluated at the start so people are supported in understanding their own reactions and expectations regarding living with GT. |

Support from HCP

HCP such as SLTs are faced with complex discussions regarding dysphagia. They should be appropriately prepared and have a specified format of managing emotionally difficult conversations. HCP should provide appropriate training in relation to GT which needs to be comprehensive (decision making, insertion, living with GT) and individualised (in content and timing). The authors propose a training pathway, where 3 training sessions are offered (1, at the time of referral for GT; 2, post-insertion in hospital; and 3, at home). It is important for HCP to understand, identify and manage the problems associated with GF. Healthcare systems should better facilitate well-timed access to GT after decision have been made.

Little is known how support from families impacts the personal experiences of living with GT – from the perspective of the person with GT. HCP play an important role in supporting people living with GT. The support should be individualised based on individual needs and provided by MDT. Training for patients and families should be delivered at different stages of living with GT, a one-off format does not seem to be sufficient.
developing third order constructs. This process is illustrated in Tables 4 and 5.

Superordinate theme 1: decision making

Seven studies reported data regarding the process of deciding whether to accept or decline a GT [35,37,53,54,56-58]. All the seven studies considered this decision as complex and individualised. A synthesised summary of subordinate themes (Table 4) which emerged across the seven studies is presented below.

Subordinate theme 1: influence of other people

The influence of other people, such as healthcare professionals or family members, was evident during the decision-making process. Having trust in healthcare professionals and feeling supported by them was important [37,56]. Some participants relied heavily on healthcare professionals to guide decision making regarding GT [58]. However, not every professional was perceived as having sufficient knowledge or experience of MND [56]. Getting inaccurate or inconsistent information from a professional caused consternation and was reported as disturbing [56]. Healthcare professionals were the main source of information regarding GT [37] and having multiple sources of information could make the decision easier [56]. Providing the right amount of information based on individual needs was important [37,53,56]. Some people felt pressured by healthcare professionals when making a decision regarding GT which subsequently led to emotional tension and stress [56]. Emotional tension was also evident in relation to the fear of the procedure [53,56,57], the altered body [53,57] and the negative perceptions of a GT as something “not normal” [37]. Liasing with other people with a GT was valuable [37,54] but it was also emotionally challenging [54].

Family also influenced decision making; family’s opinions were valued [53] and having family support “marginally” increased the acceptance of GT [56]. Aiming to reduce caregivers’ burden caused both acceptance and refusal of GT insertion [35,56]. One study reported that the presence of family can be both a source of support and increased pressure [56].

Subordinate theme 2: choice and control

Perceptions of choice and maintaining a sense of control over one’s own body emerged across all seven studies. Participants wished to have an active role during decision making and take responsibility for their choices [37,53,56,58]. They were often facing a double decision: should they accept or reject GT and when to accept or reject it [58]. Some participants appeared to take control by self-selecting different information from different healthcare professionals to influence and then change their decision for GT [58]. Some viewed a GT as essential for survival [35,51,54,56]. They perceived not having any other option but to accept a GT [53,54,56,57] as the only alternative was to “starve to death” [54].

Subordinate theme 3: influence of dysphagia

Self-perceptions of dysphagia contributed to acceptance of a GT. If a symptom of dysphagia (especially choking) was perceived as a current concern, a person was more inclined to engage in discussions about a GT and to accept insertion [35,37,54,56,57]. One study highlighted that the personal perceptions of their own swallowing ability may not be in line with clinical diagnosis [35]. In a separate study, there was limited discussion regarding the potential influence of GT on social and emotional aspects of eating and drinking and the authors suggested this was likely due to participants being interviewed while their swallowing function was unaffected by the illness and so the issue had not yet been considered by them [58].

Subordinate theme 4: GT in the wider context of MND

Uncertainty with regards to the future progression of MND complicated decision making about GT [37,58]. The onset of MND and the rate of progression affected decision making for some people (further explained in Table 4) [35,37,57]. Acceptance of MND was also reported as a factor influencing decision making regarding GT [56].

Subordinate theme 5: personal values

Personal philosophy of life and values, such as wishing to prolong one’s own life or to avoid any life prolonging intervention [35,53,56] as well as the desire to live in the here and now and not to focus on any future concerns [56] were described as factors influencing decision making in relation to GT.

Subordinate theme 6: timing

A tendency for late acceptance of a GT was noted [37,57] with some participants delaying their decision for as long as they could [57]. Time was required to arrive at a “turning point” and to see “valuable opportunities” related to GT, to change own perception of GT from a frightening “symbol of deterioration” to an “eraser of complaints and concerns” [57]. All these complex factors seemed to evolve over time. One study noted that “the right time” can only be determined retrospectively as no one can predict the exact trajectory of MND [56]. There is a fine line between preparing and traumatising people when providing early information [58].

Studies reported that healthcare professionals must understand the problems associated with the decision-making process [51] and consider both philosophical and pragmatic issues related to a GT [53]. Healthcare professionals may not have a standardised approach to discussing GT [53]. They may receive little training and learn on the job [54]. Appropriate timing of GT discussions was important [37,56] as well as ensuring a sensitive approach [58]. Early discussion of GT is recommended to factor in decisional delays [35].

Super-ordinate theme 2: living with a gastrostomy tube

The everyday experiences of living with a GT (Table 5) emerged across four studies [38,51,54,57].

Subordinate theme 1 and 2: problems and benefits

All of them reported both problems (subordinate theme 1) and benefits (subordinate theme 2) associated with GT. Some participants felt unprepared for any complications following GT insertion [54,57] and reported this procedure as “terrible” or “horror” [57]. Multiple clinical and psycho-social problems were reported in all four studies. However, participants were generally pleased with their decision to accept GT. Some perceived other issues related to MND as more significant than any problems related to GT [57]. The positive experiences of living with a GT typically developed a few months after GT insertion and were considered to outweigh any GT related problems [38,51,54,57]. The onset of MND appeared to influence participants’ experiences of GT. For people with bulbar onset (where dysphagia was present but other regions of the body were largely unaffected), GT was “a facilitator of happy events” [57] and it provided a secure way to meet nutritional needs [51]. For people with spinal onset (where other regions of the body were affected before dysphagia was present), GT was “a transformation of misery” especially if they were unable...
to administer own feeds and had no caregivers (subordinate theme 3: Support from family) [57].

Subordinate theme 3 and 4: support from family and healthcare professionals
Although there was limited information on how support from families impacts the personal experiences of living with GT, one study proposed that the availability of informal carers is important for GT effect and function [57]. Support from healthcare professionals (subordinate theme 4) was considered important also. Studies reported that healthcare professionals should be prepared to handle complex emotional discussions [54] and should recognise and manage any difficulties experienced in relation to living with a GT [51]. The authors of one study reported that individual training should be provided to people following insertion of GT [38]. Once a person has consented for GT, healthcare professionals should act immediately and there should be no delays with the insertion procedure [35].

Line of argument
This review identified that decision making in relation to accepting or refusing a GT is a complex and individualised psychological process, often developing over a long period of time. As a GT is frequently considered essential for survival, a person is not only making a decision to accept an alternative way of hydration and nutrition but to prolong their life.

A line of argument was developed to provide a comprehensive overview of the lived experience of GT in MND as an example of NDD based on the findings of this review and authors’ interpretations (Figure 2).

All studies in this review reported on the roles and influences of healthcare professionals with regards to the experiences of GT in MND. We advocate that healthcare professionals and researchers should look at the entire experience of GT holistically rather than breaking the experience into pre and post insertion. Based on the findings of the studies included in this review, the crucial times to offer people support are: (1) during the process of decision making, (2) at the time of the insertion procedure, and (3) while living with GT post insertion. In our interpretation, there is a relationship between the experience of decision making and the experience of living with GT (as demonstrated by the arrow forward in Figure 2). The perceived quality of experience during decision making, appears to influence how quickly people perceive benefits from GT following insertion. Difficulties experienced during the decision-making time combined with an often traumatic insertion procedure may affect personal satisfaction with GT during the first few months after insertion. More positive experiences tend to develop over time. Studies included in this review reported that healthcare professionals play an important role in influencing the experiences of people with MND and in our opinion they have the potential to improve patients’ experiences of GT. During the decision-making time, healthcare professionals should provide continuous support and the opportunity to collaboratively discuss the future of living with GT (as demonstrated by arrow backwards in Figure 2). The conversations should focus not only on health-related issues (i.e., weight and nutrition), but also the potential impact on participation in daily activities. Extending discussions beyond the decision-making time, providing a balanced level of information regarding how to live with GT, and optimising the future experiences can make the decision-making time less traumatic and challenging. It seems that if a person feels...
well supported, facilitated and guided by healthcare professionals during the decision-making time, the person’s level of decisional conflict may reduce.

We advocate that the input from the whole multidisciplinary team (MDT) is crucial during the decision-making process and it should not cease after GT insertion. It should be provided continuously not only to resolve any clinical complications related to the GT, but also to incorporate GT into everyday routines, to allow a person to continue oral diet as much as appropriate, and ultimately to reduce any negative experiences related to a GT. Healthcare professionals should ensure that individuals with NDD are provided with correct information (tailored to their personal needs) and that this information has been understood. This is necessary to make a fully informed decision. The support, recommendations, and information should be consistent across the team. There is no universal best timing for discussion of GT and this timing should be individually assessed by the MDT. Personal perceptions should be investigated in addition to clinical measures, especially in relation to self-perception of swallowing ability. The potential appropriateness of GT should be discussed in the context of the whole trajectory of the disease and the individual’s living context, not only from a nutritional or respiratory perspective.

The evidence related to the lived experiences of GT in NDD is lacking and only a small number of studies focus on the experiences of GT in MND. The use of GT should be guided by the personal values, beliefs, and the individual needs of the person in collaboration with the MDT. Making the decision to accept or decline GT can be challenging and can have long-lasting impact on people.

Discussion

Summary of findings

This article systematically analysed and synthesised the available qualitative literature on patients’ experiences of GT in MND (as the only type of NDD identified by this review). The findings indicate that there is a small number of studies on this topic and these studies are predominantly focused on the decision-making time before GT insertion. There is lack of representation from populations living with other types of NDD in the current body of knowledge. Vessey et al. explored factors that influence decision making for GT insertion among participants with multiple sclerosis and cerebellar ataxia. However, this study was excluded from our review as it included participants with progressive pharyngeal scarring and unknown diagnoses and the data related to NDD could not be extracted [34]. The research gap related to the lack of representation from other NDD may be explained by the differences in the management of each individual disease. For example, GT insertion is generally discouraged in advanced dementia and only recommended occasionally in the earlier stages of this disease [27,59]. Non-oral feeding is also not common in Parkinson’s disease [23], but it may be applicable if a person is in a long-term care setting [60]. However, approximately 40% of people with MS present with dysphagia and the majority of these require non-oral feeding [61]. This reflects a gap in the current research investigating the experiences of GT in NDD outside MND. MND is also distinctive in relation to survival time which is typically estimated at 3 years [62] compared with 15.8 years in Parkinson’s disease [63], 15–24 years in Huntington’s disease [64], and 6.7 in dementia [2]. It is also distinct in terms of the onset and severity of dysphagia which typically presents early and is more severe in bulbar onset of MND [12, 14, 65].

Decision making is an individualised process which can be complex for some people with MND [35,37,54,56,57] while others perceive no decisional conflict [34]. The support provided by healthcare professionals during the decision-making time can potentially influence experiences post insertion. A sense of relief following GT insertion can occur despite the complications and difficulties that often arise in the first few months [51,54,57,66]. Healthcare professionals have an important role in supporting people with MND and they can influence personal experiences of people with MND in positive and negative ways [35,37,53,54,56–58]. To ensure early development of positive experiences of GT, healthcare professionals must develop rapport, trust, and offer appropriate individualised support [38,66]. It is vital that clinicians take the time to listen to the patient’s concerns and provide them with continuous opportunities to discuss queries regarding their condition and the intervention [67]. It is clear from this research that patients value the opinions of their healthcare professionals [37,56] and a reciprocal approach in which healthcare professionals consider the patients’ perspectives, values, and beliefs is key to collaborative practice.

Clinical guidelines for GT in MND recommend MDT assessment of nutrition and hydration, severity of dysphagia and factors influencing safe swallowing including fear of choking as well as other psychological considerations [68]. Personal perceptions of dysphagia should also be investigated as they can influence the final decision to accept or refuse a GT and may differ from outcomes of clinical examination [35,58].

People with MND may delay insertion of GT in order to continue with oral diet [37,53,57] despite the fact that oral diet is often possible in the presence of GT [57]. The inclusion of oral feeding supplementary to GT appears to be often misunderstood. This emphasises the need for effective communication between healthcare professionals and people with MND [53,57]. Stavroulakis et al. recommended that healthcare professionals should provide training regarding GT three times across the entire experience of GT (before insertion, after insertion in hospital, and after discharge from the hospital at home) [38]. As there can be a great deal of knowledge for patients to absorb, it is critical to provide regular access to information allowing the person to process this at the most convenient time for them. The creation of digital GT decision-making aids, accessible online to both patients and families, may be valuable in supporting shared decision making and allowing flexible access to information [36].

Strengths, limitation, and reflexivity

This review was conducted with a great degree of rigor and is reported following the eMERge guidelines [46]. This is one of the few meta-ethnography studies which included people with first-hand experience of GT as co-authors. This provided a critical eye alongside a unique interpretation of evidence.

This review was limited to studies published in the English language only. No time limits for literature searches were applied. All studies included in this review recruited participants with MND. As MND presents with a unique disease trajectory, findings cannot be easily generalised to other NDD. The studies were carried out in developed countries with no reference to socioeconomic factors among the participants. Only four studies described accommodations applied to facilitate individuals with communication difficulties to participate in the interviews. Some of the included studies provided limited reference to qualitative methodology and the relationship between the researcher and the participants is not clearly explored. All studies had a clear aim relevant
to the topic of this review. However, the majority focused on decision making rather than living with GT. Due to the presentation of qualitative data without qualitative analysis methods in two studies, the authors were required to work with both first- and second-order data throughout the theory development. In studies that presented data from both people with MND and their carers, the authors extracted first-order concepts from people with MND only. This was not always possible for second-order concepts. A similar process was followed for the study including GT and non-invasive ventilation.

Three authors are qualified healthcare professionals, and two of them represent academic institutions and have over 10 years of experience in adult neurology and dysphagia. Although they were new to conducting meta-ethnography, they received training and had experience of other qualitative research methods. The authors followed the eMERGe guidelines [46] and the involvement of people with an NDD as co-authors improved the credibility and trustworthiness of the study. The authors acknowledge their professional background and personal experiences may have influenced their interpretations of findings. During the development of theory and the line of argument, it became apparent that the authors who are healthcare professionals had a less optimistic view of GT in comparison to the authors who live with MND and GT. The discussions of these different viewpoints were valuable as both professional and personal perspectives were compared and investigated until a consensus was reached in regard to the line of argument synthesis. The amalgamation of these viewpoints facilitated the co-production of meaningful, relevant, and accessible information in this review, highlighting the benefits of PPI involvement in health research.

**Recommendations and conclusions**

This review shows that the lived experience of GT in MND is complex and individualised. Healthcare professionals, as part of an MDT, should provide a holistic approach across the trajectory of the illness, exploring perceptions and values of people with MND. This will allow them to provide appropriate individualised support throughout the entire experience of GT (during the decision-making time as well as post-insertion). The structure of health services should ensure that both healthcare professionals and patients are provided with sufficient time to build rapport, discuss management options, including potential benefits and problems, as well as examine personal values. Flexible service provision should be advocated for in response to individual needs of those with MND.

Further research is indicated to explore the lived experiences of GT in various types of NDD. These experiences should be investigated holistically rather than being divided into pre and post-insertion. Some individuals living with NDD may also present with communication and cognitive difficulties. Future studies should ensure that these individuals are supported and included within the research design.

**Disclosure statement**

The authors declare no conflict of interest associated with this publication.

**ORCID**

Dominika Lisiecka [http://orcid.org/0000-0002-7633-3076](http://orcid.org/0000-0002-7633-3076)
Aine Kearns [http://orcid.org/0000-0002-6596-2740](http://orcid.org/0000-0002-6596-2740)

**References**

[1] Patterson C. World Alzheimer report 2018: the state of the art of dementia research: new frontiers. London: Alzheimer’s Disease International (ADI); 2018.
[2] Rait G, Walters K, Bottomley C, et al. Survival of people with clinical diagnosis of dementia in primary care: cohort study. BMJ. 2010;341:c3584.
[3] Logroscino G, Piccinnini M, Marin B, et al. Global, regional, and national burden of motor neuron diseases 1990–2016: a systematic analysis for the Global Burden of Disease Study 2016. Lancet Neurol. 2018;17(12):1083–1097.
[4] Chiò A, Logroscino G, Hardiman O, et al. Prognostic factors in ALS: a critical review. Amyotroph Lateral Scler. 2009;10(5–6):310–323.
[5] Feigin VL, Nichols E, Alam T, et al. Global, regional, and national burden of neurological disorders, 1990–2016: a systematic analysis for the Global Burden of Disease Study 2016. Lancet Neurol. 2019;18(5):459–480.
[6] Westeneng H-J, Debray TPA, Visser AE, et al. Prognosis for patients with amyotrophic lateral sclerosis: development and validation of a personalised prediction model. Lancet Neurol. 2018;17(5):423–433.
[7] McDermott CJ, Shaw PJ. Diagnosis and management of motor neurone disease. BMJ. 2008;336(7645):658–662.
[8] Gladman M, Zinman L. The economic impact of amyotrophic lateral sclerosis: a systematic review. Expert Rev Pharmacoecon Outcomes Res. 2015;15(3):439–450.
[9] Arthur KC, Calvo A, Price TR, et al. Projected increase in amyotrophic lateral sclerosis from 2015 to 2040. Nat Commun. 2016;7(1):12408.
[10] van der Graaff MM, de Jong JMBV, Baas F, et al. Upper motor neuron and extra-motor neuron involvement in amyotrophic lateral sclerosis: a clinical and brain imaging review. Neuromuscul Disord. 2009;19(1):53–58.
[11] Orrell RW, Guiloff RJ. Clinical aspects of motor neurone disease. Medicine. 2020;48(9):607–611.
[12] Dharmadasa T, Henderson RD, Talman PS, et al. Motor neurone disease: progress and challenges. Med J Aust. 2017;206(8):357–362.
[13] Phukan J, Elamin M, Bede P, et al. The syndrome of cognitive impairment in amyotrophic lateral sclerosis: a population-based study. J Neurol Neurosurg Psychiatry. 2012;83(1):102–108.
[14] Oliver DJ. Palliative care in motor neurone disease: where are we now? Palliative Care. 2019;12:1178224218813914.
[15] Robbins J. Swallowing in ALS and motor neuron disorders. Neurol Clin. 1987;5(2):213–229.
[16] Oliver D. The quality of care and symptom control – the effects on the terminal phase of ALS/MND. J Neurol Sci. 1996;139:134–136.
[17] Briani C, Marcon M, Ermani M, et al. Radiological evidence of subclinical dysphagia in motor neuron disease. J Neurol. 1998;245(4):211–216.
[18] Bensimon G, Lacomblez L, Meineinger V. A controlled trial of riluzole in amyotrophic lateral sclerosis. ALS/Riluzole Study Group. N Engl J Med. 1994;330(9):585–591.
[19] Breiner A, Zinman L, Bourque PR. Edaravone for amyotrophic lateral sclerosis: barriers to access and lifeboat ethics. CMAJ. 2020;192(12):E319–E320.
[20] Miller RG, Mitchell JD, Lyon M, et al. Riluzole for amyotrophic lateral sclerosis (ALS)/motor neuron disease (MND). Cochrane Database Syst Rev. 2002;2:CD001447.
quality appraisal in qualitative evidence synthesis. Res Methods Med Health Sci. 2020;1(1):31–42.

[56] Greenaway LP, Martin NH, Lawrence V, et al. Accepting or declining non-invasive ventilation or gastrostomy in amyotrophic lateral sclerosis: patients’ perspectives. J Neurol. 2015;262(4):1002–1013.

[57] Pols J, Limburg S. A matter of taste? Quality of life in day-to-day living with ALS and a feeding tube. Cult Med Psychiatry. 2016;40(3):361–382.

[58] Paynter C, Mathers S, Gregory H, et al. How people living with motor neurone disease and their carers experience healthcare decision making: a qualitative exploration. Disabil Rehabil. 2020;1–9.

[59] Nice G. Dementia: assessment, management and support for people living with dementia and their carers; 2018 [cited 2019 Mar 11]. Available from: https://www.nice.org.uk/guidance/ng97

[60] National Collaborating Centre for Chronic Conditions. National Institute for Health and Clinical Excellence: Guidance. Parkinson’s disease: national clinical guideline for diagnosis and management in primary and secondary care. London: Royal College of Physicians of London; 2006.

[61] Scalfari A, Knappertz V, Cutter G, et al. Mortality in patients with multiple sclerosis. Neurology. 2013;81(2):184–192.

[62] Hardiman O, van den Berg LH, Kiernan MC. Clinical diagnosis and management of amyotrophic lateral sclerosis. Nat Rev Neurol. 2011;7(11):639–649.

[63] Oosterveld LP, Allen JC Jr., Reinoso G, et al. Prognostic factors for early mortality in Parkinson’s disease. Parkinsonism Relat Disord. 2015;21(3):226–230.

[64] Rodrigues FB, Abreu D, Damásio J, et al. Survival, mortality, causes and places of death in a European Huntington’s Disease Prospective Cohort. Mov Disord Clin Pract. 2017; 4(5):737–742.

[65] Luchesi KF, Kitamura S, Mourão LF. Amyotrophic lateral sclerosis survival analysis: swallowing and non-oral feeding. NeuroRehabilitation. 2014;35:535–542.

[66] Hazzard E, Gulliver S, Walton K, et al. The patient experience of having a feeding tube during treatment for head and neck cancer: a systematic literature review. Clin Nutr ESPEN. 2019;33:66–85.

[67] Hogden A, Greenfield D, Nugus P, et al. What influences patient decision-making in amyotrophic lateral sclerosis multidisciplinary care? A study of patient perspectives. Patient Prefer Adherence. 2012;6:6829–838.

[68] National Institute for Health and Care Excellence (NICE). Motor neurone disease: assessment and management. NICE; 2016. Available from: https://www.nice.org.uk/guidance/ng42/resources/motor-neurone-disease-assessment-and-management-pdf-1837449470149