A mixed methods study exploring how multidisciplinary teams support people with motor neuron disease to make decisions about gastrostomy placement

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17th February 2025

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Contents

Abstract

Background: People with MND are supported to make decisions about gastrostomy by several healthcare professionals (HCPs) across the multidisciplinary team (MDT). Despite recommendations for early intervention, decision-making is associated with uncertainty. The aim of this research was to understand the factors that influence decision-making about gastrostomy in the context of MDT care delivery.

Methods: A mixed methods design was employed. First, a qualitative evidence synthesis of 27 papers aimed to understand the perspectives of people with MND, caregivers and HCPs of intervention decision-making. Second, a survey of 139 HCPs aimed to understand HCPs' practice and beliefs about supporting people with MND considering gastrostomy. Finally, a qualitative multiple case study including three UK MND teams, used 42 observations, 39 interviews, documentation and medical note reviews, and 3 focus groups to understand how MDTs supported decision-making of people with MND about gastrostomy in real-world settings.

Findings: HCPs were not always able to initiate discussions about gastrostomy as early as they preferred. People with MND frequently experienced emotional responses to information about the disease and gastrostomy, influencing the timing of engagement in discussions and decisions. Some people with MND sought to extend normality by avoiding decision-making presenting a barrier to HCPs' proactive preferences. HCPs are a valued source of support for people with MND, controlling initiation of discussions and guiding timing of decisions. People with MND need for concrete guidance about gastrostomy timing was not always met by HCPs. The involvement of multiple HCPs presented a challenge to delivering coordinated decision-support. A lack of a strategic approach and access to quality interprofessional communication contributed to an uncoordinated approach.

Conclusions: Decision-making about gastrostomy can be emotionally driven and heavily influenced by the MND care context in which decisions are made. Decisions are distributed over time, space and people. MND teams should develop a strategic approach, grounded on

strong lines of interprofessional communication to coordinate the decision support they deliver to people with MND.

List of abbreviations

AAC	Augmentative and alternative communication
ACT	Acceptance and Commitment Therapy
AHP	Allied Healthcare Professional
ALS	Amyotrophic Lateral Sclerosis
ALS ALLOW	Ascertain-Leave opportunity-Stratify-Anchor-Let it be-Listen in silence-Offer over time-Work together
BANS	British Artificial Nutrition Survey
CAG	Confidential Advisory Group
CASP	Critical Appraisal Skills Programme
CHERRIES	Checklist for Reporting Results of Internet E-Surveys
CNS	Clinical Nurse Specialist
EMA	European Medicines Agency
ENTREC	Enhancing transparency in reporting the synthesis of qualitative research
EPR	Electronic Patient Record
FDA	Food and Drug Administration
HCP	Health Care Professional
HEN	Home Enteral Nutrition
HRA	Health Research Association
IP-SDM	Interprofessional Shared Decision Making
MND	Motor Neuron Disease
MDT	Multi-Disciplinary Team
MHRA	Medicines and Healthcare products Regulatory Agency
NICE	National Institute for Health and Care Excellence
NIHR	National Institute for Health and Social Care Research
NIV	Non-Invasive Ventilation
NHS	National Health Service

PEG	Percutaneous Endoscopic Gastrostomy
PIG	Percutaneous Image-Guided Gastrostomy
PPI	Patient and Public Involvement
pwMND	People/person with motor neuron disease
REC	Research Ethics Committee
RETREAT	Review question–Epistemology–Time/Timescale–Resources–Expertise–Audience and purpose–Type
RIG	Radiologically inserted Gastrostomy
SDM	Shared Decision Making
SOD1	superoxide dismutase 1
SPIKES	setting, perception, invitation, knowledge, emotion and strategy
SLT	Speech and Language Therapist
UK	United Kingdom
WHO	World Health Organisation

List of tables

The numbering of the tables of Papers 1-4 (Chapters 4-7) remains the same as they are included in the published papers (see below). The tables included in the *introduction*, *background*, *methodology and methods*, and *discussion* chapters, are in their own numerical order.

Chapters 1-3 and 8

Paper 1 (Chapter 4): Qualitative evidence synthesis

Paper 2 (Chapter 5): HCP survey

Paper 3 (Chapter 6): Multiple case study: timing of discussions and decisions

Paper 4 (chapter 7): Multiple case study: Coordination of MDT decision support

List of figures

The numbering of the figure of Papers 1-4 (Chapters 4-7) remains the same as they are included in the published papers (see below). The figures included in the *introduction*, *background*, *methodology and methods*, and *discussion* chapters, are in their own numerical order.

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Paper 4 (chapter 7): Multiple case study: Coordination of MDT decision support

Dissemination

The research included in this thesis has been disseminated in a number of forums and formats as described below.

Peer reviewed publications

White, S, O'Cathain, A, Halliday, V, Croot, L, McDermott, CJ. Factors influencing decisions people with motor neuron disease make about gastrostomy placement and ventilation: A qualitative evidence synthesis. Health Expect. 2023; 1- 18. doi:10.1111/hex.13786

White S, O'Cathain A, Halliday V, Bradburn M, McDermott CJ. Supporting people with Motor Neuron Disease (MND) to make decisions about gastrostomy feeding tube placement: a survey of UK healthcare professionals' practice and beliefs. Amyotroph Lateral Scler Frontotemporal Degener. 2024 Feb 9:1-9. doi: 10.1080/21678421.2024.2314061. Epub ahead of print. PMID: 38337170.

Conference presentations

Gastrostomy placement in MND: a simple or complex decision? Invited speaker at NNNG annual conference in Nottingham on 14/6/23.

Gastrostomy placement in MND: a complex decision distributed over time and people. Invited speaker at BAPEN annual conference on 29/11/23.

How do multidisciplinary teams support people with MND to make decisions about gastrostomy placement: a qualitative multiple case study. Abstract accepted for oral presentation at International MND Symposium. Presentation delivered on December 8th 2024 in Montreal.

Conference posters

White S, O’Cathain A, Halliday V, Bradburn M, McDermott C (2023). Decision making about gastrostomy placement in MND: a survey of UK healthcare professional beliefs and practice. Poster presented at ENCALS Annual conference in Barcelona on 12-14th July 2023.

White S, O’Cathain A, Halliday V, Croot L, McDermott C (2023). Decision-making about gastrostomy placement and ventilation in motor neuron disease (MND) care: a qualitative evidence synthesis. Poster presented at ENCALS Annual conference in Barcelona on 12-14th July 2023.

Other presentations

Supporting patient-centred decisions about gastrostomy placement. Nutricia education event. Dublin. (invited speaker). 26th May 2022.

Exploring how the multidisciplinary team supports people with MND making the decision to have a gastrostomy feeding tube placed. Sheffield BRC training academy presentation day. 22nd June 2022.

Supporting people with MND to make decisions about gastrostomy placement. MND Association Community of Practice Knowledge exchange online event. 28th June 2022. (invited speaker)

Gastrostomy? Supporting patient-centred decision making. Invited speaker at MSA Annual study day in Sheffield (SITRAN) on 19/10/23.

Supporting decision making within the context of MND. Invited speaker at Zambon study event in Nottingham on 5/12/23.

Multiple presentations to a variety of decision making and MND academic and clinical audiences, during 2-week placement at Aarhus, Denmark on doctoral research. 19/4/24 - 5/5/24.

How does the motor neuron disease (MND) multidisciplinary team support people with MND to make decisions about gastrostomy? Invited speaker at Sheffield Teaching Hospitals NHS Foundation Trust Combined Community and Acute (CCA) Care Group Research Meeting on 15/6/24.

Contribution to the academic work

The conception, design, acquisition of funding (via my NIHR Clinical Doctoral Research Fellowship), data collection, analysis, or interpretation of data for the research presented in this thesis was my responsibility. I received support through monthly supervision meetings and feedback on draft paper manuscripts and this thesis from Professor Christopher McDermott, Professor Alicia O’Cathain and Dr Vanessa Halliday, all of whom were co-authors of the publications. Dr Liz Croot kindly supported me as a second reviewer, screening 10% of the title/abstracts and the selected full texts for the qualitative evidence synthesis and was included as co-author for the publication (White *et al.*, 2023). Mike Bradburn (statistician) supported me with completing the comparative statistical tests for the HCP survey and was included as co-author for the publication (White *et al.*, 2024).

I will be including as co-authors of the multiple case study papers, Professor Bekker (Professor of Medical Decision Making) from The University of Leeds as she supported me with the conceptualisation of the project and Sarah Birchall (Research Coordinator) from Sheffield Teaching Hospitals NHS Foundation Trust as she has contributed through developing the protocol, support with gaining NHS research ethics committee (REC) approval and setting up the research sites.

Acknowledgements

I have many people I would like to thank who have supported me throughout the last three years. Completing my doctoral fellowship and this thesis has been an emotional journey of highs and lows for me. I am certain this would not have been possible without the constructive academic feedback, and moral support when needed, from my amazing team of supervisors including Professor Christopher McDermott, Professor Alicia O'Cathain and Dr Vanessa Halliday. Each supervisor provided me with a different source of support, which together complimented each other perfectly. I would like to thank my thesis advisors, Dr Susan Baxter and Dr Johnathon Cooper Knox, for their critical appraisal at my confirmation review and thesis plan milestones whose advice with hindsight enabled me to make some important pragmatic decisions needed to submit within my funded period. I would also like to acknowledge the helpful discussions I had with my tutor, Professor Tim Chico, who gave reassurance and motivation to keep going.

Throughout my fellowship I received excellent peer review and advice from a group of academics and healthcare professionals who bought a massive amount of experience and knowledge to the fellowship. I would like to thank Emma Wood, Kathryn Webster, Margaret Boddy, Professor Hilary Bekker, Professor Markus Reuber, Dr Anne Hogden, Professor Bobbie Farsides. I would especially like to acknowledge the additional support that Professor Hilary Becker has given me prior to and during my fellowship. Her insight into preparing the research plan and mentorship throughout the fellowship has been invaluable and I hope to continue to collaborate with her in the future.

Dr Liz Weekes who kindly agreed to be a mentor has provided a shoulder to lean on including exploring how I could develop a clinical academic career. Liz's significant clinical

and academic experience has been a great source of guidance and supported me to plan for the next steps in my career.

This has been my first experience of having to set up a multi-centre study and it came with a lot of research governance challenges which I was naïve to. Sarah Birchall, my amazing research coordinator at Sheffield Teaching Hospitals Clinical Research and Innovation Office, has guided me through the research governance requirements, including preparing applications for ethical and HRA approvals, setting up sites and dealing with any issues associated with these.

I am very grateful for the people living with MND and their families who have helped develop this research plan and provided some honest reflections and advice on the research design and interpretations. I have met with some amazing people who have given up their time and assisted me to make some key decisions throughout the research. This research would not have been possible without them.

I have been lucky enough to be part of an amazing research team at SITRaN. Professor McDermott, as part of his NIHR professorship has assembled an experienced group of academic and clinical researchers all focused on trying to improve the care that people with MND receive. I won't name them all, but I have found our trips to Elm coffee shop a great source of academic and moral support and hope to collaborate with them again in the future.

I would like to give thanks to all the people with MND and their caregivers, who were kind enough to allow me to observe their intimate and difficult discussions with their HCPs, and then talk about their experience in such candid detail. I would also like to acknowledge the HCPs who allowed me into their workplace and gave up their valuable time to take part in interviews and focus group. I would like to thank the principal investigators (PI) within each case (I am unable to name them to protect the anonymity of them as research participants and of the location of the cases). I would not have gained access to the cases or met my recruitment targets without them.

I would like to thank my wife Lucy and my two children, Eve and Ben. They have all been extremely understanding and tolerable of the time I have had to dedicate to completing this doctoral fellowship over the last three years. It has involved me being away for prolonged periods of time to attend conferences, research placements and site visits, and spending many evenings and weekends locked away in my writing cave. I appreciate that life does not stop because I am doing a PhD and therefore, I am thankful that I have had my families support to get this completed.

Finally, I would like to dedicate this thesis to the memory of my Grandad, Fred White (pictured below), who sadly passed away with MND when I was 15 and continues to be my motivation for trying to make life for people with MND slightly less awful than it is today. Continue to rest in peace Fred.

Figure 1. My Grandad, Fred, who sadly passed away with MND when I was 15.

1. Introduction

1.1 Chapter outline

The following chapter provides an overview of the topic area including a summary of MND, and why many people with MND face the difficult decision to have a gastrostomy at some stage of the disease course. I summarise how the clinical, psychosocial and organisational factors when combined create a complex context within which healthcare professionals and people with MND collaborate about the options available. A justification is made for studying how MND teams support people with MND making decisions about gastrostomy in real-world settings. A description and justification for the recursive thesis format used is provided, which is a hybrid of traditional and publication thesis formats. An overview of the content each chapter is described. Finally, a reflexive account of my own clinical and personal experience is provided, to allow an understanding of the position I was in when commencing this doctoral fellowship and how it has informed the development of the research presented.

1.2 Introduction to the clinical and research topic area

Motor neuron disease (MND) (also known as amyotrophic lateral sclerosis (ALS)) is a neurological condition that involves the progressive loss of the motor neuron function (Talbot, 2002). Motor neurons carry the electrical signals from the brain, via the spinal cord and onward to the muscles, to allow for the functions relied on for daily function and to sustain life itself. The devastating consequences for most people with MND include the onset and then progressive loss of the ability to speak, move, breath and swallow. People with MND can also experience non-motor symptoms including 30-50% developing cognitive impairment and 10% frontotemporal dementia (Phukan *et al.,* 2007; Benbrika *et al.,* 2019). MND is often referred to as a rare disease, with a prevalence of 4.02-4.91 per 1,00,000 population in the UK (Hoppitt *et al.,* 2011). This is comparable with a global incidence of 1.59 (95% CI 1.39-1.81) per 1,00,000 person-years and prevalence of 4.42 (95% CI 3.92-4.96) per 1,00,000 population (Xu *et al.,* 2020). However, an individual's life-time risk is just 1 in 350(males)-472(females) (Alonso *et al.,* 2009), which is explained by a short average survival of just two to five years after diagnosis (Logroscino *et al.,* 2008; McDermott and Shaw, 2008).

There is currently no cure for MND, and the only pharmaceutical treatment available in the UK is Riluzole which extends survival by a moderate 2-3 months (Miller *et al.,* 2012). Therefore, therapeutic options currently remain focused on compensating for the consequences of motor neuron denervation (McDermott and Shaw 2008). Many of the physical, social and psychological consequences of MND present a barrier to people with MND being able to eat and drink safely or in adequate quantities to meet their nutritional requirements (Coates *et al.,* 2023). Dysphagia, which affects 85% of people with MND, presents a risk of malnutrition and of aspirating food into the lungs and is associated with increased morbidity and mortality (Worwood and Leigh 1998; Sorenson *et al.,* 2007; Genton, 2011; Burkhardt *et al.,* 2017). There are growing signals in the literature that weight loss is associated with reduced survival and quality of life and is currently the target of nutritional interventions aimed at slowing the disease course (NIHR [RP-PG-1016-20006], 2018; Ning *et al.,* 2019; Janse van Mantgem *et al.,* 2020; Ludolph *et al.,* 2020). However, strategies to protect the airway and optimise the nutritional content of oral dietary intake only remain effective for a limited period of time before people with MND need to consider alternative routes of nutrition, hydration and medication administration (Stavroulakis *et al*., 2013).

The main alternative route of nutrition routinely offered to people with MND is that of gastrostomy tube placement (Stavroulakis *et al*., 2013). Gastrostomy placement involves a short hospital admission to surgically place a narrow plastic tube into the stomach allowing the administration of enteral nutrition, fluids and medications (Stavroulakis *et al.,* 2013). People with MND and their caregivers can usually be trained to care and use the gastrostomy in their own homes, though some may need support from formal carers or transfer into care homes (Lisiecka *et al*., 2021a; Lisiecka *et al*., 2022).

Evidence for efficacy of enteral tube feeding in MND in terms of nutritional, quality of life and survival outcomes remains equivocal with a randomised controlled trial of enteral nutrition in MND unlikely to be feasible for ethical reasons (Sulistyo *et al.,* 2023). Observational studies have found a survival benefit associated with enteral nutrition or improved outcomes when gastrostomy is placed earlier in the disease course (ProGAS study, 2015; Lopez-Gomez *et al.,* 2021; Castanheira *et al.,* 2021; Yang *et al.,* 2024). In addition to weighing up the risks and benefits of enteral nutrition, people with MND also need to consider the procedural risks. Respiratory failure and increased weight loss is associated with worse procedural outcomes (Kasarskis *et al.,* 1999; ProGAS study, 2015). However, gastrostomy can be safely placed even in the presence of significant respiratory failure, when additional precautions are taken to manage the risks (Sancho *et al.,* 2010; van Neerven *et al.,* 2024). This research and professional consensus have informed professional guidance that encourages people with MND, whose informed preference it is proceed with enteral nutrition, to do so prior to experiencing significant dysphagia or malnutrition and/or before the risks associated with placing a tube become prohibitive (Miller *et al*., 2009a; Burgos *et al.,* 2018; NICE, 2019; Van Damme *et al.,* 2024). The decision about the timing of gastrostomy placement is complicated by a lack of consensus about when enteral feeding should commence in relation to the indications of dysphagia, nutrition, respiratory failure and mealtime burden (Van Eenennaam *et al.,* 2021). A further organisational consideration to consider is the time it takes healthcare services to organise the procedure (Labra *et al.,* 2020). Delays placing a gastrostomy once clinically indicated exposes people with MND to the increased risk of poorer outcomes associated with worsening malnutrition and aspiration risk (ProGAS study, 2015; Son *et al.,* 2024). There is a lack of guidance that synthesises the range of clinical and organisational factors that healthcare professionals and people with MND need, to inform when discussions should start, the optimal timing of decisions and the timing of commencing enteral nutrition (Miller *et al.,* 2009a; Burgos *et al.,* 2018; NICE, 2019; Van Damme *et al.,* 2024).

On the face of it, the decision to have a gastrostomy placed may appear to be a simple one: people with MND faced with a significant and progressive health threat (i.e. dysphagia and/or malnutrition) have a relatively low risk intervention available (i.e. gastrostomy placement and enteral nutrition) to address it. So why do people with MND not immediately accept the option when it is offered by healthcare professionals (Labra *et al*., 2020; Son *et al*., 2024)? While there is a strong clinical argument for gastrostomy placement, people with MND do not make decisions about gastrostomy in isolation, with consideration of clinical indications alone or without being influenced by the context in which they are making them. Decisions about care and intervention options in MND have been described as ‘the worst-case decision-making context' (Hogden, 2014). Emerging qualitative literature has sought to better understand this context from the individual perspectives of people with MND , caregivers' and healthcare professionals' (King *et al.,* 2009; Hogden *et al.,* 2012; Foley *et al.,* 2014a; Foley *et al.,* 2014b; Stavroulakis *et al.,* 2014; Greenaway *et al.,* 2015; Labra *et al.,* 2020). Throughout the disease course, people with MND are asked to make multiple, and sometimes simultaneous, decisions about their care (e.g. house adaptions) and treatments (e.g. NIV), in addition to considering the option to having a gastrostomy placed. The response of people with MND to the physical and psychosocial consequences of MND have been shown to evolve over the disease course as they learn to live with, and accept, the diagnosis (Albert *et al.,* 1999; King *et al.,* 2009; Foley *et al.,* 2014a). Individually, the perceptions of people with MND about what the intervention involves and will mean for them and their caregivers, are influenced by their own personal histories, values and priorities (Stavroulakis *et al.,* 2014; Greenaway *et al.,* 2015; Labra *et al.,* 2020). Healthcare professionals and caregivers are an important and influential part of the context in which people with MND are making decisions. Both can protect or threaten individuals' autonomy, through enabling people with MND to understand their preferences for intervention, or by exerting pressure on them to make a decision (Foley *et al.,* 2014b; Greenaway *et al.,* 2015). Decisions about the timing of gastrostomy placement have been associated with delays the time it takes people with MND to make decisions (Labra *et al.,* 2020; Son *et al*., 2024). A potential source of decisional conflict could be the lack of consensus between healthcare professionals about the optimal timing of gastrostomy placement and how this is communicated by the multidisciplinary team (MDT) over multiple clinical interactions (Van Eenennaam *et al.,* 2021; Chapman *et al.,* 2021). Decisions about if, and when, to have a gastrostomy are influenced by many disease, patient-centric and organisational contextual factors (Stavroulakis *et al.,* 2014; Labra *et al.,* 2020), which have not, to date, been studied in a real-world context.

Following diagnosis, people with MND are recommended to receive support from a MDT of healthcare professionals, to address the physical, psychological and clinical consequences of MND (NICE, 2019). MND teams can include a range of professional disciplines working across several different settings including the clinic and in the homes of people with MND (OPM, 2016; Kierkegaard *et al.,* 2021). In the UK, people with MND are often supported by one of the 22 MND care centres or networks that have been established by the MND Association with other people with MND being supported by general neurology and therapy services operating within acute and community settings (MND Association (no date)). An integration of tertiary, secondary and primary care is employed to allow people with MND to have access to MND expertise and care delivered closer to home (WHO, 2006). When needed people with MND may utilise secondary care (i.e. hospital admissions), to access tests, undergo procedure (e.g. gastrostomy placement) or to address an acute clinical issue (e.g. chest infection) (WHO, 2006). However, access to specialist tertiary care is limited for people with MND living in developing countries due to a lack of expertise and funding (WHO, 2006). Attending specialist MND MDT clinics is associated with improved survival of 7-10 months (Ng *et al.,* 2009; Aridegbe *et al.,* 2013). While MDT care is recommended in professional guidance, there remains a lack of evidence about the mechanisms behind this positive effect on outcomes and how healthcare professionals should collaborate to deliver coordinated care (Jannsens *et al.,* 2016).

Earlier and quicker access to life-prolonging interventions is likely to be a key mechanism for the beneficial impact of MDT care in MND. An argument could be made that earlier access to interventions is supported by the timing, coordination and quality of the decision support delivered by healthcare professionals, individually and collectively. Any delays in services offering interventions, or in decision-making attributed to how the options are discussed, could lead to delays in starting the intervention. The decision to place a gastrostomy is unique in that it comes under the remit of many different professional disciplines across the MND team. Neurologists, respiratory services, MND nurses, dietitians, and speech and language therapists all play a role in supporting the decision-making of people with MND about gastrostomy placement (Van Eenennaam *et al.,* 2021). Published surveys have revealed a lack of consensus about the use and timing of gastrostomy placement among healthcare professionals and different healthcare disciplines (Rio *et al.,* 2007; Oliver *et al.,* 2011; Ruffell *et al.,* 2013; Van Eenennaam *et al.,* 2021). There remains a lack of evidence that has sought to understand the variations in practice and beliefs of the multiple professions involved and how they coordinate the decision support they deliver across the MDT.

There is a paucity of research conducted to understand how people with MND make decisions about gastrostomy in the context of real-world practice, from the perspectives of the different people involved. The following thesis presents a three-component mixed-methods study that has aimed to produce an in-depth and rich account of how decisions are made about gastrostomy in MND care, including how MND teams coordinate the support they provide to people with MND, and identify potential targets for future decision-support interventions or service improvements. The three components of this research presented sought to answer the following overarching doctoral research question:

> ‘How do motor neuron disease (MND) multidisciplinary teams support people with MND to make decisions about gastrostomy tube placement?’

1.3 Thesis format

A recursive thesis format has been used to present the research completed during my doctoral fellowship. This is a hybrid of a conventional and publication style thesis. In common with a conventional format thesis, it begins with an introduction and review of the key literature to make the argument for the research conducted, followed by a methodology and methods chapter justifying the methods used. However, the findings are included in four publication format chapters (Chapter 4-7), each including their own introduction, methods, findings and discussion sections. Finally, in the last chapter, the findings of each study are integrated and compared to the published literature.

Included in this thesis are two published papers presenting the findings from a qualitative evidence synthesis (White *et al.,* 2023) and HCP survey (White *et al.,* 2024). While the findings from the multiple case study are yet to be published, they are also presented in publication format and will be worked up for submission following the submission of this thesis. Using the publication format for my findings chapters has challenged me to present the key findings and supported the publication (or drafting) of papers that can be disseminated quickly within or soon after the clinical doctoral research fellowship, before I return to my clinical role as an NHS Dietitian in September 2024. With the papers focusing on the same overarching doctoral research question, there is the risk of some repetition particularly with regards to the methods sections of both multiple case study chapters. However, effort has been taken to ensure the introductions, findings and discussions are distinct from each other with each providing further depth to answering the research questions for each component of the study and the overarching doctoral research question.

A challenge associated with the thesis format chosen is with regard to a mix of referencing styles used and the numbering of tables and figures. Both journals within which the qualitative evidence synthesis (Paper 1) and survey (Paper 2) papers have been published required authors to use a two different reference styles, with Health Expectations using AMA style and Amyotrophic Lateral Sclerosis and Frontotemporal Degeneration using Vancouver style which involve numbered citations. For the purposes of this thesis, the referencing format has not been altered for these two published papers. For the two unpublished papers (Papers 3 and 4) the Harvard reference style has been used with each including their own reference lists. For the remaining chapters (i.e. introduction, background, Methods and Discussion) the Harvard reference style is used, as per my university departments referencing guidance, with the reference list available at the end of the thesis.

1.4 Summary of thesis chapters

The thesis is presented in 8 chapters. A summary of each chapter is given below.

Chapter 1 introduces the topic area including the condition of MND and how the consequences of the disease lead to people with MND needing to consider the decision to have a gastrostomy placed. A case is made for better understanding the contextual complexity of decision-making in MND about gastrostomy, including the supporting role played by MND healthcare professionals and teams. I acknowledge my position as an healthcare professional with significant clinical experience in the topic area including a summary of my personal and academic history. Finally, a summary of the thesis format and content is provided.

Chapter 2 presents a detailed background about the problem and identifies the gap in current understand that research aimed to address. Published research and guidance is summarised and critiqued to describe the complexity of the decision-making process taking place within the current models of MND MDT care provision. Current models of shared decision making are reviewed and applied to the MND context. Particular attention is given to the published qualitative literature, capturing people with MND, caregivers’ and healthcare professionals' experiences, perceptions and views of making decisions about gastrostomy placement and makes the argument that we are at a stage where the findings of these studies can be synthesised. An argument is made for studying the phenomena of decision-making occurring within real-world practice, using multiple perspectives to describe the contextual complexities at play.

Chapter 3 provides a rationale and description of the methodology used for the review and primary research conducted. I describe the subtle realist position I have taken for this research and how this has justified and guided the design, conduct and analysis of the research. This will include a justification for including each method included in this 3-component sequential mixed-methods design and how each contributes to a deeper understanding of the topic under study. A rationale for the qualitative review, survey and multiple case study methods is presented including additional information about each research method used that could not be included within the limited word count in publications.

Chapter 4 presents a qualitative evidence synthesis (Paper 1, White *et al.,* 2023), that synthesised the findings from qualitative studies focused on understanding how people with MND, caregivers' and healthcare professionals' experience and perceived making decisions about gastrostomy or ventilation in MND care. The synthesis identified the contextual challenges that needed to be better understood and informed the development of the survey and the multiple case study protocols.

Chapter 5 presents a cross-sectional survey of healthcare professionals' practice and beliefs about supporting people with MND making decisions about gastrostomy placement (Paper 2, White *et al.,* 2024). The survey findings identified sources of consensus and contention in healthcare professionals' views, including between the different professions involved in supporting the decision-making of people with MND about gastrostomy. Combined with the qualitative evidence synthesis, the survey findings identified concepts for further exploration during the multiple case study and informed the development of the *a priori* framework.

Chapters 6 and 7 present the findings of the multiple case study relating to the initiation of discussions, timing of decisions and coordination of MDT decision support. Chapter 6 (Paper

3) focuses on the timing of discussions and decisions about gastrostomy, and Chapter 7 (Paper 4) on how the MND team coordinate the decision support they deliver to people with MND about gastrostomy across the MDT.

Chapter 8 integrates the findings from the three components of research and compares the findings to the published literature and theory. The strengths and limitations of the study are discussed followed by an account of my reflexivity throughout the conduct of each study. The implications for further research and for practice are proposed. Finally, my main conclusions are presented.

1.5 Reflexivity prior to commencing doctoral research

The following reflexive account provides some background about myself, my clinical career and my journey towards conducting the research presented in this thesis. It is important to be transparent about how my personal history, experience and values influenced the design of the research presented in this thesis. I am a 47-year-old male, married to Lucy with two teenage children, Eve and Ben. I qualified as a dietitian in the NHS in 2001 and have worked for Sheffield Teaching Hospitals NHS Foundation Trust ever since, covering a variety of clinical areas. For most of my dietetic career (2004-2021) I have been a home enteral nutrition (HEN) dietitian, supporting people living at home on enteral nutrition, usually via a gastrostomy tube. The role involved visiting patients and their families in their own homes, advising on enteral feeding plans and managing tube related complications. The 17 years as a HEN dietitian has not only given me considerable experience of the clinical application of the intervention but spending time with people on HEN from leaving hospital with a tube in-situ until the end of their lives, has informed my understanding and empathy for the impact HEN has on patients' lives and my preference for qualitative research. My passion for the HEN role lead me to becoming the HEN clinical lead for the Parenteral and Enteral Nutrition Specialist Group of the British Dietetic Association (PENG), a role I still hold today. Over the years I have contributed and lead many local and national HEN related projects. I take a reflective approach to practice, learning from my own experience, other healthcare professionals and from people on HEN. Understanding and improving patients experience and quality of life has always been a priority for me and informed my decision to have a naso-gastric (NG) tube placed and be nil by mouth for a week in 2015. This experience further informed the empathy I have for patient's unfortunate enough not to have the choice that I had to remove the tube at the end of a week. The experience provided me with an insight into incorporating enteral nutrition methods into my daily life, the responses of other people to the NG tube, and having any oral diet or fluids.

It is through the HEN role that I developed a clinical and subsequently academic interest in how people make decisions about gastrostomy placement. As a team we recognised that people being discharged into our service had not always been well informed or prepared for life on HEN, and believed we could use our clinical experience to help inform peoples decisions pre-gastrostomy placement. We started to offer a 'pre-gastrostomy counselling service' to those people considering an elective placement of a gastrostomy, which mainly included those with progressive neurological conditions, including MND. It was a role valued by patients and those specialist teams that referred patients to us. However, I started to question whether our involvement was effective in supporting people to make informed choices or resulted in better decisions i.e. decisions that were aligned with peoples' preferences and values. These reflective thoughts were the beginning of my academic journey which commenced in earnest in 2014.

In addition to my clinical and academic interest in the nutritional management of people with MND, it is important to disclose that I have personal experience of the disease through my grandad, Fred, sadly passing away with MND when I was 15 years old. I like to refer to Fred in every presentation I do so he is not forgotten. Fred drives my motivation for the research I do and my passion for trying to make life a little less horrific for people with this awful disease. I would like to dedicate this thesis to his memory and all the other people who have passed away with MND.

I completed the NIHR funded MSc Clinical Research in 2015, including completing a qualitative study exploring the role of values in decision-making about gastrostomy placement (White and Brereton, 2018). The MSc confirmed my ambition to pursue a clinical academic career. Since completing the MSc I have worked closely with Professor Christopher McDermott, who is now my PhD primary supervisor. In 2015, Professor McDermott invited me to be part of the team co-designing with people with MND and their caregivers, the mytube website (available at: https://mytube.mymnd.org.uk/), which aimed to translate recent research findings (ProGAS study, 2015) into an online resource aimed at supporting the understanding of people with MND about the gastrostomy decision (White *et al.,* 2017). Following this, Professor McDermott invited me to be co-applicant on the £2.3 million NIHR programme grant for applied research, HighCALS. I was involved in each work package including supporting the design, conduct, interpretation and dissemination of the primary research and the development of a complex nutrition support intervention now being evaluated in a multi-centre randomised controlled trial (RCT). More recently I was invited to be on the expert advisory group for a research project developing a decision aid for people with MND considering gastrostomy (Maunsell *et al.,* 2019). Involvement in the projects above, in addition to my clinical experience, have helped improve my understanding and identify the gaps in practice and in the literature, on which this thesis is based. In 2021, following collaboration with my supervisors, peers, people with MND and caregivers, I was successful in applying for a 3-year full-time NIHR Clinical Doctoral Research Fellowship (CDRF), which the following thesis is the result of.

The above reflexive account is presented to be upfront about my position when I started this doctoral research. I was conscious of the need to be reflexive throughout the process of qualitative research design, conduct, reporting and dissemination. Throughout the research I have kept a reflexive journal and had discussions with my supervisors and colleagues, that have enabled me to challenge the decisions and interpretations I have made. I have included a further reflexive summary related to the research completed in the discussion (Chapter 8; Section 8.4.3).

1.6 Summary of chapter

The above has introduced the topic area in which this research is situated and identified the gap in the current literature in terms of understanding how the MND MDT support people with MND to make decisions about gastrostomy. The key points include that gastrostomy is routinely offered in MND care to address the risks associated with dysphagia and malnutrition. People with MND, caregivers and HCPs reporting finding decision-making complex highlights a need to better understand the context within which decisions about gastrostomy are being made. MND care is delivered by teams of professional disciplines, many of whom take an interest in the decisions people with MND make about gastrostomy.

A recursive thesis format is used which has challenged me to disseminate the findings of the research within (or soon after) the clinical doctoral fellowship. My previous experience as a HEN dietitian and personal contact with MND have informed my interest in the topic and the development of the research presented in this thesis. The following chapter involves a review and critique of the current literature, further clarifying the justification for the research planned.

2. Background

2.1 Chapter outline

The following chapter reviews and critiques the literature about MND, its consequences and decisions people with MND make about gastrostomy placement, including expanding on some of the literature included in the introduction provided in Chapter 1. Evidence is presented for why people with MND are routinely offered the gastrostomy option including addressing the disease-related risks of continuing without the intervention. Gastrostomy placement provides an alternative option and is itself associated with a range of outcomes and experiences that people with MND and healthcare professionals deliberate about during decision-making. The evidence that has focused on the procedure to place a gastrostomy, its outcomes and people with MND or caregivers' experience of life on enteral nutrition are reviewed, to provide an understanding of the intervention that people with MND are asked to consider. The decision about gastrostomy placement has been reported to be influenced by factors much wider than the clinical outcomes of the disease or intervention. Therefore, the mostly qualitative literature is reviewed describing how decisions are made in general in MND, and more specifically in relation to the option to have a gastrostomy. Finally, the gap in the literature is identified providing a justification for the mixed-methods study that was proposed.

2.2 What is motor neuron disease?

Motors neurons are nerves originating in the brain and travelling through the spinal cord, carrying the signals to the muscles to perform their function. MND is a progressive neurological condition which involves the degeneration of motor neurons and the associated progressive loss of multiple essential physiological functions. People can present with symptoms of upper, lower or mixed motor neuron loss. Amyotrophic lateral sclerosis (ALS) is the most common form of MND with other presentations including primary lateral sclerosis (PLS) and progressive muscular atrophy (PMA) (Kiernan *et al.,* 2011). About 20% of people with MND have bulbar onset disease which typically presents with slurred speech followed by the subsequent presentation of swallowing problems (dysphagia) (Kiernan *et al.,* 2011). The most common site of onset is limb onset, with early symptoms including weakness in the hands and lower limbs (e.g. foot drop). The aetiology of MND remains a major source of academic enquiry with improved understanding of the causes of the disease providing potential targets for pharmaceutical and genetic interventions. While the majority of diagnoses are sporadic, 5-10% are familial (Kiernan *et al.,* 2011). The identification of genetic causes of MND are the target of future treatment development that have the potential to slow, stop or even prevent disease (Shatunov and Al-Chalabi, 2021; Miller *et al.,* 2022).

In the UK the incidence of MND is 1.06-2.4 per 1,00,000 person-years and prevalence 4.02-4.91 per 1,00,000 population (Hoppitt *et al.,* 2011). This is comparable with estimates of global incidence of 1.59 (95% CI 1.39-1.81) per 1,00,000 person-years and prevalence of 4.42 (95% CI 3.92-4.96) per 1,00,000 population (Xu *et al.,* 2020). However, there are global variations, with South Asia reporting the lowest incidence (0.42) and prevalence (1.57) and Western Europe the highest (incidence: 2.76; prevalence: 9.62) (Xu *et al.,* 2020). The global prevalence of MND increase with age peaking at 70-79 years (Xu *et al.,* 2020). Again, this pattern is matched in the UK, with a study using a UK population-based sample finding that MND was more likely to present between the ages of 75-79 in both sexes (Alonso *et al.,* 2009). While MND is often considered a rare disease due it's low point prevalence (which remains low because of the short survival), the life-time risk of being diagnosed with MND is 1 in 350 in men (2.9 per 1000) and 1 in 472 in women (2.1 per 1000) (Alonso *et al.,* 2009).

A model has been developed and externally validated with a European cohort of people with MND, to predict prognosis (Westeneng *et al.,* 2018). However, due to the heterogeneity of the disease, it is difficult to give individuals accurate predictions at diagnosis and care is required with how such predictions are communicated to people with MND (Van Eenennaam *et al.,* 2020; Van Eenennaam *et al.,* 2021). The majority of people with MND have a predicted survival of three to five years after diagnosis (McDermott and Shaw, 2008). While most people with MND will experience limb weakness and bulbar symptoms at some point in the disease course, those with bulbar onset disease experience speech (dysarthria) and swallowing (dysphagia) problems earlier in the disease course than those with other sites of onset (Kiernan *et al.,* 2011). People with MND will often experience a range of other consequences of the disease including drooling (sialorrhoea), speech problems, respiratory failure and reducing mobility (upper and lower limbs).

Despite the huge amount of research taking place world-wide, in the UK there remains just one pharmaceutical option for people with MND which is riluzole. A survival benefit of 3 months of taking riluzole has been confirmed by the meta-analysis of results from four trials (Miller *et al.,* 2012). An RCT of tofersen has been shown to reduce Superoxide dismutase 1 (SOD1) in cerebral spinal fluid and neurofilament light chains in plasma (biomarker of motor neuron degeneration) but did not slow disease progression as measured by the ALSFRS-R score (a functional score for MND disease progressions commonly used as an outcome measure) (Miller *et al.,* 2022). However, an open-label extension found that people with MND starting tofersen earlier reported a slower decline in ALSFRS-R scores (Miller *et al.,* 2022). Tofersen was recently given approval by the Food and Drug Administration (FDA) in the US and the European Medicines Agency (EMA) in the European Union. However, tofersen is yet to be authorised by Medicines and Healthcare products Regulatory Agency (MHRA) or National Institute for Health and Care Excellence (NICE) in the UK, a decision that has caused consternation within the MND Association and anxiety for people with MND that the treatment could benefit (MND Association, 2024). The lack of therapeutic options to slow or stop the disease is a major source of upset for those being diagnosed, leads to a lack of hope for the future and is an important contextual factor in relation to how people with MND evaluate the utility of care and intervention options.

In the absence of many effective pharmaceutical options, much of the focus of MND care is on managing, or compensating for, the many physical consequences of the disease including interventions to support mobility, communication, respiratory failure and dysphagia (McDermott and Shaw, 2008). Mobility aids including walking aids and wheelchairs support people with MND to have some independence with their mobility. Adaptions to the homes of people with MND are often required to enable them to remain independent and safe for as long as possible. Augmentative and alternate communication (AAC) devices and strategies can support people with MND to communicate, including the recording of their voice (i.e. voice banking) to be used with electronic AAC devices (Cave and Bloch 2021). Non-invasive ventilation (NIV) is routinely offered to people with MND with declining respiratory function. NIV is one of the few interventions that has been proven to extend survival in MND and is routinely offered to people with MND presenting with respiratory symptoms or a decline in markers of respiratory function (Bourke *et al.,* 2006). NIV can only support the respiratory needs of people with MND for so long before they either pass away or consider invasive ventilation via a tracheostomy. The use of mechanical ventilation via a tracheostomy in MND varies around the world with higher prevalence reported in Japan compared to the US or Europe (Turner *et al.*, 2019). Mechanical ventilation is rarely used in the UK.

2.3 How is MND care organised and delivered?

International MND guidelines consistently recommend that people with MND have access to a range of professions who can provide support for the diverse psychological, social and physical consequences of the disease (Miller *et al*., 2009b; NICE, 2019; Hogden *et al*., 2020; Shoesmith *et al*., 2020; Berlowitz *et* al., 2023; Petri *et al*., 2023; Van Damme et al 2024). The models of MND care vary around the world and are dependent on the geography and resource of the different countries (WHO, 2006; Kierkegaard *et* al., 2021; Bublitz *et* al., 2024). The research presented in this thesis is focused on UK models of MND care delivery and how it impacts on the delivery of decision-support about gastrostomy. However, the conceptual findings are likely to be transferable to MND care settings outside of the UK. The UK's NICE MND guideline recommends service should: '*Provide coordinated care for people with MND, using a clinic-based, specialist MND multidisciplinary team approach. The clinic may be community or hospital based.*' (NICE, 2019). People with MND receive care in a variety of settings including clinic, hospices, hospital wards and their own home (OPM, 2016). To account for the low point prevalence of MND and to facilitate the maintenance of a source of MND expertise, care is often delivered using a tertiary care model (WHO, 2006). In the UK, this involves care centred around a specialist MND clinic, often hosted in hospitals or hospices, which provide an efficient method by which healthcare services can assemble a range of HCPs with MND expertise. However, acknowledging the increasing difficulty people with MND may experience travelling sometimes long distances to clinic as they become more physically disabled, there is a need to bring care closer to the homes of people with MND. Community healthcare teams are often able to deliver care closer to where people with MND live, including through domiciliary visits. Recently, service, technology and research efforts have focused on the implementation and assessment of care provided remotely including the use of telemedicine (Hobson *et al.,* 2019; Helleman *et al.,* 2020).

There is evidence for improved health outcomes for people with MND who receive MDT MND care (Aridegbe *et al.,* 2013; Paipa *et al.,* 2019), informing the global professional recommendation for people with MND to attend a specialist MND clinic (Miller *et al.,* 2009b, NICE 2019). Mechanisms for these improved outcomes have been proposed including improved clinical decision making and earlier access to interventions such as riluzole, NIV and gastrostomy (Miller *et al.,* 2009b). Though improved outcomes are frequently attributed to attendance at a specialist MND clinic, MND care is delivered, and interventions accessed, through interaction with healthcare professionals across all settings including community services visiting people with MND at home (OPM, 2016). No RCTs were identified in a Cochrane systematic review of the effectiveness of MND MDT clinics, with low level evidence for improved quality of life, reduced admissions and improved disability and mixed findings in relation to survival (Ng *et al.,* 2009). More recently, a meta-analysis found significantly improved survival, with a mean difference of 141.67 (CI 95%, 61.48 to 221.86, p=0.0005) between people with MND attending a multidisciplinary clinic and those attending general neurology care (De Almeida *et al.,* 2021). However, the meta-analysis was based on results from just two prospective cohort studies with high heterogeneity (I^2=87%). The absence of evidence does not mean MDT care is not effective (Ng *et al.,* 2009). The complexity and variability of MND care, and the establishment of the MDT care model as a gold standard of practice will make it difficult evaluate using RCTs. It is my hypothesis that earlier access to interventions could be enabled through the timing and quality of the decision support people with MND receive from the MDT about such interventions, justifying making understanding the decision-making process a research priority. Future research is required to better understand how people with MND and healthcare professionals reach decisions about treatment options and the impact these interactions have on the timing of interventions and on health outcomes.

2.4 Why do people with MND need to consider gastrostomy placement and enteral nutrition?

As introduced above MND is a complicated, multisystem disease associated with many physical challenges. Many of the consequences of MND present a barrier to people with MND being able to eat and drink enough safely or in adequate quantities and each can represent a clinical indication for considering gastrostomy placement and commencement of enteral nutrition (Stavroulakis *et al*., 2013). Having multiple indications contributes to the complexity of making decisions about gastrostomy, including uncertainty about when each may present and the rate of progression. Table 1 lists the different indications and why they indicate a need to consider gastrostomy.

Table 1 Common indications for gastrostomy and reasons why they indicate a need to consider a gastrostomy.

Indication for gastrostomy	Reason why indicates need to consider gastrostomy
Dysphagia	Dysphagia presents a risk to people with MND of aspiration and choking. Aspiration can lead to chest infections and pneumonia, which is associated with significant morbidity and mortality risk. Dysphagia can also result in a poor mealtime experience including prolonged times to finish meals and avoidance of eating in public.
Malnutrition	Weight loss can be the consequence of taking inadequate calorie intake (in addition to the loss of muscle mass due to motor neuron denervation). Weight loss before and after diagnosis is associated with poorer outcomes. Increasing frailty also increases the risks associated with the procedure to place a gastrostomy.
Mealtime burden	Weakening upper limb strength makes it harder for people with MND to be independent with feeding themselves and increasingly reliant on help from others to prepare and eat meals. Weakening upper limb and handgrip strength can lead to prolonged and stressful mealtime experience for people with MND and caregivers, which can be compounded by the impact of dysphagia.
Respiratory failure	Respiratory failure and being on ventilation can make it harder for people with MND to eat and drink, particularly as the number of hours per day ventilation (and the wearing of a mask) increases. Respiratory failure also increases the risk associated with the procedure to place a gastrostomy.

In common with any clinical intervention, people with MND have a legal right to choose how they wish to manage any given symptom or consequence of MND, including the option to continue without any intervention (BMA, 2024). Therefore, during decision-making people with MND should understand the benefits, risks and outcomes of choosing to continue without or declining a gastrostomy, and weigh these up against those of having a gastrostomy and starting enteral nutrition. Each option will be associated with its' own set of health, quality of life, experiential and care burden outcomes which each individual person with MND will have their own views on. Summarised below is the evidence for the common indications identified in the literature as reasons to consider gastrostomy placement: dysphagia, malnutrition, mealtime burden and respiratory failure, and the outcomes associated with them.

Dysphagia affects the majority of people with MND at some point in the disease course (Vesey 2017). Dysphagia presents a risk of aspirating food and fluids into the lungs and of choking, both of which are associated with increased morbidity and mortality (Sorenson *et al.,* 2007). Initially, people with MND experiencing dysphagia take compensatory action by modifying the texture of their diet and fluids to make them easier and safer to chew and swallow, adjusting head positioning when swallowing and reducing distractions (Lisiecka *et al.,* 2021b). However, as the swallowing muscles become increasingly weak, the risk of aspiration or choking can reach critical levels. In addition to the risk of aspiration, dysphagia is associated with reduced enjoyment of eating which is also a strong indicator of people with MNDs' decisions to accept gastrostomy (Johnson *et al.,* 2012). The presence of dysphagia has a significant impact on caregivers, increasing the burden of carer associated with meal preparation, assisting with eating and anxiety about choking (Lisiecka *et* al., 2022; Rangira *et al.,* 2022). Assessing the prevalence of dysphagia is challenging as it depends on the disease phenotype, site of onset, measure or definition used, and stage of disease. A retrospective cohort study of a large trials database found that for those with no dysphagia at trial enrolment the incidence of dysphagia after 1 year was 44% or 85% and after 2 years was 64% or 92%, for those with spinal or bulbar onset respectively (Perry *et al.,* 2021). Similarly, people with MND with fast progressing or bulbar onset disease at diagnosis were found to be at significantly higher risk of developing dysphagia and of being indicated for gastrostomy (Mariani *et al.,* 2022). More severe dysphagia has been found to be correlated with weight loss and respiratory function (other indications for gastrostomy) and with changes in functional status (Shijo *et al.,* 2023). Despite the challenges estimating the prevalence of dysphagia in MND, it is clear that it is a symptom that most people with MND will experience particularly those with bulbar onset disease, that once presented is likely to progress and is the primary clinical indication for healthcare professionals and people with MND to consider gastrostomy placement (Stavroulakis *et al.,* 2013; Van Eenennaam *et al.,* 2021). In addition to the aspiration and malnutrition risks presented by dysphagia, swallowing problems also disrupt the lives of people with MND and their caregivers (Lisiecka *et al.,* 2021b). A qualitative interview study of people with MNDs' experience of dysphagia described the adaptions people with MND put in place to compensate for any challenges with swallowing experienced (Lisiecka *et al.,* 2021b). Progression of dysphagia was reported to be associated with increased fear of choking, reduced enjoyment of eating and signalled disease progression. When evaluating the outcome of the option to continue without a gastrostomy, it is important that people with MND consider the health, quality of life and experiential outcomes of progressive dysphagia and how these may compare to those associated with life on enteral nutrition.

Mealtimes can become prolonged, burdensome and stressful for people with MND as a result of dysphagia and weakening upper limb strength. Weakening hand grip affects people with MNDs' ability to prepare meals, hold cutlery or cups and can prevent people with MND from being able to lift food or drink to the mouth. Therefore, the presentation and progressions of upper limb weakness presents a significant barrier to being able to eat and drink enough, increases their independence on others and impacts on people with MNDs' experience of mealtimes. Caregivers can need to become increasingly involved in purchasing and preparing food, and eventually can be required to assist people with MND at mealtimes. Prolonged and burdensome meals have been identified as a major indication for offering gastrostomy placement to people with MND in surveys of healthcare professionals (Stavroulakis *et al.,* 2013; Van Eenennaam *et al.,* 2021). There is a paucity of research that has explored the association of mealtime length or burden on malnutrition risk and with the decisions of people with MND about gastrostomy, and a lack of outcome measures used in practice that could be used to inform how mealtime burden could inform timing of the intervention.

Weight loss in MND is multifactorial with causes including the inevitable loss of muscle mass because of motor neuron denervation, immobility and malnutrition (Genton *et al.,* 2011). Reduced nutritional intake due to dysphagia, weakening hand and arm strength and other physical and psychosocial factors can result in an energy deficit placing people with MND at risk of fat and fat-free body weight losses i.e. caused by malnutrition. Currently there is a lack of sensitive biomarkers that are feasible for use in routine practice and able to distinguish between disease and nutrition related losses of body mass (Ludolph *et al.,* 2023). The lack of measures to accurately monitor nutritional status and identify energy deficit remains a barrier to understanding how nutritional status is affected by the disease and the impact of any interventions as described below:

> '…the proportion of loss related to malnutrition versus disease or immobilisation is unclear' (Genton *et al.,* 2011).

A reduction in calorie intake for any reason places people with MND at risk of malnutrition. Similar to dysphagia, there are challenges with estimating the prevalence of malnutrition in MND, associated with the amount of symptom burden, stage of disease, different definitions of malnutrition and the range of measures of nutritional status used (Genton *et al.,* 2011). Malnutrition can be defined in terms changes in total weight or specific body mass compartments (e.g. fat or fat-free body mass), cross-sectional assessment of weight (e.g. BMI), comparisons of intake against estimated requirements, or patient reported measures (e.g. subjective global assessment). Weight loss or a lower weight in MND is an independent predictor of reduced survival. A systematic review including 9991 people with MND from 17 studies found that each additional gain of one BMI point was associated with significantly longer survival (HR 0.95, 95%CI 0.93–0.97; $p < 0.001$) (Ning *et al.,* 2019). Similarly, a French study found 30% increased risk of death for every 5% loss of weight at diagnosis (compared to usual weight), and 34% increased risk of death for every 5% weight lost after diagnosis (Marin *et al.,* 2011). In another study, weight loss at diagnosis was an independent prognostic indicator, with the risk of death increasing by 23% for every 10% weight lost since symptom onset (Janse Van Mantgem *et al.,* 2020). Increased weight loss prior to gastrostomy has also been associated with poorer outcomes. Post-hoc analysis of data from a large prospective cohort study of 330 people with MND found that losing greater than 10% weight from diagnosis was associated with significantly poorer survival (ProGas study, 2015). The negative nutritional and prognostic outcomes associated with weight loss have informed recommendations for earlier gastrostomy placement in people with MND, ideally prior to losing more than 5% weight since diagnosis (ProGAS study, 2015;

NICE 2019). The increasing signals in the literature that malnutrition is an independent prognostic indicator have made it a target for nutrition support intervention evaluation and development. The post-hoc analysis of an RCT including 201 people with MND found significantly improved survival and maintenance of weight in people with MND with fast progressing disease, taking high calorie supplementation (Ludolph *et al.,* 2020). Currently a large UK RCT is evaluating the effectiveness of a complex nutritional intervention to slow disease progression (NIHR [RP-PG-1016-20006], 2018). Nutrition support interventions are routinely used in practice including dietary enrichment and oral nutritional supplements (NICE, 2017). Gastrostomy placement is another nutritional intervention that in theory should support people with MND to meet their nutritional requirements to prevent or resolve malnutrition and help manage the aspiration or choking risks associated with dysphagia (Stavroulakis *et al*., 2013).

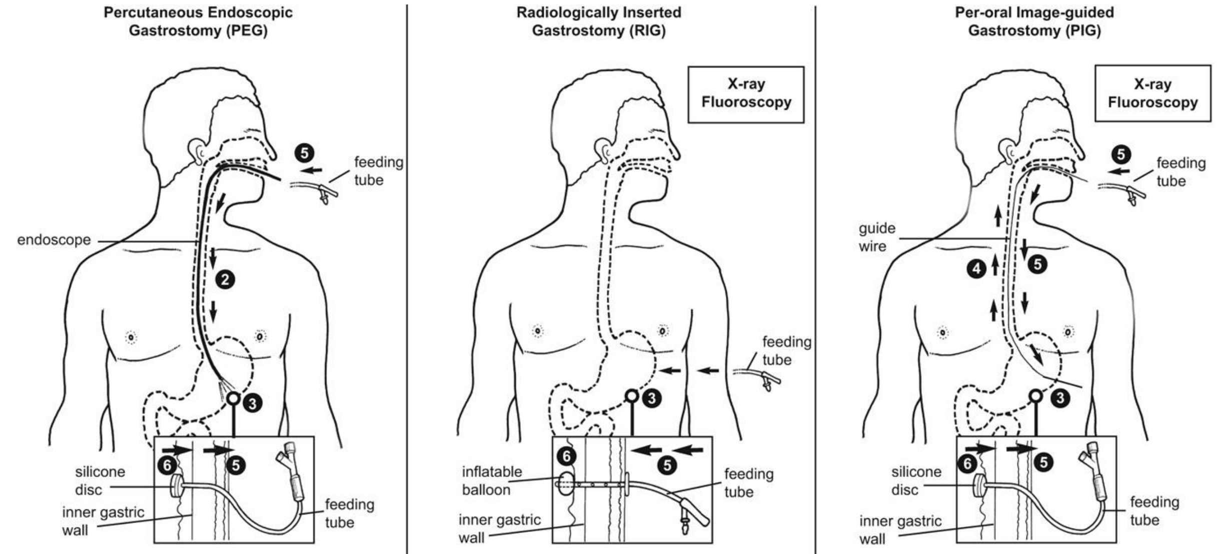
Respiratory failure is a common feature of MND and the main cause of death in people with MND as a result of the loss of motor neurons innervating the diaphragm (Corcia *et al.,* 2008; Hardiman 2011; Burkhardt *et al.,* 2017). People with MND are routinely offered NIV when their respiratory function can no longer clear the high blood levels of carbon dioxide that are associated with poor sleep, fatigue and ultimately death (Dorst and Ludolph, 2019). NIV which involves people with MND wearing a mask and receiving mechanical assistance to inflate and deflate the lungs, is one of the few interventions proven to extend survival in MND. An RCT including 41 people with MND found that survival was improved by 205 days in people with MND without bulbar symptoms (Bourke *et al.,* 2006). More recently, a retrospective study found a survival benefit of 15.5 months in all people with MND and 10 months in those with severe bulbar symptoms compared to a control group of people with MND who had declined NIV (Sancho *et al.,* 2018). The presentation and progression of respiratory failure is highly relevant to the decisions of people with MND about gastrostomy placement in relation to the procedural risks it infers (Kasarskis *et al.,* 1999; Miller *et al.,* 2009a). Therefore, decisions about gastrostomy placement are not just informed by the risks of aspiration and malnutrition, but also by the increasing procedural risks associated with respiratory failure. It is important for people with MND and healthcare professionals to understand the outcomes associated with the decline in all the clinical factors described above when considering gastrostomy placement. Together these represent the outcomes of continuing without a gastrostomy, and should be compared, during decision-making, with the predicted outcomes of agreeing to gastrostomy placement for administering enteral nutrition.

2.5 Gastrostomy placement in MND care

The procedure

Gastrostomy placement is routinely offered in MND care primarily in response to the risks associated with aspiration and malnutrition (Miller *et* al., 2009a; NICE, 2019; Shoesmith *et al*., 2020). The prevalence of gastrostomy placement in MND cohorts varies widely around the world between 7-69% (Sulistyo *et al.,* 2023). Figures reported from a national home enteral nutrition registry in 2015 found that MND was the main degenerative neurological condition indication for home enteral nutrition (HEN) in the UK (BANS, 2018). Of the total number of new registrants on the database, 6% (194/3216) had a diagnosis of MND; an increase from 2% (60/3430) in 2010 (BANS 2011; BANS, 2018). The prevalence of gastrostomy placement has also been shown to vary between MND centres in the same country (Mitsumoto *et al.,* 2003). The variance could be explained by differing practices, availability of expertise, cultures, criteria used to indicate a need for enteral nutrition and methodology used to assess prevalence. Gastrostomy placement involves the surgical placement, under conscious sedation, of a narrow plastic tube (~0.5cm in diameter) into the stomach enabling the administration of nutrition, hydration and medications when people with MND are unable to safely take adequate amounts orally (Stavroulakis *et al.,* 2013; NICE, 2019). There are several methods by which a gastrostomy can be placed with endoscopic and radiological methods being most common, each of which have a similar 30-day mortality associated with them of 3-7% (figure 1) (Stavroulakis *et al.,* 2013; ProGas study, 2015). The endoscopic method (i.e. percutaneous endoscopic gastrostomy (PEG)) includes the passing of an endoscope via the mouth and oesophagus and pull through of the gastrostomy out of an incision made in the abdominal wall. Alternatively, the radiological method (i.e. radiological inserted gastrostomy (RIG) or percutaneous image guided gastrostomy (PIG)) involves the placement of a gastrostomy under x-ray guidance, with or without the use of sutures to secure the stomach to the abdominal wall.

Figure 2 Comparative illustration of the main gastrostomy insertion methods in patients with MND (PEG vs. RIG vs. PIG). Stavroulakis et al (2013) Gastrostomy use in motor neurone disease (MND): a review, meta-analysis and survey of current practice, Figure 1, pp. 98. Figure reused with permission from Taylor and Francis.



Percutaneous Endoscopic Gastrostomy (PEG)

1. Conscious sedation (not shown in picture)
2. Endoscope insertion and stomach inflation
3. Transillumination with endoscope to locate puncture site
4. Incision, guide wire insertion (not shown in picture) through puncture site into stomach, up the oesophagus, exiting mouth
5. Feeding tube passed over the guide wire through the mouth, oesophagus, into stomach and pulled through the abdominal wall incision
6. Secure fixation with robust silicone disc
7. Endoscopic check

Advantages: Large bore tube placement, secure fixation, stiches not needed
Disadvantages: Endoscope use, conscious sedation, NIV use not possible during the procedure

Radiologically Inserted Gastrostomy (RIG)

1. Conscious sedation not necessary (not shown in picture)
2. Stomach inflation with NGT(not shown in picture)
3. Location of puncture site under fluoroscopic guidance
4. Stomach stitched to the abdominal wall (gastropexy), guide wire pushed into stomach from outside, track enlargement through a series of dilators (not shown in picture)
5. Feeding tube pushed into stomach from outside through enlarged track
6. Feeding tube fixation with inflatable balloon
7. Fluoroscopic check with contrast medium injection

Advantages: No endoscope use, NIV use possible, conscious sedation not required
Disadvantages: Small bore tube placement, stitches needed

Per-oral Image-guided Gastrostomy (PIG)

1. Minimal conscious sedation (not shown in picture)
2. Stomach inflation with NGT(not shown in picture)
3. Location of puncture site under fluoroscopic guidance
4. Guide wire insertion through puncture site into the stomach, up the oesophagus, exiting mouth
5. Feeding tube passed over the guide wire through the mouth, oesophagus, into stomach and pulled through the abdominal wall incision
6. Secure fixation with robust silicone disc
7. Fluoroscopic check with contrast medium injection

Advantages: Minimal sedation, no endoscope use, NIV use possible, large bore tubes, secure fixation, stitches not needed
Disadvantages: More complex procedure

Gastrostomy placement is a procedure that takes place in the hospital setting and can be completed as a day-case or requires a short hospital admission. The choice of placement method is often influenced by the expertise available within each MND centre and the clinical condition of the person with MND, particularly their respiratory status (Stavroulakis *et al.,* 2013; Benstead *et al.,* 2016; Marsden *et al.,* 2016). As with any surgical procedure there are morbidity and mortality risks to be aware of and manage, some of which are particularly relevant to people with MND (Blomberg *et al.,* 2012). In common with all gastrostomy placement procedures, there are risks during or immediately after the procedure of infection, bleeding, pain and death (Blomberg *et al.,* 2012; Zhang 2012; ProGas Study, 2015). A 30-day survival post gastrostomy placement of 5.31% (PEG) and 6.00% (RIG) was reported in a recent systematic review including 603 people with MND from 7 studies (Castanheira *et al.,* 2021). These mortality rates are similar to those reported for patients having a gastrostomy for any clinical indication with a 30-day survival post gastrostomy placement of 5.5% (PEG) and 10.5% (RIG) (Lim *et al.,* 2016). Following placement, people with MND can experience further complications associated with living with a gastrostomy tube including infection, leakage, granulomas and blocked, damaged or displaced tubes (ProGAS study, 2015). These complications are not unique to the MND indication for gastrostomy.

The respiratory failure commonly experienced by people with MND presents an added procedural risk factor to consider. The increased risks associated with placing gastrostomy tubes in people with MND in respiratory failure (Kasarskis *et al.,* 1999) has informed professional guidance recommendations for placing gastrostomy prior to reaching more significant levels of respiratory failure (i.e. before FVC<50%) (Miller *et al.,* 2009a). However, the same guidance makes the point that a low FVC is not necessarily a contraindication for the procedure. More recent research has confirmed that gastrostomy tubes can be placed without a significant increase in complications in people with MND in significant respiratory failure if the required precautions are in place (van Neerven *et al.,* 2024). Procedural precautions or adaptions help manage the risks include using experienced interventionists, respiratory or anaesthetic support, NIV use and selecting radiological rather than endoscopic methods (Stavroulakis *et al.,* 2013).

Outcomes of enteral nutrition

It remains unclear what impact enteral nutrition placement has on quality of life and survival in people with MND, with studies presenting mixed results (Castanheira *et al.,* 2021). Even though identified as a gap in the literature by published guidance, there is a paucity of controlled trails evaluating the effectiveness of enteral nutrition or the timing of the gastrostomy placement (Burgos *et al.,* 2018). This is not surprising with enteral nutrition being routinely used in practice worldwide and there being significant ethical considerations in terms of recruiting to a suitable control group. Attempts at evaluating the effectiveness of enteral nutrition have included recruiting people with MND who decline gastrostomy as a control group (Mazzini *et* al., 1995; Son *et al*., 2024). However, these studies are at high risk of selection bias with those declining gastrostomy likely to have different characteristics to those in the intervention arm. Therefore, retrospective and prospective observational cohort studies are the most frequently published research evaluating gastrostomy outcomes which are themselves associated with confounding factors and significant sampling or information bias (Grimes and Schulz, 2002; Son *et al*., 2024).

A controlled study from 30 years ago found that people with MND with gastrostomy experienced reduced survival after 6 months and improved weight gain compared to a group of people with MND refusing the intervention (Mazzini *et al.,* 1995). However, the trial only included 30 people with MND and there were differences in the groups disease severity and nutritional status at baseline that could have biased the results. A prospective multicentre study evaluated the outcomes of 89 people with MND post gastrostomy placement (Dorst *et al.,* 2015). Gastrostomy placement was associated with weight stabilisation and a mean 19-month survival including longer survival in those living longer than 12 months who were taking >1500kcals/day (23.6 v 21.1 months). A prospective study found no difference in survival when comparing people with MND who accepted or refused gastrostomy placement, where the intervention was deemed clinically indicated (Vergonjeanne *et al.,* 2020). However, a meta-analysis of 1578 people with MND found a significant increase in survival associated with gastrostomy placement (Bond *et al.,* 2019). While these findings are of interest, they provide limited information that could guide the decisions of people with MND about if or when to have a gastrostomy.

The timing of gastrostomy appears to be associated with a prognostic benefit, with later placement, in terms of progression or the disease or specific indications, being associated with shorter survival (ProGAS study, 2015; Castanheira *et al.,* 2021). More significant respiratory failure and weight loss at the time of gastrostomy placement have been associated with reduced survival. A retrospective cohort study found people with MND with predicted FVC>60% had significantly improved survival ($P<0.05$) (Bond *et al.,* 2019). However, improved survival could be influenced more by disease progression than the placement of a gastrostomy, with respiratory failure often being the cause of death in (Burkhardt *et al.,* 2017). In relation to nutritional outcomes, people with MND who had lost more than 10% weight at the time of gastrostomy placement, had significantly reduced survival, further fuelling the hypothesis that earlier gastrostomy placement could improve outcomes (ProGas study, 2015). Therefore, the timing of gastrostomy placement, in relation to stage of disease, respiratory function and nutritional status influences the clinical and survival outcomes of the intervention, informing the professional guidance recommending earlier placement before significant nutritional or respiratory decline (Miller *et al.,* 2009a; Burgos *et al.,* 2018; NICE, 2019; Shoesmith *et al.,* 2020). Communicating risk information relating to interventions is an important part of informed decision making (Thornton, 2003). In relation to the decision to have a gastrostomy, this involves understanding the risks associated with each indication and of placement in the present but also how these risks may change with time if the intervention is delayed. The lack of conclusive evidence about the clinical and quality of life outcomes associated with enteral nutrition in relation to clinical markers contributes to uncertainty and presents a challenge to healthcare professionals and people with MND seeking to make informed decisions.

An interesting attempt at evaluating the timing of gastrostomy placement was referred to in a qualitative paper (Pols and Limburg, 2016) but has not since been published (author contacted but did not receive a reply). Dutch researchers had aimed to compare the outcomes of people with MND randomised to have the timing of gastrostomy placement decided by their clinicians, with a control group who were allowed to make their own decision (i.e. usual practice). However, the study was reported to have failed because they were unable to recruit, with people with MND describing not being ready to accept the intervention despite the clinical recommendation. While the failure of the study must have been upsetting for the researchers, the interesting finding here was that people with MND were not willing to give up control for their decisions to their healthcare professionals. The concept of control appears frequently in the qualitative literature and points to the fact that decisions are based on more than the clinical indications for gastrostomy or the preferences of healthcare professionals.

A further factor to consider in relation to the optimal timing of gastrostomy is the time it takes people with MND to make a decision and then the time it takes for healthcare services to arrange tube placement following a decision having been made. An Australian cohort study found that people with MND took a median of 6 days (range 0-57 days) to make a decision following a clinical recommendation to have a gastrostomy and then waited a further median of 52 days (range 18-163 days) for gastrostomy placement after being referred (Labra *et al.,* 2020). In a French study, gastrostomy was placed a mean of 2.7 months (range: 0.9-5.8) after the intervention was indicated and recommended by the MDT (indication defined as reduced intake, >10% weight loss, mealtimes taking >45 minutes and risk of aspiration) (Vergonjeanne *et* al., 2020). Similarly, a recent cohort study in South Korea identified a delay of 8 months from gastrostomy being recommended to it being placed and during this time people with MND experiencing an average weight loss of 13.7% and accelerated decline in functional scores (i.e. 1.7 ALSFRS-R points per month) (Son *et al.,* 2024). Therefore, when considering the clinical need for gastrostomy there is a need to factor into decision-making how long it takes for people with MND to make decisions, how responsive local services are in placing gastrostomy tubes and the potential for continued disease decline during this time.

The lived experience of being on home enteral nutrition

In addition to the clinical or nutritional outcomes of gastrostomy placement and enteral nutrition in MND, the interventions are also associated with experiential outcomes that are relevant to the deliberations of people with MND about the option. A recent meta-ethnography aimed at understanding people with MNDs' experience of life on enteral nutrition identified less in-depth exploration of the post-gastrostomy experience in the qualitative literature than that of the pre-gastrostomy decision-making stage (Lisiecka *et al.,* 2021a). People with MND on enteral nutrition and their caregivers experience a range of clinical, practical, logistic and psychological challenges (Stavroulakis *et al.,* 2016; Lisiecka *et al.,* 2021a; Lisiecka *et al.,* 2022). However, many people with MND were content with their decision to have a gastrostomy placed, reporting benefits such as reduced anxiety at mealtimes, weight maintenance and a perception of improved survival that outweighed the negative consequences the intervention (Stavroulakis *et al.,* 2016). When responding to a post-gastrostomy survey, despite reporting inconveniences and complications, no people with MND wished for their gastrostomy to be removed after 1 year, with most reporting an improved quality of life and acknowledged the benefits once experiencing the consequences of dysphagia (Chhetri *et al.,* 2017). Similarly, a further qualitative study described how the perceptions of people with MND of living with a gastrostomy shifted *'from Symbol of Deterioration to Eraser of Complaints and Concerns'* in response to the progression of their disease (Pols and Limburg 2016). These studies capture that although people with MND face many challenges associated with the disease, gastrostomy placement and enteral nutrition the intervention can, with time, be viewed positively in comparison to the alternative of continuing without. With people with MND becoming increasingly dependent on others for care, enteral nutrition is another task that caregivers can be expected to take on. Though not specifically focused on MND, a previous meta-ethnography described the challenges associated with caring for patients on HEN including feeling overwhelmed, lacking choice and a disruption of their home lives (Lisiecka *et al.,* 2022). The importance of coordinated professional support is highlighted as essential for supporting their ability to care for patients on enteral nutrition (Stavroulakis *et al.,* 2016; Lisiecka *et al.,* 2022).

2.6 Decision making in healthcare settings

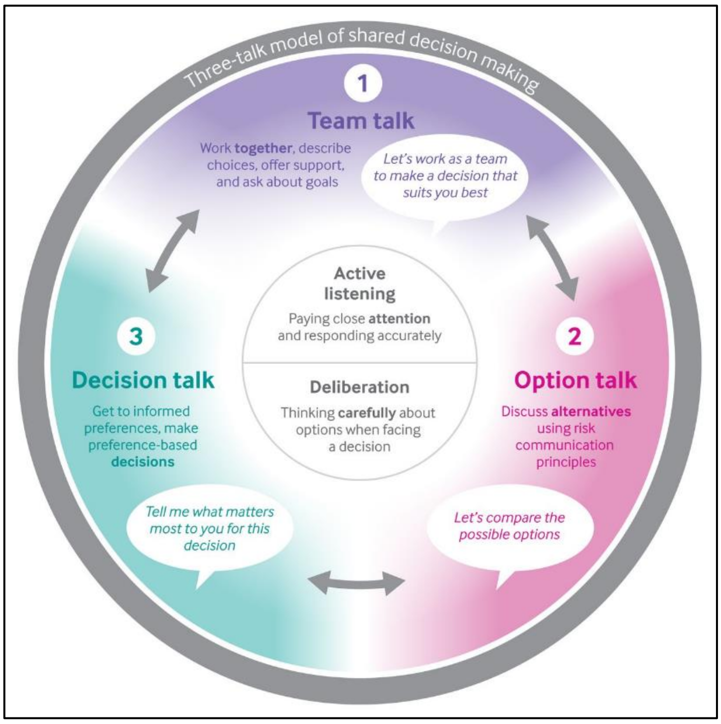
A legal framework exists that guides how people in the UK should be supported to make informed decisions about any treatment options by their healthcare professionals (MCA, 2005). The Mental Capacity Act (MCA) is an important piece of legislation mandating that people with the mental capacity are supported to make their own decisions, including in healthcare protecting their right to refuse treatments (MCA, 2005). Much of the professional guidance relating to treatments or intervention used in healthcare are underpinned by the principles of the Mental Capacity Act (BMA, 2024). The British Medical Association (BMA, 2024) consent guidance recommends that doctors share enough information with patients to enable them to understand their options and make informed decisions, including the option to continue without treatment. While the BMA guidance describes doctors' responsibilities when seeking informed consent, it is important to acknowledge that many healthcare professional disciplines are involved in presenting, discussing and deciding about treatment options, not just medical professionals.

While the MCA and associated guidance details healthcare professionals' legal and professional requirements, there is also a significant evidence base in relation to the process by which patients should be supported to make informed decisions that are consistent with their values and preferences. A collaborative process of deliberation between healthcare professionals and patients is commonly accepted as the gold standard approach to reaching decisions that are consistent with patients' values; a concept referred to as 'shared decision making' (SDM) (Coulter and Collin, 2011). Essential components of SDM identified through a review of publications on the topic included defining the problem, presenting options, discussing risks and benefits, clarifying patients' values and preferences, discussing patients' ability and self-efficacy, doctors sharing knowledge and recommendations, assessing understanding, explicitly making or deferring decision and arranging review (Makoul and Clayman, 2006). A professional recommendation to adhere to SDM is observed in most of the guidelines developed by NICE in the UK and is now the main focus of its own NICE guideline, making it a requirement of healthcare practice in the UK (NICE, 2021).

Shared Decision Making models

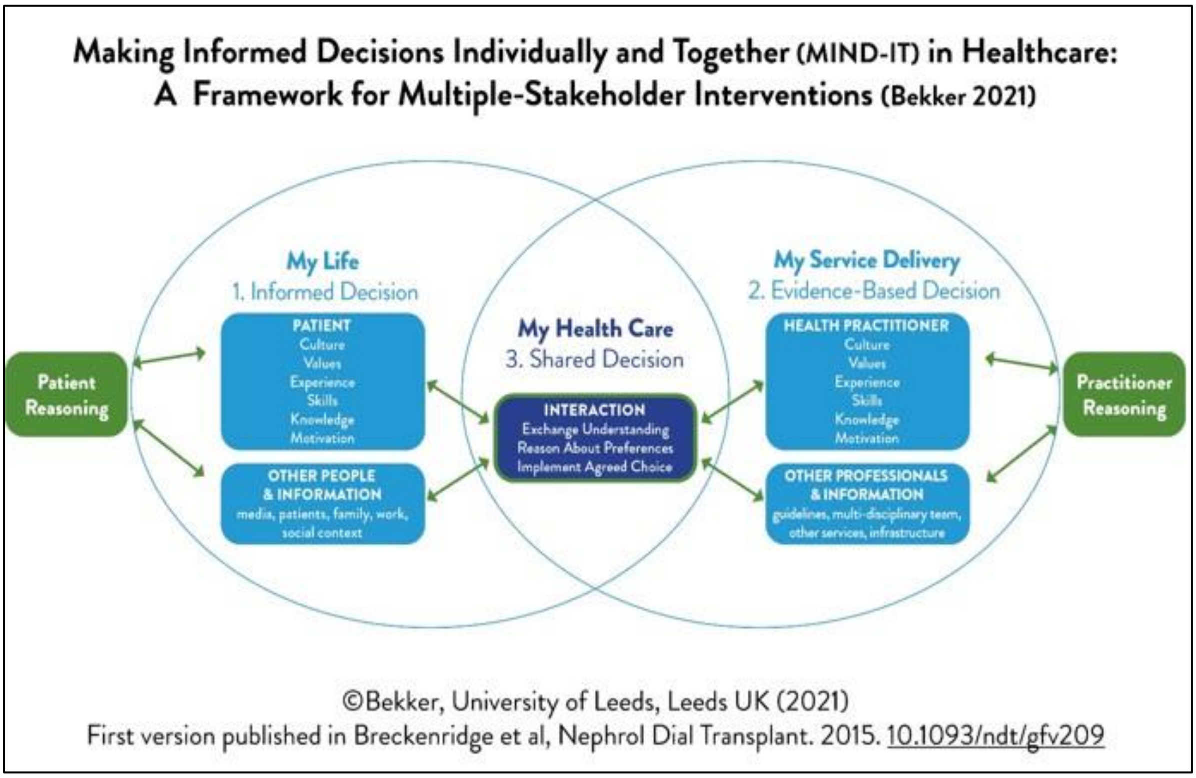
A variety of SDM models have been developed to help guide practice about the process of SDM and inform the development of decision support interventions. The frequently cited three talk model (figure 3), describes a cyclical process of collaboration between healthcare professionals and patients to come to a shared understanding of the options available (Team talk), what the options involve and their outcomes (Option talk) and making decisions aligned with patients' preferences (Decision talk) (Elwyn *et al.,* 2017). This model simplifies the SDM process into three clear stages that healthcare professionals can use to structure and deliver the decision support to patients, encouraging a collaborative approach, discussion about all options and the use of advanced communication skills e.g. active listening. However, what is absent from the model is the cognitive processes, behaviours and reasoning of the individuals involved. There is also a lack of reference of the social, clinical, relational or organisational context within which SDM takes place. While such issues may be of less significance for some simpler healthcare decisions, in relation to complex decisions about gastrostomy in MND, they are of high importance and particular relevance.

Figure 3 Three talk model of shared decision making in Elwyn, G. *et al.,* (2017) A three-talk model for shared decision making: multistage consultation process, figure 3, p. 5, is licenced for reuse under CC BY-NC 4.0.



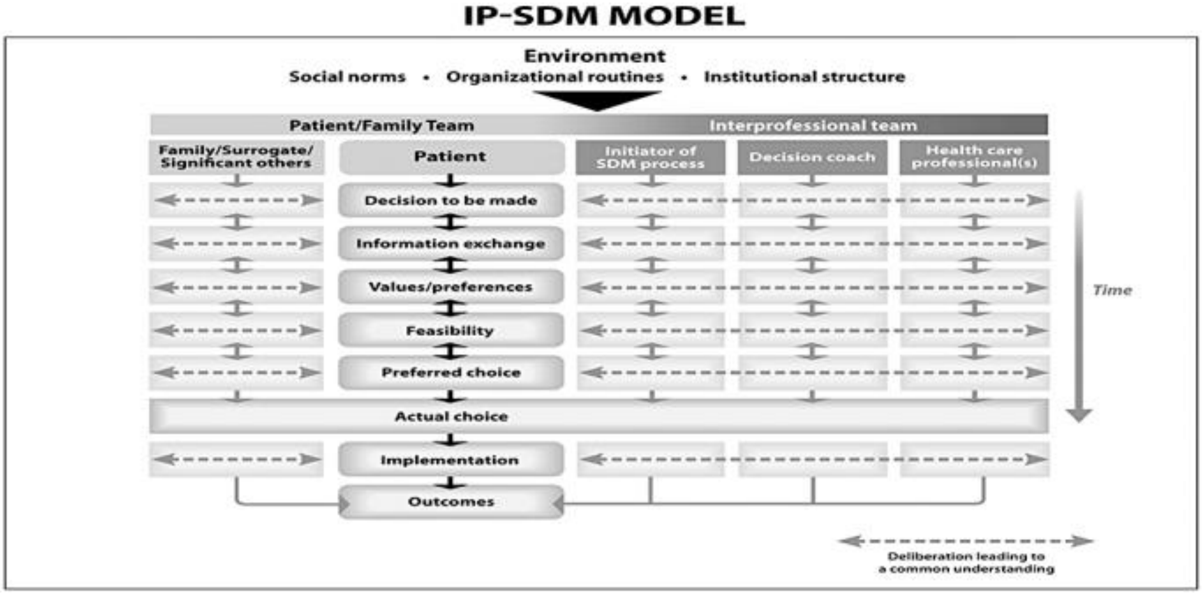
The Making Informed Decisions Individually and Together (MIND-IT) in Healthcare framework captures how patients and healthcare professionals enter the collaborative process of decision-making each with their own specific knowledge, experience, skills, motivations and values which they use to reason about their choices (see figure 4) (Breckenridge *et al.,* 2015; Bekker *et al.,* 2023). SDM require healthcare professionals and patients to share with each other their understanding and reasoning in relation to the decision problem to establish preferences for the options available. MIND-IT also explicitly acknowledges that these interactions do not occur in isolation and are influenced by the organisational, social and policy contexts in which they are taking place. The framework can be used to inform the development of interventions to promote reasoning and collaboration within complex clinical and social contexts (Bekker *et al.,* 2023). The MIND-IT framework has the potential to be adapted to decision-making in MND care, with decisions often being associated with uncertainty, multiple outcomes and a complex clinical and healthcare context.

Figure 4 Making Informed Decisions Individually and Together (MIND-IT) framework. Toft *et al.*, (2022) Measures used to assess interventions for increasing patient involvement in Danish healthcare setting: a rapid review, Figure 1, pp. 3. is licenced for reuse under CC BY-NC 4.0. (First published in Breckenridge *et al.*, 2015)



Decisions in healthcare are not always made as a dyad of just a single patient and a single healthcare professional. Often, several stakeholders are involved, including caregivers and multiple healthcare professionals; this is certainly the case for decisions about gastrostomy placement as will be discussed later. A theory analysis of three systematic reviews found that most SDM models failed to consider the involvement of other stakeholders, such as family members, the wider MDT and organisational factors (Stacey *et al.,* 2010). The interprofessional model of SDM (IP-SDM) (figure 5) integrates the principles of SDM taking place over time, the environment (i.e. context) in which decisions are being made and the involvement of the MDT (Légaré *et al.,* 2011a). Such models appear to be relevant to MND care where the MDT model is common, with many different healthcare professional disciplines operating in different settings initiating and continuing discussions the care options available and in view of the influential role caregivers play with regards the reasoning and choices of people with MND.

Figure 5 The inter-professional shared decision-making model (IP-SDM). Légaré *et al.,* 2011b Validating a conceptual model for an inter-professional approach to shared decision making: a mixed methods study, Figure 1, p. 557, is licenced under Creative Commons license.



Some of these models acknowledge that the decision-making process is bound within and influenced by the specific context in which it is taking place (Légaré *et al*., 2011b; Breckenridge *et al*., 2015). It is essential that these contextual factors are fully understood and accounted for, to support the implementation of decision-support interventions such as decision coaching or decision aids into routine practice. MND and MND care provision presents a particularly complex context within which decisions are made about care and treatment, including many challenges which could impact on the ability to meet the principles of good quality SDM.

Decision support interventions have been developed that involve patients in decision-making and better inform them about the problem, intervention and decision, weigh up the risks and benefits, check understanding, clarify values and facilitate the implementation of a preference-based decision. A recently updated Cochrane review confirmed the use of decisions aids supported patients to make values aligned decisions, improved knowledge and increased involvement in decision-making (Stacey *et al.,* 2024). Despite the recent development of a decision aid to support people with MND making decisions about gastrostomy placement in the UK and Australia (Hogden *et al.,* 2016; Maunsell *et al.,* 2019), previous research has confirmed such interventions are often poorly integrated into routine practice (Légaré *et al.,* 2018; Stacey *et al.,* 2019). A better understanding of how people with MND make decisions in real-world contexts could support the integration of interventions into practice that aim to promote engagement in, or improve the quality of, decision-making.

2.7 Motor neuron disease: 'the worst-case decision-making context' (Hogden 2014)

There is emerging qualitative literature that has sought to better understand the context of decision-making in MND care from the perspectives of people with MND, caregivers and healthcare professionals. Following diagnosis people with MND and their families have little time to psychologically and physically adapt to the decline in function prior to being asked to engage in decision-making activities (Greenaway *et al.,* 2015; Martin *et al.,* 2016). People with MND are asked to consider a range of choices about their care and treatment while experiencing a progressive loss of function, independence and control (Oliver *et al.,* 2011; Foley *et al.,* 2014c). Qualitative studies have described the inter-related clinical, psychosocial, organisational and relational factors influencing decision-making from the perspective of people with MND, their caregivers and healthcare professionals (Hogden *et al.,* 2012a; Hogden *et al.,* 2012b; Hogden *et al.,* 2013; Foley & Hynes 2018). Many of these factors can promote, interfere or form a barrier to people with MND understanding the consequences of the disease and the care or treatment options available to them. The rapid disease progression many people with MND experience make some decisions time-limited in terms of their effectiveness or feasibility. Therefore, there is a need to better understand the contextual factors and develop strategies or interventions that support people with MND to engage in early discussions, be fully informed about their choices, establish their reasoned preferences and facilitate timely decisions.

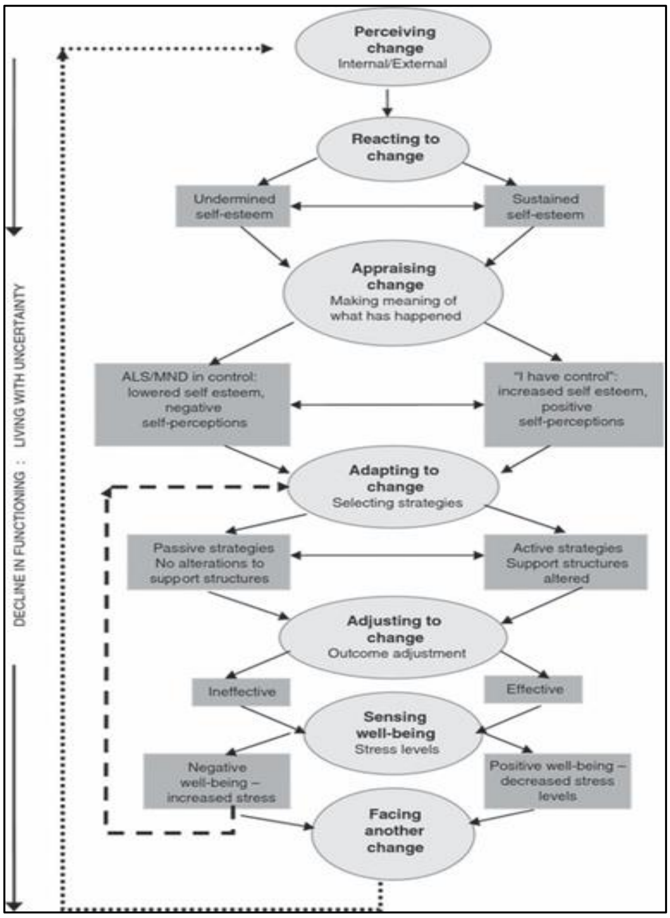
The reaction people with MND have to receiving the MND diagnosis influences their engagement in and response to, discussions about the disease course and treatment options. The route to being diagnosed can be stressful and associated with uncertainty about the cause of the symptoms presenting. It can take 10-16 months for people with MND to receive the diagnosis following the first onset of symptoms, with delays being associated with misdiagnosis, waiting for referrals, the lack of biomarkers and arranging clinical tests

(Richards *et al.,* 2020; Gwathmey *et al.,* 2023). Being given the diagnosis of MND and living with the consequences of the disease has been equated to being given a death sentence and leaves people with MND experiencing a loss of hope and control (King *et al.,* 2009; Foley *et al.,* 2014a). While often wanting to be well informed, people with MND can feel uncomfortable learning about how their condition will deteriorate in the future, and what this will mean for them and their caregivers (Hogden *et al.,* 2015). The response to the diagnosis and subsequent adaption to living with MND presents a particular challenge when it comes to supporting people with MND to understand the disease and the options they have to improve their quality or length of life (King *et al.,* 2009). Even once engaged in learning about the disease course, uncertainty about the rate of progression can lead to people with MND deferring discussions and decisions which has the potential to impact on outcomes if this results in later commencement of the intervention (Paynter *et al.,* 2020).

The psychological impact of MND and how people with MND respond and seek to cope with the disease influences the response to and perceptions of the care options offered to them throughout the disease course (Foley *et al.,* 2014a). Such discussions highlight the direction their disease will take and brings the attention of people with MND to a future associated with increased disability, dependence and an earlier death (Foley *et al.,* 2014a; Paynter *et al.,* 2020). A commonly reported strategy used by people with MND to cope with living with the disease is to focus on what they can control in the present and a preference to deal with future challenges as they are experienced (Foley *et al.,* 2014a). This presents a challenge to opening discussions about the future including establishing people with MNDs' preferences in advance of disease progression. Healthcare professionals have interpreted this avoidant behaviour as people with MND being in denial, actively choosing to defer such discussions to allow them cope with the distressing consequences of the disease (Hogden *et al.,* 2012a). The emotional readiness of people with MND to engage in discussions about interventions influences the timing of information exchange between healthcare professionals and people with MND (Hogden *et al.,* 2012b; Martin *et al.,* 2016). While avoiding discussions may be perceived by people with MND to be an effective coping strategy to reduce psychological distress, information seeking behaviour has been found to be associated with a better quality of life (Matuz *et al.,* 2010). Such avoidant behaviours (i.e. deferring discussions or decisions) have the potential to postpone the timing of when people with MND gain an understanding of the care or treatment options that may enable them to live better with MND and may delay the commencement of treatments which could impact on their health and quality of life outcomes (Hogden *et al.,* 2015). In relation to some decisions in MND care, including about gastrostomy, there is a powerful clinical rationale for supporting people with MND to understand their options prior to significant disease progression and for encouraging earlier intervention.

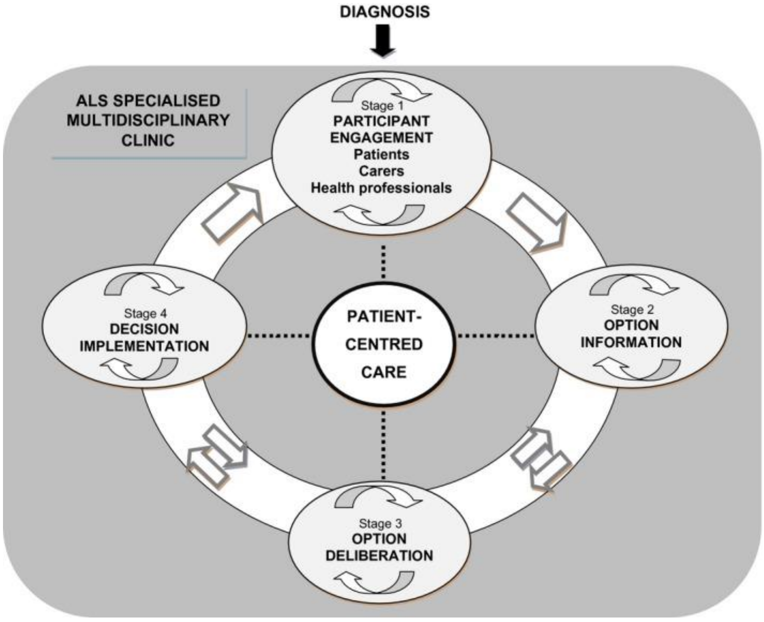
A number of conceptual models have captured a longitudinal, cyclical process through which people with MND perceive and respond to the disease and the options presented to them (King *et al.,* 2009). King *et al.* (2009) used a grounded theory study to develop the 'On-going change and adaptation model of decision making' (figure 6) mapping a process of adaption and acceptance onto disease progression and how each are associated with positive or negative responses and coping strategies that could influence decision-making. The adaptive responses people with MND have to disease progression were described as active or passive (including denial). The evolving acceptance and adaptation of people with MND to the progressive functional losses influences their perceptions of interventions and engagement in discussions and the decisions made (Albert *et al.,* 1999; King *et al.,* 2009; Foley *et al.,* 2014c). A population-based study found that people with MND with an active approach to interventions are more likely to make decisions about NIV or gastrostomy than those with passive attitudes (Martin *et al.,* 2014). This research highlights the importance of seeking to understand and address people with MNDs' emotional response and coping strategies to the diagnosis, experience of disease progression and having treatment options presented, when trying to engage in decision-making discussions.

Figure 6 On-going change and adaptation model of decision making. King *et al.,* (2009) Living with amyotrophic lateral sclerosis/motor neurone disease (ALS/MND): Decision-making about 'ongoing change and adaptation', Figure 1, pp. 748. Figure reused with permission from John Wiley and Sons.



A further cyclical model of decision-making in MND was developed following analysis of interviews with people with MND, caregivers, and healthcare professionals from two MND centres in Australia (Hogden *et al.,* 2015). The model is consistent with the models of SDM discussed in section 2.6 including the sharing of information, deliberation about options and the implementation of decisions (figure 7) (Légaré *et al.,* 2011b; Elwyn *et al.*, 2017). The importance of facilitating engagement in discussions and the fluid nature of deliberation when people with MND are considering their options is also captured. The model places MND decision making within the context of the MND clinic and therefore fails to recognise the role that HCPs operating within other settings play in supporting people with MND to make their decisions. There is a need for research to explore how people with MND are supported to make decisions as a result of interactions with all healthcare professionals in all settings in which they take place, not just those taking place in the MND clinic.

Figure 7. Model of patient-centred decision making in specialized ALS multidisciplinary care. Hogden *et al*., (2015) Development of a model to guide decision making in amyotrophic lateral sclerosis multidisciplinary care, Figure 1, pp. 1778. Figure reused with permission from John Wiley and Sons.



Control

Throughout the qualitative literature focusing on decision-making in MND care, control is a commonly reported concept proposed to influence how people with MND engage in discussions, deliberate about their options and the decisions they implement. A loss of control to the disease was reported to leave some people with MND feeling they lack a choice, perceiving interventions as being inevitable due to disease progression (Stavroulakis *et al.,* 2016; Paynter *et al.,* 2020). pwMND have described varied preferences for information ranging from wishing to be fully informed early in the disease course, through to only wanting to learn about interventions when they perceive there is a need (Greenaway *et al.,* 2015). A discrete choice experiment found that people with MND preferred to receive all information about MND at diagnosis, particularly those with less progressed disease (Tobin *et al.,* 2021). There is a period of time between being having an option presented and then needing the intervention, where people with MND reported they perceived they have a choice and before the disease is forcing them to make a decision (Paynter *et al.,* 2020). These findings support an argument for allowing people with MND to consider their options before their autonomy may be threatened by the pressure placed on them by disease burden. People with MND can differ in their preferences for maintaining control over their decisions and when to give some control to supporting healthcare professionals (Foley *et al.,* 2014a; Greenaway *et al.,* 2015). An individualised approach to decision-making is required that supports people with MND to be in control of their discussions, choices and healthcare services.

Role of others

Healthcare professionals and caregivers are an influential part of the context in which people with MND make decisions. While people with MND seek to take control of their decisions, they do not make decisions alone and can release responsibility, preferably on their own terms, for decision-making to supporting healthcare professionals and caregivers (Foley *et al.,* 2014a). Healthcare professionals play an integral role during the decision-making process in terms of being gatekeepers to interventions and the support they provide to people with MND to understand the disease and the options available. Healthcare professionals are a valued source of information, counselling, reassurance and guidance (Foley *et al.,* 2014a; Greenaway *et al.,* 2015). However, the trust in healthcare professionals can be threatened by a lack of coordinated care, poor communication skills or inadequate MND knowledge (Greenaway *et al.,* 2015). While people with MND prioritise the information they receive from healthcare professionals, they are also influenced by the approach taken by healthcare professionals towards having these difficult discussions (Paynter *et al.,* 2020). Decision-making is in a large part influenced by the dialogue between people with MND and healthcare professionals. Facilitators of effective decision-making discussions included healthcare professionals giving people with MND time to deliberate about their options, particularly those with speech difficulties or using communication aids, and using advanced communication skills (Paynter *et al.,* 2020). However, allowing time for these discussions can be challenging within the time constraints of busy MND clinics or in relation to the rate of disease progression (Paynter *et al.,* 2020).

Equally, caregivers play a varied role, not only during the decision-making process, but also through being a rationale for people with MNDs' choices because of the perceived burden the disease and/or the intervention will have on caregiver. Caregivers play a supportive role during decision-making, including screening and sourcing information; providing emotional support; supporting communication; and enabling people with MND access to expert advice (Hogden *et al.,* 2013; Paynter *et al.,* 2020). Interventions, including gastrostomy placement, inevitably have consequences on caregiver burden which influences how people with MND perceive the decisions they are making (Greenaway *et al.,* 2015; Foley and Hynes, 2018; Labra *et al.,* 2020; Castanheira *et al.,* 2021). Healthcare professionals and caregivers consistently acknowledge and support the rights of people with MND to make autonomous decisions (Hogden *et al.,* 2015). However, caregivers' involvement can present a threat to the autonomous decision-making of people with MND when there is a difference in preference for any given option (Foley and Hynes, 2018).

2.8 Making the decision to have a gastrostomy placed

The experience of people with MND and caregivers making decisions about gastrostomy

Guidance recommends healthcare professionals have early discussions about gastrostomy placement with people with MND and revisit the option as the disease progresses and indications present (Burgos *et al.,* 2018; NICE, 2019). However, psychosocial, existential and cognitive challenges have been reported that influence decisions people with MND make about gastrostomy (Stavroulakis *et al.,* 2014; Greenaway *et al.,* 2015; Martin *et al.,* 2016; Labra *et al.,* 2020). A recent meta-ethnography identified a wide range of factors that people with MND reported influenced the decisions they made about gastrostomy placement including interactions with other people; perceptions of choice and control; the influence of dysphagia on decisions made; the role of personal values; timing of gastrostomy placement; impact of emotions and feelings; and the place of gastrostomy within the wider context of MND (Lisiecka *et al.,* 2021a).

In an interview study people with MND and caregivers experienced uncertainty about disease progression, attached value to continuing oral diet, and reported a lack of understanding about the risks and benefits of the intervention when considering gastrostomy placement (Stavroulakis *et al.,* 2014). The idea of not being able to eat and drink is a major source of loss for people with MND and contributes to the negative representation of gastrostomy placement (Stavroulakis *et al.,* 2014; Van Eenennaam *et al.,* 2023). The control people with MND have reported to value in previous MND studies (Foley *et al.,* 2014a), have also been found to be relevant to the decision to have a gastrostomy. People with MND reported seeking control by delaying decisions about gastrostomy but then perceived a lack of choice when placed under pressure by increasing symptoms (Van Eenennaam *et al.,* 2023).

Many people with MND agree in principle to have a gastrostomy when it is needed (Van Eenennaam *et al.,* 2023), but they are then required to make a second decision about when they wish to place the tube. Healthcare professional recommendations for gastrostomy placement are often made in response to observing functional decline (Van Eenennaam *et al.,* 2021). In contrast, people with MND have been reported to be guided by how they are experiencing the disease, sometimes willing to tolerate symptom burden rather than start an intervention (Stavroulakis *et al.,* 2014; Greenaway *et al.,* 2015). People with MND often report not wishing to consider future disease progression or the prospect of being in a position when gastrostomy needs to be considered, preferring to delay decision-making (Pols and Limburg, 2016; Labra *et al.,* 2020; Van Eenennaam *et al.,* 2023). People with MND can be reluctant to accept the placement of gastrostomy before they perceive there is a physical need (i.e. dysphagia) for enteral nutrition (Paynter *et al.,* 2020). The symptomatic triggers that prompt people with MND to make the decision to have a gastrostomy include weight loss, burdensome mealtimes, and dysphagia (Stavroulakis *et al.,* 2014). An exploratory population-based study found that people with MND with more advanced disease at recruitment were more likely to make intervention decisions, including about gastrostomy (Martin *et al.,* 2014). This is consistent with previous research where lower scores of eating derived pleasure predicted acceptance of gastrostomy (Johnson *et al.,* 2012). The resistance to early discussions or decisions creates a challenging juxtaposition between healthcare professionals wishing to be proactive and people with MND wishing to delay until they perceive a need (Greenaway *et al.,* 2015). Many qualitative studies have explored the perceptions of people with MND with regards decision-making about gastrostomy after a decision has been made to accept or decline gastrostomy or after the tube had been placed (Stavroulakis *et al.,* 2014; Greenaway *et al.,* 2015; Martin *et al.,* 2016; Labra *et al.,* 2020; Van Eenennaam *et al.,* 2023). However, it is equally important to explore decision-making before and while making decisions to gain a better understanding about how people with MND experience the phenomenon contemporaneously.

The association of non-illness factors with decision-making has not only been identified in the qualitative literature. Higher intelligence, increased time in education, and an active attitude (i.e. actively seeking information) were associated with people with MND being more likely make decisions to accept or decline interventions, including gastrostomy (Martin *et al.,* 2014). These findings are consistent with the qualitative research presented above indicating that people with MND do not make the decision based on the clinical indications alone and other contextual factors are at play. The lack of clinical evidence to guide decisions about timing and the wider contextual factors has implications for what decisions are made and when. These need to be better understood if people with MND are going to be better supported to make informed and timely decisions.

Healthcare professional experience of supporting people with MND *to make decisions about gastrostomy*

Previous research has identified the range of issues that may influence healthcare professional clinical reasoning about discussing gastrostomy placement including markers of function or health state (Van Eenennaam *et al.,* 2021), perceptions about 'social factors, emotional coping, and acceptance' (Hogden *et al.,* 2012a; Martin *et al.,* 2016), and previous clinical experience (Greenaway *et al.,* 2015). Several healthcare professional surveys have provided an insight into practice and attitudes towards gastrostomy placement, including evidence of variability in their responses (Ruffell *et al*., 2012; Stavroulakis *et al*., 2013; Van Eenennaam *et al.,* 2021). A survey of 20 neurologists, acting as primary investigators for a UK multicentre trial of gastrostomy use in MND care, captured the range of indications used to guide when they offered gastrostomy to people with MND (Stavroulakis *et al.,* 2013). All neurologists responding would wait for the presentation of nutritional or swallow indication for gastrostomy. However, this survey did not explore 'when' the neurologists introduced the option to people with MND, the views of the wider MDT or the process of decision-making. An MDT survey revealed differences between medical and allied health professionals about the timing of gastrostomy discussions, policy compliance, and how informed they believed people with MND are about enteral nutrition (Ruffell *et al.,* 2012). In a Dutch cross-sectional survey, there was variation in what clinical cut-offs (e.g. % weight loss) physicians referred to and ‘when’ the option was introduced, with half preferring to discuss gastrostomy soon after diagnosis and others waiting for the presentation of clinical indicators (Van Eenennaam *et al.,* 2021). Healthcare professionals have reported taking into consideration the emotional readiness of people with MND prior to opening discussions about intervention options (Hogden *et al.,* 2012a). Further qualitative research is required to better understand healthcare professionals reasoning in relation to how, why and when they decide to present the option of gastrostomy to people with MND.

2.9 MDT decision support

As described in section 2.3, MND care is recommended to be delivered by MDTs experienced in MND care, with such care models being associated with improved survival and quality of life (Ng *et al.,* 2009; Aridegbe *et al.,* 2013; NICE, 2019). Good communication, evidence-based healthcare and adequate healthcare professional resource have been identified as attributes of an effective MND MDT (Hogden and Crook, 2017). Timely and effective decision support has the potential to be a mechanism by which MND teams infer a prognostic benefit, through facilitating earlier commencement of interventions in those people with MND who choose to proceed. Maybe more so than any other decision that people with MND are asked to make, discussions about the option to have a gastrostomy placed comes under the professional remit of many different healthcare professions (Ruffell *et al.,* 2012; Van Eenennaam *et al.,* 2021). MND physicians replying to a survey identified 12 different professions who have discussions with people with MND about gastrostomy placement including neurologists, specialist nurses, dietitians and speech and language therapists (Van Eenennaam *et al.,* 2021). The MDT nature of decision support reflects the variety of possible clinical indications for the gastrostomy which are of relevance to these professions (Berlowitz *et al.,* 2023). While each healthcare professional may have different experience, perspectives or knowledge that can help inform the decisions people with MND make about gastrostomy, the MDT is challenged to deliver consistent and coordinated decision support. The challenge is made all the more difficult in view of healthcare professionals, teams and services being separated in terms of their geography and organisation (OPM 2016). Previous qualitative studies have identified the MND MDT clinic as the ideal place for making decisions through the availability of healthcare professionals with expertise in MND and their ability to coordinate other services (Hogden *et al.,* 2015).

However, the healthcare professionals involved in supporting people with MND to make decisions about gastrostomy are often not part of the clinic MDT, with care having to be coordinated across settings (Hogden *et al.,* 2017). Issues such as care coordination are not comprehensively addressed in international MND management guidelines (Janssens *et al.,* 2016).

While people with MND prioritise and value the support they receive from healthcare professionals about the decision to have a gastrostomy, research has identified challenges within MDT care including poor communication between healthcare professionals and inconsistent information about the disease and interventions (Hogden *et al.,* 2012a; Hogden *et al.,* 2013; Hogden *et al.,* 2017; Chapman *et al.,* 2021). A lack of MND or intervention specific knowledge, poor communication between healthcare professionals, and a lack of manpower may explain why some people with MND and healthcare professionals experience inconsistent delivery of decision support across the MDT (Hogden *et al.,* 2012a; Hogden *et al.,* 2017). Healthcare professionals and people with MND have reported mixed messages being delivered about the need for interventions which has the potential to contribute to people with MND experiencing decisional conflict (Greenaway *et al.,* 2015; Chapman *et al.,* 2021). Differences in opinion were found in responses to a survey between UK medical and allied healthcare professionals when asked about the timing of gastrostomy discussions, policy compliance, and how informed people with MND and caregivers are about gastrostomy placement (Ruffell *et al.,* 2013). While Dutch physicians responding to a survey were consistent in relation to the indications and goals of gastrostomy placement, there was further evidence of varying healthcare professionals' beliefs about the timing of discussions with people with MND (Van Eenennaam *et al.,* 2021). Any variations in how healthcare professionals within a single MDT initiate, share or frame information about the gastrostomy decision has the potential to cause decisional conflict for people with MND they are supporting as a collective. The MND MDT has the complex task of delivering a consistent approach to decision support across healthcare professionals acting in different settings, teams and organisations, each of whom have their own priorities, values, experience affecting their reasoning. The MDT should act as a coordinated unit of decision support accounting for the many clinical and contextual factors known to influence how people with MND make decisions. Therefore, when studying decision-making about gastrostomy in MND care, enquiry should extend to exploring how decisions are made across people, settings and time.

2.10 Why is the proposed research needed?

In summary, the above review of the current literature conveys the following key messages:

- Nutritional status is an independent prognostic indicator for people with MND and nutrition support interventions have the potential to improve outcomes.
- Gastrostomy placement is one such nutrition support intervention that is recommended in professional guidance and routinely offered to people with MND.
- Healthcare professionals are recommended to have early discussions about gastrostomy with people with MND and to communicate the benefits of placement before significant functional or clinical decline.
- A process of shared decision making is recommended to enable patients to reach informed decisions aligned with their values.
- Interactions between people with MND, caregivers and healthcare professionals influence when, how and what decisions about gastrostomy are made.
- Findings from qualitative research signal the contextual complexities associated with making decisions about gastrostomy.
- Decision support is delivered by a range of healthcare professionals from different professions, teams, and organisations, and care is delivered in different settings.
- Attendance at a specialist MND MDT clinic is associated with improved outcomes.
- Little is known about how the MND MDT coordinates the decision support it delivers across healthcare professionals, settings, teams and organisations.

There have been several qualitative studies published exploring people with MND, caregivers and HCPs experience of making decisions in MND in general and in relation to gastrostomy specifically. A number of similar or related concepts (e.g. control, avoidance) have been identified across this evidence base in relation to the contextual factors influencing how and when decisions are made. There is now an opportunity to synthesise these findings to construct deeper understanding of this context and help prioritise the direction of future research and intervention development. Healthcare professional reasoning, experience and practice plays an important role in relation to when gastrostomy is offered, how people with MND understand their options and the timing of decisions. There remains a gap in the literature that has sought to understand how people with MND and healthcare professionals undertake the process of decision-making about gastrostomy in real-world practice. There is also a paucity of research that has explored how the healthcare professionals coordinate the decision support they deliver across the different settings in which they support people with MND.

2.11 Doctoral research question

The overarching research question for my doctoral research was:

> ‘How do MND MDTs support people with MND to make decisions about gastrostomy tube placement?’

2.12 Research questions for each component of the study

A sequential mixed-methods design was employed to answer the doctoral research question, in including three components: Qualitative evidence synthesis, HCP survey and multiple case study. The specific research questions for each of the three components of the study were:

- Qualitative evidence synthesis:
  > ‘What are the organisational, team and individual level factors that influence decision making by people with MND when making the decision to start an invasive healthcare intervention?’
- HCP survey:
  > ‘What are healthcare professionals’ practice and beliefs in relation to supporting people with MND making decisions about gastrostomy placement?’
- Multiple case study:
  > ‘How healthcare professionals, operating within MDTs, support people with MND to make decisions about gastrostomy placement?’

2.13 Overview of research plan

A three-component mixed-methods sequential study was undertaken:

1. Qualitative evidence synthesis focusing on people with MND, caregivers and healthcare professionals' experience of making decisions about invasive interventions.

A qualitative evidence synthesis identified the qualitative evidence exploring the factors that influence decision-making about invasive interventions, from the lived experience of people with MND, caregivers and supporting healthcare professionals.

2. A cross-sectional survey of UK healthcare professionals' practice and attitudes.

The analytical themes from the review informed the development of a cross-sectional survey exploring healthcare professional practice and attitudes towards supporting gastrostomy decision-making both as individuals and as MDTs.

3. Multiple case study exploring the real-world delivery of MDT decision support

Finally, a multiple case study approach, triangulating data collected from a people with MND, caregivers and healthcare professionals from within three cases, using a range of qualitative methods, provided a rich description of how MND MDTs facilitate decision making in real-world practice.

While the research conducted was focused on the UK MND care models, particularly the case studies which are designed to focus on the 'particular' of the cases, the findings are likely theoretically or analytically transferable to other MND settings both within the UK and internationally (Stake, 2006; Yin, 2018).

2.14 Summary of the chapter

The above chapter has reviewed the literature that is relevant to understanding and justifying the research questions. The key points include that MND is a progressive neurological disease associated with the loss of multiple functions, many of which represent a health threat for which gastrostomy placement is indicated to manage. The placement of a gastrostomy involves a hospital admission, a surgical procedure, followed by the care and use of the tube for enteral nutrition in the home setting. There is a paucity of evidence supporting the efficacy of enteral nutrition in MND, including uncertainty about the timing of gastrostomy placement. MDT care is recommended as best practice for people with MND and associated with improved outcomes. Many professional disciplines within the MND MDT can get involved in supporting people with MND decisions about gastrostomy. To date, there has not been any research focusing on how the MDT coordinate the decision support they deliver to people with MND considering gastrostomy. Interactional, clinical, psychological, nutritional and organisational factors contribute to a complex context within which decisions are being made and are in need of better understanding to guide the development of interventions to improve the quality of decision-making by people with MND. A three component, sequential mixed methods design has been used to answer the research question, which are described in more detail in the following chapter.

3. Methodology

3.1 Chapter outline

The following chapter provides further information about the methodology and methods used during the research conducted. Firstly, I justify the subtle realist position I have taken for this research including its compatibility with the mixed-methods approach. A summary of the explanatory sequential mixed-methods approach is described. I then provide justifications for and further detail about the methods used in the qualitative evidence synthesis, HCP survey and multiple case study. The information provided in this chapter adds to the information included in the methods section of Papers 1-4. For each component of research, a justification for including the method, detail about how the methods were implemented, and a summary of the ethical considerations associated with the method are described. Finally, I provide some background into how I have collaborated with several PPI groups and individuals with MND, and how these have informed the design of the research and implementation of the plan.

3.2 Philosophical position

The design of research should be guided by the researcher's philosophical position in terms how they define the reality of social entities being studied (i.e. ontological position) and what knowledge or understanding it is possible to generate about that social entity (i.e. epistemological position (Mason, 2018). Below I the position I took in relation to this research.

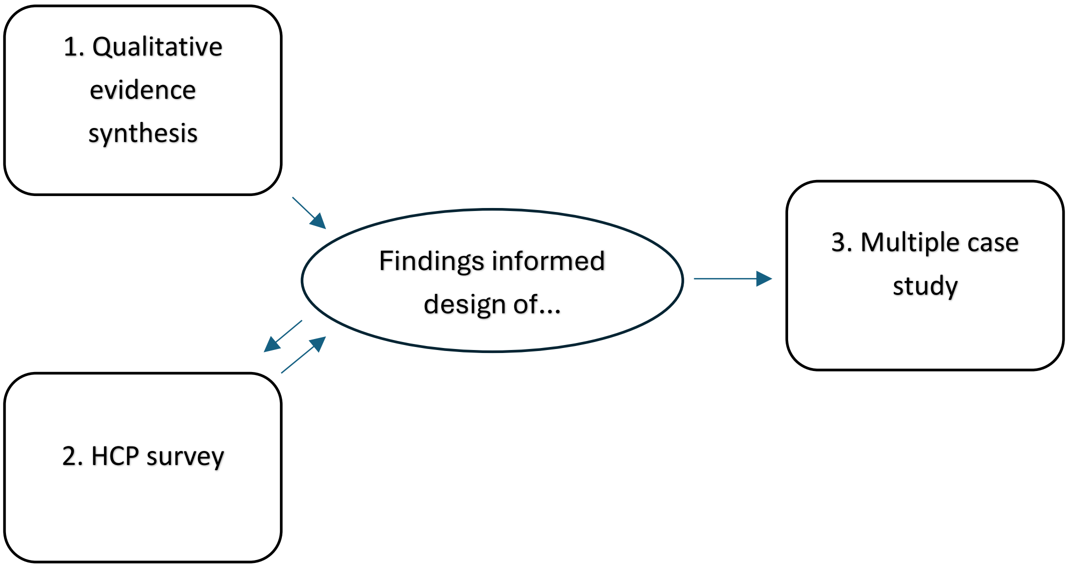
There are several positions that could guide research about this topic of interest. An objectivist position would view the process of decision-making a social reality independent of the people involved. This would be consistent with researchers with a positivist epistemology seeking to quantify the quality of decision-making or participants attitudes and practice to generate theory or laws that are available to future inquiry (Bryman 2018). Such an approach would provide objective evidence about the process of decision-making but would not generate knowledge about how the intrinsic action of decision-making of people with MND interacts with the context of the disease and the health service organisations within which decisions are made. I believe decisions about gastrostomy placement are socially constructed by people with MND through their interactions with caregivers, healthcare professionals and the external context (Robson and McCarton, 2016). This position is aligned more with a constructionist position dismissing the positivist view that contexts and people do not influence each other. Taking a constructionist or subtle realist position, the process of decision making is an external, independent reality about which knowledge can generated through accessing participants' experiences, perceptions and reasoning (Pope and Mayes, 2020). Hammersley (1992) argues for a subtle form of realism where researchers can have confidence in the credibility of any knowledge claims accepting that certainty about a social reality is not achievable. Academic inquiry can represent an independent social reality that exists external to the observations about it, but any given representation of the social reality remains that of the individual researcher and could differ from the representations of others (Hammersley 1992, p. 50-54; Pope and Mayes, 2020, pp. 17-18). Taking this position justifies the integration of multiple perspectives of the decision-making and the use of mixed methods to obtain an in-depth understanding about the phenomenon and context (Duncan and Nicol, 2004; Murphy *et al.,* 1998). I adopted a subtle realist position; in practice this requires reflexivity throughout the whole research process (May and Perry, 2017).

3.3 Mixed methods research

The components of this research study include quantitative (i.e. healthcare professional survey) and qualitative (i.e. qualitative evidence synthesis and multiple case study) methods. The two main justifications for using quantitative and qualitative methodology were completeness and enhancement (Bryman 2016, p. 556-561). First, in relation to completeness, the survey was included to generate knowledge about healthcare professionals' practice and beliefs in relation to how they support people with MND making decisions about gastrostomy. The survey findings would identify trends or differences within the healthcare professional population that would not be possible within the case study. The survey enhanced the project through identifying conceptual targets to be explored in depth within the case studies, through informing the case study objectives, sampling strategy, topic guides and the a priori analytical framework. For example, the survey finding that not all healthcare professionals believed they were able to initiate gastrostomy discussions as early as they would like was a concept explored in depth during the case studies, seeking to generate a contextual understanding for why this may be the case.

A combination of a fixed and emergent mixed methods design was used for these studies, following a predetermined research plan (fixed) but with the findings from the earlier research informing the design of the subsequent component (emergent) (Cresswell and Plano Clark, 2018). The study follows an explanatory sequential design, one of the core mixed method designs described by Cresswell and Plano Clark (2018) including three distinct and sequential studies with subsequent studies seeking to explain or explore the findings from the earlier studies. The concepts presented in the qualitative evidence synthesis and the findings from survey, are explored in depth within the multiple case study, with the findings from each being integrated and interpreted within the discussion section of this thesis.

Figure 8. Mixed method explanatory sequential study design.



3.4 Ethical approvals

Ethics approval has been granted for the primary research included in this thesis. The HCP survey received approval from the University of Sheffield ethics panel (reference: 045355) on 5/5/22 and Health Research Authority (HRA) (reference: 308744) on 10/5/22. The multiple case study received NHS research ethics committee approval from West of Scotland Research Ethics Service (REC reference: 22/WS/0171) on 14/12/22 and from HRA and HCRW on 30/1/2023. A substantial amendment was approved by HRA and HCRW

Approval on 4/7/2023, to remove the requirement for every healthcare professional present at an MDT meeting to have given consent to be observed, allow the audio recording of interactions between healthcare professional and people with MND (an option that was not able to be used), and to allow people with MND to be sent reminders about expressing an interest in being observed.

3.5 Patient and public involvement (PPI)

Patients and the public can have input at every stage of the research process and such involvement has been associated with improved quality and relevance research (Brett *et al.,* 2014). There are several reasons to seek to collaborate with patients and public as described in the NIHR’s briefing notes for researchers (NIHR, 2021). These include for ethical reasons, with most health research involving the recruitment of patients, they should have a say in how such research is planned, conducted and disseminated. Collaborating with patients who have lived experience of a disease, care delivery or treatments, provides a valuable insight into both the design of the research question and design, but also experience of how the intended subjects of research will respond and what their information needs are. Finally, research grant and ethics panels now require researchers to describe how PPI has been and will continue to be involved in the project.

I have collaborated with people with MND and caregivers both during the conception of this research and during its design and conduct. While developing the application for the NIHR Doctoral Fellowship, I met with 4 different PPI panels, each with a different type of experience (general, palliative care, multiple sclerosis and MND) to develop the research questions and proposed research plan. In addition to these group meetings, I also met with two people with MND who had experience of making the decision to have a gastrostomy. The outcome of these meetings influenced the focus of the proposed research and the choice of research methods including highlighting some of the practical considerations I would need to consider. The meetings with people with MND confirmed that the experience of making the decision to have a gastrostomy was challenging and particularly highlighted the influence that their healthcare professionals had on their ability to make a decision. This was one of the reasons for wanting to widen the scope of the research further than just the experience of people with MND to include the perceptions of caregivers and healthcare professionals.

Prior to commencing the NIHR Doctoral Research Fellowship I recruited my own PPI panel through advertising via the MND Association (see appendix 1). I recruited several people with MND and a past carer, who I have met with each year at key time points throughout the fellowship. The group have been a valuable and critical source of support for the on-going conduct of the research. The PPI group were particularly involved in the design of the case study. We discussed how to recruit people with MND early in their decision-making process (ideally the first discussions). This resulted in an in-depth debate about how to approach people with MND about a study exploring decision-making about gastrostomy when they were not aware such a discussion may take place. While acknowledging the covertness of the proposal, it was agreed that it was not appropriate to inform people with MND about the topic of the research until after an interaction when gastrostomy had been discussed and where full consent could then be sought. The PPI group also highlighted the value of purposively sampling people with MND at different stages of decision-making (i.e. first discussions, subsequent discussions, making decisions), to gain contemporaneous insights through observations and interviews about how people with MND experience these different scenarios. Finally, the group highlighted the importance of recruiting healthcare professionals and observations of interactions outside of the MND clinic, whom they identified as also having detailed discussions with them about gastrostomy.

I have sought to inform my PPI group about the progress of the research through sending out ‘newspaper’ style updates (see appendix 2). Sadly, some members have died throughout the time of the fellowship, and I would like to acknowledge the valuable contribution they made to this thesis. This was always going to be a possibility given the nature of MND and even was a scenario that the group have joked about. In line with NIHR’s payment guidance, all PPI members were offered cash or voucher reimbursement for their time.

3.6 Overview of methods

There were three phases to the mixed methods research presented in this thesis. Firstly, the qualitative literature published was systematically synthesised, aiming to understand the experiences and perceptions of people with MND, caregivers and healthcare professionals in relation to making decisions about gastrostomy or ventilation. A thematic synthesis was the chosen method of synthesis. The findings are presented in Paper 1 and have since been published (White *et al*., 2023).

The outcomes of the qualitative evidence synthesis were used to develop a questionnaire aiming to understand healthcare professionals' practice and beliefs towards supporting people with MND making decisions about gastrostomy placement. Following pilot testing, the questionnaire was distributed using a snowball sampling approach as a cross-sectional online survey. The survey remained open for 10 weeks before being analysed using descriptive and comparative statistics to identify and compare differences between responses from the healthcare professional disciplines within the MDT. The findings are presented in Paper 2 and have since been published (White *et al*. 2024).

Three MND teams were selected to take part in a multiple qualitative case study. Within each case a range of qualitative data collections methods were used to explore how MND teams support people with MND to make decisions about gastrostomy including the perspectives of purposively sampled people with MND, caregivers and healthcare professionals. The Framework approach (Ritchie *et al.,* 2014) was used to manage and analyse the large quantity of textual data from non-participant observations, interviews, focus groups, medical note review, and documentation review. The findings from two key themes are presented in Papers 3 and 4.

In view of the limited word count available to include in the publication style chapters, further justifications for the choice of each research method used are provided below with additional information about the implementation of the methods included in the appendices.

3.7 Qualitative evidence synthesis

3.7.1 Rationale for qualitative evidence synthesis

The rationale for beginning with a qualitative evidence synthesis was borne out of a scoping of the literature that identified several concepts consistently reported in the findings of published studies focusing on decision-making in MND care. To date these findings had yet to be synthesised to develop an understanding of the intrinsic, interactional and organisational influences on the decision-making of people with MND. A further rationale for performing this review was to identify any gaps in the literature that the survey and multiple case studies could seek to address. Qualitative evidence synthesis involves systematically identifying and synthesising qualitative studies that will address the research questions under examination (Seers, 2012). Synthesising the findings from multiple studies allowed for the generation of a higher analytical representations of how decisions are made in MND care (Howell Major and Savin-Baden, 2010 p. 11-14). A paucity of quantitative evaluation of decision-making process or quality in MND care made a systematic review of quantitative studies unfeasible and would not have met the aim of understanding the contextual influences.

It was important to ensure a similar review had not already been published. A recently published meta-ethnography (Lisiecka *et al.,* 2021) was designed to synthesise the qualitative findings of studies that had focused on the experiences of people with neurodegenerative disease (not people with MND or other stakeholders e.g. caregivers and healthcare professionals) of being on enteral nutrition (not decision making). The searches conducted only identified studies relating to MND, although it did identify a theme focused on decision-making rather than experience of enteral nutrition due to the lack of rich data available. The review did not include the experience of other important stakeholders i.e. caregivers and healthcare professionals, only focused on the experience of people with MND making decisions about gastrostomy placement (i.e. no other interventions) and did not include search terms to capture studies exploring decision-making in the search strategy. People with MND make several decisions about interventions that have similarities in that they address significant health threats, are associated with being life-prolonging and have a significant impact on the experience of patients and caregivers living with MND. Indeed, several of the primary qualitative research papers have focused on the decisions people with MND make about gastrostomy and ventilation (Lemoignan and Ells, 2010; Greenaway *et al.,* 2015; Martin *et al.,* 2016). Previous evidence has also identified how people with MND, caregivers and several healthcare professional disciplines within the MDT are involved in the process of decision-making about gastrostomy and that there are challenges associated with this (Hogden *et al.,* 2012a; Chapman *et al.,* 2021; Van Eenennaam *et al.,* 2021). Therefore, there remains a gap in the literature to prospectively synthesise the qualitative literature focusing on how decisions are made about interventions in MND care from the range of stakeholder perspectives.

3.7.2 Overview of method

The protocol for the review was registered with PROSPERO (CRD42021283314). The design and conduct of the qualitative evidence synthesis was guided by the criteria included in the enhancing transparency in reporting the synthesis of qualitative research (ENTREC) statement (Tong *et al.,* 2012). See Appendix 3 for a summary of how the review met the criteria of the ENTREC statement. The development of the scope of the review and the research question was informed by scoping searches, discussion with supervisors, PPI and expert panel members and guided by the SPICE research question framework, the results of which are outlined in table 2 (Booth, 2006). Three key decisions were made during the design of the review protocol. First, was whether to include other progressive neurological conditions. The rationale for only including MND was the unique decision-making context of living with a rapidly progressive disease without effective treatments or a cure. Including qualitative accounts from other conditions lacked relevance to the overarching doctoral research question. Second, was to focus only on decisions about gastrostomy and ventilation with these being contemporaneous decisions routinely made by people with MND that have a significant impact on future outcomes and experience of living with MND compared to for example, decisions about genetic testing. Third, was to include the perspectives of people with MND, caregivers and healthcare professionals, taking the position that decisions are constructed through interactions between each and to capture the experiences and views of each about the decision support delivered by the MDT.

Table 2. The development of the review research question using the SPICE framework (Booth, 2006).

Setting	**P**erspective	Phenomena of **I**nterest	**C**omparison	**E**valuation
MND care	People with MND, caregivers, healthcare professionals, observations	Decision-making about commencing invasive healthcare interventions	None	attitudes, information needs, experience
Research question: What are the organisational, team and individual level factors that influence decision making by people with MND when making the decision to start an invasive healthcare intervention?				

Appendix 4 provides a summary of how the synthesis methodology was chosen using the RETREAT framework (Booth *et al.,* 2018). Framework synthesis and meta-ethnography were methods of qualitative evidence synthesis considered for this review. Candidate decision-making theoretical frameworks were identified (Waldron *et al.,* 2020) but were not well established and not a great fit for the data available (Booth *et al.,* 2016). While meta-ethnography is commonly used in healthcare qualitative synthesis (Cahill *et al.,* 2018), it requires significant time, resource and expertise. This level of interpretation was not a priority, and I aimed to focus on organising the key factors into interpretative themes (Booth *et al.,* 2016). For this reason, thematic synthesis using the method described by Thomas and Harden (2008) was employed.

A rigorous approach was taken throughout the design of the review protocol including in relation to the development of the search strategy guided by the STARLITE framework (see appendix 5) (Booth, 2006). The use of EndNote and Excel databases supported an audit trail including recording the removal of duplicates and reporting the study selection process using the PRISMA flowchart (see Figure 1, Paper 1) (Page *et al.,* 2021). Dependability of study selection was supported through a colleague reviewing 10% of the abstracts and full texts. The decision to only extract data from the finding's sections (not discussion sections) was to ensure that the data informing the themes remained grounded in the original data and authors analysis.

The CASP qualitative checklist (CASP, 2018) was chosen to assess the rigour with which the methods were implemented, and the assessment was reported for each study (see Table 1, Paper 1) (Long *et al.,* 2020). However, the outcome of the quality assessment did not influence decisions about whether a paper was included in the review or the weighting given to each study's findings (Pawson, 2006; Dixon-Woods *et al.,* 2007; Booth & Carroll 2015). Concerns about using such quality assessments of qualitative research include the ability for such tools to account for the different philosophical positions taken, data collection methods and methodologies used when conducting qualitative research (Dixon-Woods, 2004). The value of quality assessments is also limited by the quality or comprehensiveness of reporting in publications. If used to guide study selection quality assessment may exclude studies, a particular stakeholder's perspective or potentially important concepts that would threaten the credibility or transferability of the findings (Carroll and Booth, 2015; Long *et al.,* 2020).

3.8 Cross-sectional HCP Survey

3.8.1 Rationale for survey

Surveys provide an opportunity to standardise the collection of data about healthcare professionals' practice from a sample of a larger defined population and therefore allow findings to be generalised (Kelley, 2003; Weinstein and Sinkowitz-Cochran, 2013). However, generalisation of findings is only achievable if there is confidence in the validity and reliability of the data collection instrument used to collect the data. The survey presented here is interested in evaluating current practice and beliefs of different healthcare professionals in relation to supporting people with MND making decisions about gastrostomy placement. A cross-sectional survey achieves this aim and allows similarities or differences in current practice to be identified (Aarons, 2021). The emergence of online self-administered surveys has allowed researchers to increase the reach of the data collection and helps to manage some of the resource and time constraints that are commonly associated with survey research (Calleggaro *et al.,* 2015 p. 18-23). Self-administered surveys provide more reassurances about confidentiality and less risk of social acquiescence bias than telephone or face-to-face interview administered surveys (McColl *et al.,* 2001).

3.8.2 Overview of method

Methodological challenges associated with survey research include being able to collect data that is a valid and reliable representation of the sample population (Lau, 2017). There are several different methodological texts describing the sequential approach to good quality survey design which were adapted for the purposes of designing this survey (Kelley, 2003; Willis, 2005; Artino *et al.,* 2014). The design process included identifying evidence-based concepts from the findings of the qualitative evidence review, reference to wider literature and in consultation with topic experts (PPI, academic and HCP) which were developed into indicators and dimensions (see appendix 6) that could be measured through items included in the questionnaire (Kelley, 2003; Ziniel *et al.,* 2019; Aarons 2021). Crucial to the finalisation of the questionnaire was the feedback from nine healthcare professionals from the target population to confirm the face validity, reliability and usability of the survey (Artino *et al.,* 2014).

Key to the validity of this survey was the targeted sampling strategy that sought to recruit professional disciplines identified in previous surveys (Oliver *et al.,* 2011; Ruffell *et al.,* 2012; Van Eenennaam *et al.,* 2021) as being involved in supporting people with MND to make decisions about gastrostomy. The absence of a discrete and appropriate sampling frame led to the use of the snowball sampling technique using key gatekeepers to recruit a convenience sample of healthcare professionals meeting the inclusion criteria and therefore attempting to reduce sampling bias (Blair *et al.,* 2014; Ziniel *et al.,* 2019). In survey research, sampling bias is a more important issue than sample size, when aiming to generalise the findings to the wider population (Blair *et al.,* 2014). In addition to descriptive statistics to report the frequencies of responses, comparative statistical tests met the research objective to capture how the beliefs and practice of different healthcare professionals within the MDT differ which could be explored in more depth during the multiple case studies.

3.8.3 Ethical considerations

Survey research is required to adhere to the same ethical standards as any other type of research (Hammer, 2017; MRC, 2018). Identifying potential participants, informed consent, protecting participant anonymity and confidentiality, transfer and storage of data are particularly relevant to online survey research (Kelley, 2003; Fink, 2017). It is not possible to have direct discussion with potential participants recruited to an online survey (Hammer, 2017). Consent was obtained through participants indicating agreement with statements on a consent form included at the beginning of the survey, confirming they met the inclusion criteria. Even though no participant identifiable data was collected, there remains a risk of participants being identified through the responses they give e.g. by providing data about their profession or years of experience. Therefore, participants were not asked to provide any personal information that may be used to identify them supporting the anonymity of participating. All data collected was securely stored in password protected files University of Sheffield servers only accessible to the research team, as per the data management plan approved by University of Sheffield Ethics panel.

3.9 Multiple case study

3.9.1 Rationale for qualitative multiple case study

Quantitative evidence (Stavroulakis *et al.,* 2013; Martin *et al.,* 2014; Labra *et al.,* 2020; Van Eenennaam *et al.,* 2021) fails to answer 'why' and 'how' people make decisions about gastrostomy in the context of living with MND and in receipt of MDT care provision. The process by which people with MND make decisions cannot be separated from the social, emotional, psychological and clinical context in which it is occurring. While qualitative studies have explored the experience of decision-making about gastrostomy, they are limited to the experiences and perceptions of a specific group (e.g. people with MND, caregivers or healthcare professionals), and do not seek a case-based understanding of the interactions between the phenomenon and context. Ethnography was a qualitative research approach considered, which has similarities with the chosen case study design including the use multiple data collection methods. What delineates the two, is the intent and focus of the approach (Cohen and Court, 2003). Ethnographies involve long times spent in the field with a focus on understanding the cultural and social world from the perspective of the participants under study through researchers immersing themselves in the social entity being studied (Cohen and Court, 2003; Pope and Allen, 2020). The time spent in the field with case studies is often shorter and more focused than when conducting ethnographies (Crispin *et al.,* 2017). Case study research involves less time in the field seeking a deep case-based understanding of a phenomenon and context (and where they interact) through the exploration of different perspectives and using a range of research methods. The complexity of the disease, decision and organisation of healthcare provision are a rationale for the employment of qualitative case study research design.

What are case studies?

The aim to gain an understanding about how people with MND are supported to make decisions within the context of MDT care delivery is consistent with the qualities of case study research. Case studies have previously been used in healthcare research including nursing (Anthony and Jack, 2009), palliative care (Dodd *et al.,* 2020), emergency departments (Edwards *et al.,* 2021a) and decision making in dementia care (Smebye *et al.,* 2012). Several methodological texts have been published, including different definitions for case study research which recommend the integration and case-based reporting of findings from multiple sources using a range of research methods, including single or multiple cases (Luck *et al.,* 2006; Stake, 2006; Anthony and Jack, 2009; Yin, 2018). The current study has adapted methodological approaches from a number of these sources, but mostly adheres to the methods described by Yin (2018). While Yin (2018) provides a useful practical guide to planning and performing case study research, it should be acknowledged that his text is oriented to a more empirical variant of realism that is not completely aligned with my subtle realist position.

Integration of a range of data sources enables a detailed understanding of a phenomenon and particularly when multiple cases are studied, theoretical generalisation (Aerie, 2003; Stake, 2006; Anthony and Jack, 2009; Crowe *et al.,* 2011; Walshe, 2011; Yin, 2018). However, it remains a mode of academic enquiry that has struggled to establish itself as a valid approach including a range of misconceptions about the quality of the findings generated (Flyvbjerg, 2006). Many of the criticisms of case study research are also in fact its strength such as the lack alignment with a specific research paradigm allowing the flexibility to use data collection methods or data sources that can represent how a phenomenon of interest exists within the real-world context in which it is occurring. This pragmatic approach does not mean that case study research should lack the same rigour expected from any other research design. Rigour within the present study is supported by the development of a detailed protocol that can be viewed in appendix 7 which includes a detailed account of the methods that were planned to be used. While case studies have been described as not being bound by any single research paradigm or philosophy, for this study the research design is consistent with a subtle realist position (Hammersley, 1990; Brogan *et al.,* 2019). Qualitative case studies allow for the exploration of decision-making about gastrostomy occurring within real-world MND care delivery, through time spent in the field observing and interviewing those involved (Crowe *et al.,* 2011; Crispin *et al.,* 2017).

3.9.2 Overview of method

Further information about the methods used during this multiple case study can be read in the protocol (appendix 7) and in the methods section of Papers 3 and 4. A summary of the approach to recruitment and the methods employed are summarised in figure 9 and figure 10. Figure 9 shows sequential approach to the methods used and recruitment on each case. Figure 10 adapts Stake's (2016 p. 5) graphic design of a case study to summarise the settings, case boundaries, data collection and questions being ask within each case. Further information is included below about the methods I used that was unable to be covered in detail in Paper 3 and 4, including the justifications for some of the methodological decisions made and reflections on their implementation.

Figure 9. Sequential approach to recruitment and data collection

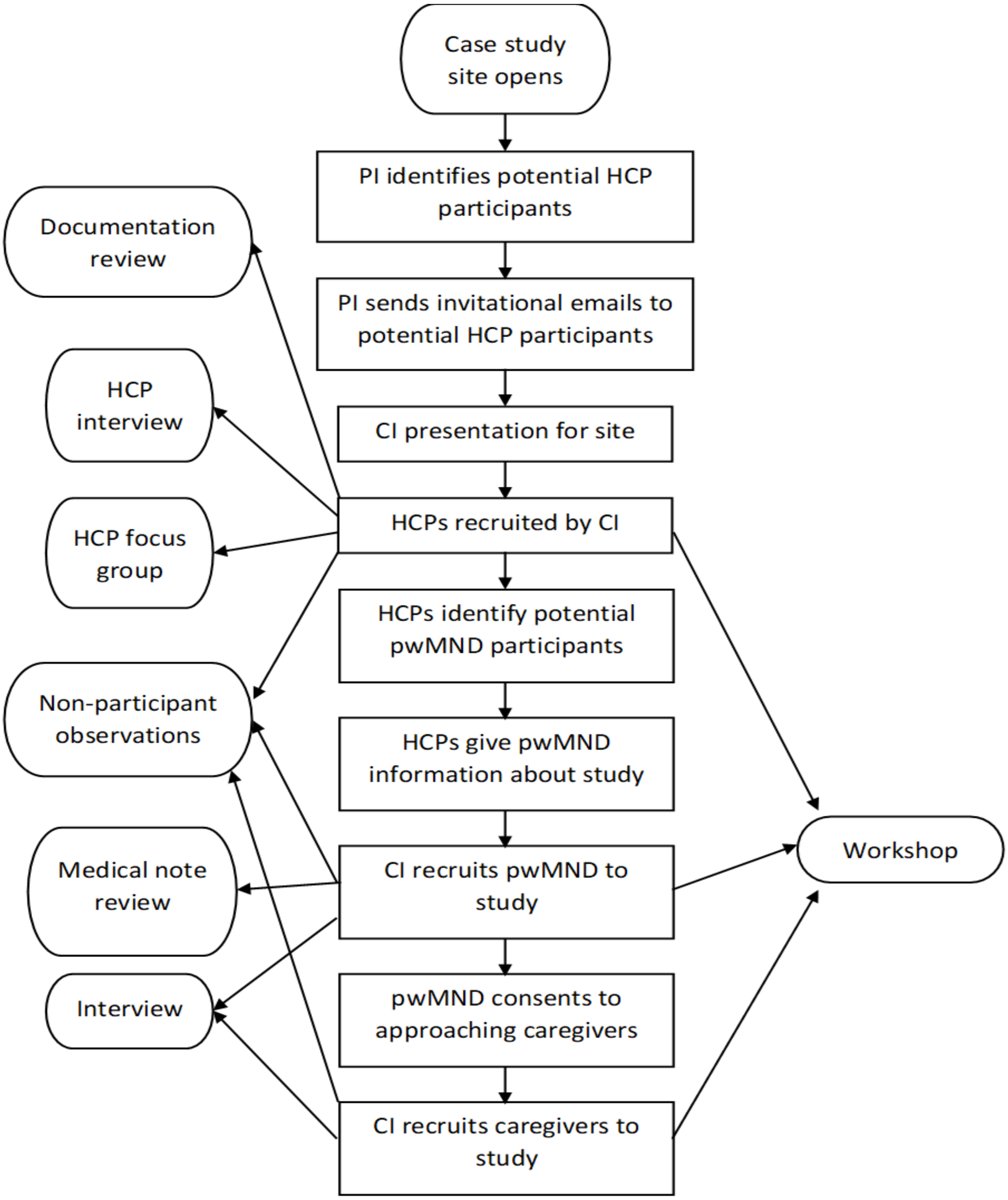
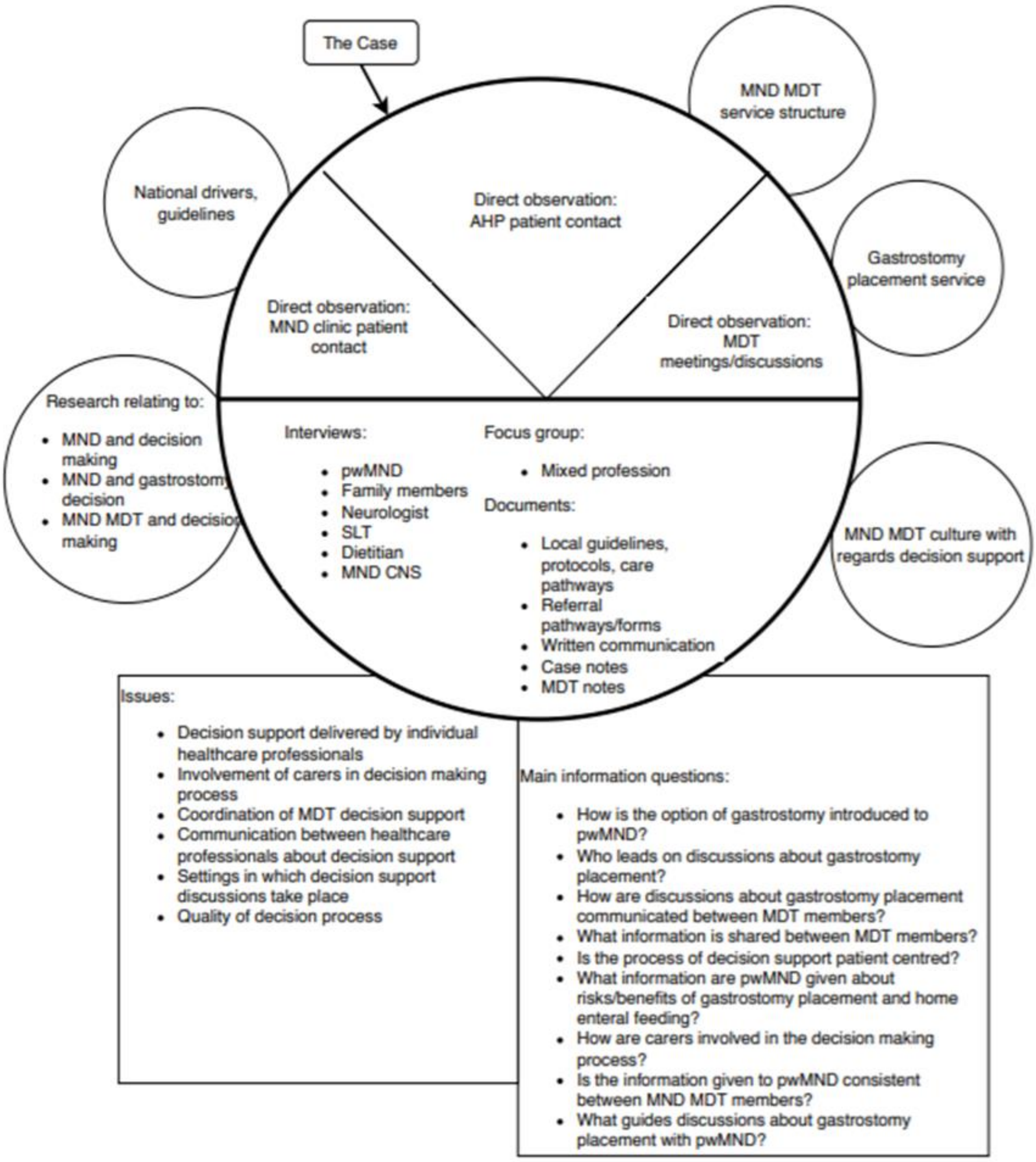


Figure 10. Graphic design of the case study. Adapted from Stake (2006) p. 5 with permission from the author within the book.



Number of cases

A key question during the design of case study research is whether to use a single or multiple case design. Yin's (2018) multiple case study approach was followed with the aim to compare cases to identify where there are consistencies (i.e. replication) and differences between cases and seek to explore reasons for these in greater depth. Yin's (2018), deductive reasoning for including multiple cases includes aiming to replicate findings in more than one case to support generalisability. This view is not particularly relevant when conducting a qualitative case study from a constructionist position. The rationale for including several purposively selected cases in this study is that it allows for comparison of the emerging concepts within and then between cases, enabling the development of richer analytical interpretations and supports the theoretical transferability of the study (Stake, 1995). The theoretical transferability of the findings is further supported by the rigour of study design and conduct, description of methods and the contextually rich reporting of findings (Glette and Wiig, 2022). Using a multiple-case design allows for the same phenomenon known to exist in all cases to be explored and compared across different contexts, organisations and settings (Brogan *et al.,* 2019) observing for replication and atypical cases, strengthening the credibility of the findings (Stake, 2006; Yin, 2018 p. 54-64). The selection of cases based on the characteristics of the size and organisation of MND care supports the rationale for including multiple cases. Stake (2006) recommends selecting 4-10 cases, with too few preventing access to observe how the phenomenon interacts with the context and with too many making it unfeasible to understand the 'uniqueness' of each case. The decision to include three cases was based on the resource a single researcher would have to spend within each case to collect enough data to be able to develop a credible account of each case and that it allowed for comparison of theoretically selected models of MND care.

Defining the case

An important requirement of case study research is that the case is clearly defined including the boundaries which limit the case (Ragin and Becker, 1992; Yin, 2018). For this study the definition of the case was 'the MND multidisciplinary team operating across the clinic and community setting that supported people with MND to make decisions about gastrostomy' and were restricted (i.e. boundaries) to the perceptions and actions of people with MND, caregivers and healthcare professionals within these cases. A broad definition of MND team is used to define the case and boundaries of the context, the case study is investigating. MND care is often delivered through an integration of tertiary, secondary and primary care provision, particularly in the UK (WHO, 2006; OPM, 2016; Berlowitz *et al.,* 2023). People with MND are supported by a regional specialist MND MDT clinic, followed by other healthcare teams nearer to home. People with MND will also occasionally need to attend hospital for tests, procedures or to address an acute clinical issue. Discussions about gastrostomy can take place in all these settings and therefore the case study sought to observe and recruit from within them all.

Case selection

The selection of case study sites should be theoretically or empirically informed while also include confidence about access to sites and the availability of relevant data (Stake, 2006 p. 24-27; Yin, 2018 p. 55). There were no data sets or research to provide *a priori* knowledge about how different cases support the decision-making of people with MND. Following a rigorous screening exercise, ‘representative cases’ of MND care provision were selected that included differences in their MND caseloads and the organisation of MND care delivery (Yin, 2018).

Recruitment and consent of cases

Seeking to recruit and observe interactions about gastrostomy across the tertiary, primary and secondary care MND healthcare system came with some practical and governance challenges. While it was possible to recruit all people with MND through their attendance at the central MND clinic, challenges were met in relation to observing interactions or recruiting healthcare professionals belonging to different organisations, which was the case for the community services in Case 1 and 2. It was not possible to recruit healthcare professionals practicing within hospices and other NHS organisations without opening them as a research site. This was a particular issue in Case B where all the community services belonged to a different NHS Trust to that of the MND clinic. With regards Case 3 the same healthcare professionals provided tertiary and primary care and within Case 1 some of the community services belonged to the same organisation as the MND Clinic and therefore could be recruited. While opening additional organisations as research sites was pursued in Case 2, the ethics and research governance processes took time and delayed data collection. No secondary care settings were accessed during the case studies. While discussions about gastrostomy were reported to happen on hospital wards, people with MND were often more unwell at these times and it would have been difficult to capture these discussions when they occurred. However, accounts of discussions on the wards were captured through accounts in the interviews, focus groups and medical note review.

Data collection

Further challenges were associated with seeking to observe interactions that included discussions about gastrostomy. The original restrictions set by the NHS REC permissions requiring all healthcare professionals present during an MDT meeting to have consented to allow an observation to take place was prohibitive. Healthcare professional attendance at MDT meetings could be transient and were mostly online making it difficult to obtain consent from all healthcare professionals beforehand. This barrier became less of an issue with time as more of the MDT were recruited and with the lifting of some restrictions granted as part of an NHS REC amendment. These are learnings for future research focusing on care delivered across care settings and organisations, including ensuring the researcher has a good knowledge of local services beforehand, opening all research sites from the start and constructing convincing arguments to ethics panels.

Originally, due to concerns that the number of people with MND meeting the inclusion criteria would be low and during the short time spent in the field it would be a challenge to meet the recruitment target, the intention was to recruit a convenience sample of people with MND within each case. This could have weakened the credibility of the findings. However, once in the field this was adapted to a purposive maximum variation sampling strategy because there was clearly scope for this to be implemented successfully (Bryman 2018). People with MND were recruited to observations and interviews who were at different stages of decision making (i.e. declined, deferred or made a decision) to generate data based on these perspectives and be able to compare responses between for example those who have had the option introduced for the first time against those who have made a decision. As a result of the important preparatory work for each case, including meeting with the case principal investigator and the healthcare team, and then further familiarisation once in the field (Chesluk and Holmboe, 2010; Pope and Allen, 2020), the healthcare professionals involved in supporting people with MND to make decisions about gastrostomy were able to be identified within each case. Again, a purposive sampling strategy was employed seeking for maximum variation in professional disciplines involved. Ritchie *et al.,* (2014, p. 259) discuss recruiting 'guides' within the organisation, who can aid with the familiarisation of sites and facilitate opportunities for recruitment and data collection. The gatekeepers for this study were the principal investigators (Pope and Allen, 2020). It was crucial to the success of each case that the principal investigator was someone with good knowledge of the local MND caseload and links with the local MDT (Millen, 2000).

A sequential approach was taken to data collection within each case (see figure 9). The order of data collection was guided by how each could inform the data collection of latter methods e.g. by adapting topic guides or informing purposive sampling of participants. In common with all research, it is important that methods are aligned with the researchers' philosophical position and in terms of case study research design, rationales can be provided for why each method is included. These justifications are provided below.

Non-participant observations

Observations used in healthcare research allow the researcher to generate an account of phenomena as they occur in real-world practice, generating different and possibly more naturalistic data than interviews or focus groups (Ritchie *et al.,* 2014 p. 245; Mason, 2018; Pope and Mayes, 2020). The method is consistent with my philosophical position that knowledge about how decisions are made could be generated through observing participants' interactions, including their responses, normative processes, behaviours and social norms (Mason, 2018). Observations were also used to inform the line of questioning during subsequent interviews and increased familiarity with the organisation of local healthcare services allowing future observation opportunities to be identified (Ritchie *et al.,* 2014 p. 249).

The main challenge with observations was that it was not always possible to predict when discussions about gastrostomy would take place, therefore necessitating observing several healthcare professional interactions with people with MND or other healthcare professionals that did not generate any data relevant to the research. While this was less of a problem with online MDT meetings which could be observed remotely, it did represent inefficiencies in relation to the limited time I had available to spend in the field, requiring some 'thinking on my feet' about where to position myself physically within clinics or which people with MND to observe, to optimise the likelihood of observing the interactions relevant to the research.

Semi-structured interviews

Consistent with my philosophical position, interviews allowed access to how participants experienced and perceived the social reality of decision-making about gastrostomy in MND care (Silverman, 2011 p. 131-148; Ritchie *et al.,* 2014 p. 178-180; Mason, 2018). The interview aimed to explore how people with MND respond to discussions about MND and gastrostomy, how they reason about the options available and the role played by the MDT in their deliberations. The series of interviews exploring the perspectives of different individuals within each case generated an understanding of the 'meaningful properties' associated with the reality under investigation (Mason, 2018 p. 111).

The interviews were conducted after several observations had taken place in each case for a number of reasons. First, the observations helped inform the purposive sampling of the participants to be interviewed. Second, similar to another MND study, it allowed the concepts emerging from the observations to be explored with those involved (Seeber *et al.,* 2019). Third, it allowed for differences to be identified between participants' accounts in interviews with what was observed occurring in observations or reported in the focus groups (Sampson *et al.,* 2020).

While participants were offered the choice, the interviews were ideally conducted in-person. There are clear practical benefits for both the researcher and participant associated with conducting interviews remotely including convenience, less costs, less travelling and where sensitive topics are to be discussed. However, careful thought is required with regards to the impact the setting may have on the data collected (Ritchie *et al.,* 2014 p. 182-183). In-person interviews provide greater opportunity to generate rapport, read body language, and engage in meaningful exploration of the issues. Particularly, in relation to interviews with people with MND and caregivers, in-person interviews were preferred as people with MND were more likely to have speech problems and there was increased potential for participants to be upset by the topics being discussed. All but one person with MND and two caregivers agreed or were available for an in-person interview. The in-person interviews were valuable for those people with MND with speech problems who either wrote or typed their responses to questions. A mix of in-person (n=6) and online (n=12) interviews were conducted with healthcare professionals, mainly for convenience and participant availability reasons.

Topic guides informed by the review, survey and the study objectives were developed for the start of the fieldwork. The topic guides for each participant type were iteratively updated to explore interesting observations in the field and concepts emerging from the analysis that was been conducted concurrently to data collection. An example of the initial interview topic guide used with people with MND can be viewed in Appendix 8. I feel I benefited from my years of clinical practice in terms of the experience of speaking to people with MND and employed advanced communication skills including active listening to identify and 'put a pin' in conceptually interesting and relevant disclosures that could be explored later in the interview (Mason, 2018). These interview and communication skills were important to ensure each interview contributed to the richness of data I was seeking to generate.

Medical note review

The aim of the medical note review was to understand what information healthcare professionals document and share in relation to the option people with MND have to place a gastrostomy. There were a number of rationales for including a medical note review in the case data sets which generated data about people with MND, healthcare professionals and the organisation of healthcare. First, the medical notes provided a source of longitudinal data (Yin, 2018 p. 114) that had been recorded contemporaneously by healthcare professionals about the participants with MND compared to the cross-sectional retrospective data generated during the interviews and focus groups. Second, healthcare professionals include in their documentation some insight into their reasoning and experience of discussing gastrostomy with people with MND. These are available to compare to the observations and accounts shared in the interviews and focus groups. Third, the healthcare professionals also document information about the response of people with MND to discussions and their reasons given for any decisions made. Again, these were available to be compared with the other units of analysis within each case. When interpreting the significance of the documentation and communication in the medical notes it is important to acknowledge that they were not produced for the purposes of academic enquiry (Yin, 2018 p. 116). The documentation of healthcare professionals was for the purpose of a professional requirement for keeping accurate records and to support the delivery of healthcare. The degree with which these represent a credible representation of healthcare professionals clinical reasoning or beliefs about the decision-making of people with MND should be viewed critically.

Focus groups

The aim of the focus groups was to understand the MND MDTs perceptions of how they, as a team and as part of the wider healthcare system, support people with MND to make decisions about gastrostomy locally. The focus groups were an opportunity to explore the concepts emerging from the other data collection methods with the local MDT as a collective, with a particular interest in how the team reason, function and perform rather than on individual healthcare professionals' practice or perceptions. The data generated by focus groups was the result of more naturalistic interactions (though not necessarily natural interactions) between participants rather than solely between the participant and researcher as occurred in the interviews (Green and Thorogood 2014 p. 127; Ritchie *et al.,* 2014 p. 212-213). In line with another study focusing on the role of the MDT in decision-making, a decision was made to assemble a group of mixed professional disciplines rather than those from single professions (Sohi *et al.,* 2015). MND teams are generally small in number and all participants of the focus groups were known to be involved in supporting people with MND decision-making about gastrostomy. An objective of the case study was to understand how the team coordinate and deliver decision support and therefore justified bringing together a group of healthcare professionals from different professions known to each other. Assembling a natural group of healthcare professionals familiar to each other has its risks though, including less depth of discussion about shared norms accepted as a given by the group and how comfortable participants will be sharing their views in front of their peers, particularly any that could be perceived as contentious or when there are any hierarchical power imbalances (Richie *et al.,* 2014 p. 233). There is also a risk of participants' beliefs changing as a direct result of the group discussion threatening the credibility of the data generated (Green and Thorogood, 2014 p. 137).

During each focus group a topic guide informed by the study objectives and the emerging analysis sought to encourage the group to discuss how they as a team supported people with MND making decisions about gastrostomy. The five phases of group interactions including forming, storming, norming, performing and adjourning guided the facilitation of the focus groups (Tuckman and Jenson, 1977). Each group were able to reach the ‘performing’ stage relatively quickly where the group were leading the discussion with very little prompting from myself, likely supported by the previous professional relationships they had clearly established.

Documentation review

Healthcare professional participants were all asked to share any documents including care pathways, guidelines, protocols or policies that may guide or have any relevance to how people with MND are supported to make decisions about gastrostomy placement. Information about each document was recorded using a document review proforma (see appendix 9), each of which was imported in NVivo and integrated into the Framework analysis.

Analysis

The Framework approach developed by Ritchie and Spencer (1994) is a method of data management and thematic qualitative analysis developed originally for use in social policy research but now used extensively in healthcare research (Gale *et al.,* 2013). Framework approach has been used to analyse data collected during case study research conducted in a variety of healthcare settings, including emergency care (Edwards *et al.,* 2021a); palliative care (Dodd *et al.,* 2020); evidence-informed decision making (Lukeman *et al.,* 2019); care delivery in Parkinson’s disease (Todd *et al.,* 2016) and decision making in dementia care (Smebye *et al.,* 2012). Framework is ideally suited to analyse case study data for several reasons, including supporting a deductive and inductive approach; managing a large volume of different data types; and facilitating the comparison of findings within and between cases.

The Framework approach included the 5 steps described by Richie and Spencer (1994): 1. Familiarisation; 2. Identifying a thematic framework; 3. Indexing; 4. Charting; 5. Interpretation of patterns (Ritchie and Spencer 1994). The following methodological papers and texts, describing the process of the Framework approach, have been used to inform analysis approach of this multiple case study (Gale at al 2013; Richie and Spencer *et al.,* 2014 p. 270-345; Parkinson *et al.,* 2016; Bonello and Meehan 2019; Goldsmith 2021). The transcriptions and field notes were imported into NVivo computer assisted qualitative data analysis software (CAQDAS). Familiarisation of each interview and focus group was supported through listening to the audio recordings and reading through the transcriptions. A process of indexing included the initial data being coded onto an a priori framework informed by the qualitative evidence synthesis and survey (Dodd *et al.,* 2020), broadly coding onto four descriptive themes i.e. initiating discussions, the process of decision-making, making decisions and MDT decision support. Time was spent organising the data into descriptive themes and sub-themes, rather than moving too quickly onto the analytical interpretations (Parkinson *et al.,* 2016). Broader or new descriptive themes and sub-themes were iteratively developed as new concepts emerged from the data. The same framework was applied within each case. The 'charting' stage involved using the Framework function in NVivo. A central matrix was developed that included data summaries for each unit of analysis (rows) under each subtheme (columns) (Gale *et al.,* 2013). Each row included a unit of analysis i.e. a single observation, interview, focus group, medical note review or document review. The organisation of the analysis in this way, allowed for the analytical interrogation of the cases and the themes, to identify relationships, that provided theoretical explanations about the phenomenon (Goldsmith 2021). It is these higher analytical and more abstract interpretations that supported the transferability of the findings. The development of higher-level explanations involved referring back to the primary data, published research and discussion with supervisors (Ritchie and Spencer 1994). The development of the matrices supported the implementation of a process of pattern matching as an analytical technique (Yin, 2018, p. 175-178). A case-based approach was taken to the cross-case analysis, to compare the data representing each theme across the three cases, looking for replication and differences (Yin 2018, p. 196). Returning to the data summaries in the NVivo matrices (including linking back to read the source data – a significant benefit of using NVivo) to interrogate the data further across units of analysis or down specific sub-themes allowed for patterns to be identified and supported the development of the analytical themes presented in Papers 3 and 4.

3.9.3 Ethical considerations

There were many ethical considerations associated with the multiple case study which are discussed below.

Consent

A significant ethical issue that required much thought, debate and PPI consultation involved obtaining informed consent from people with MND prior to the observations of healthcare professionals and people with MND. People with MND are often not aware that gastrostomy would be discussed during a consultation with their HCP ahead of that interaction taking place. Equally, while some healthcare professionals may have planned to discuss gastrostomy, it may also spontaneously arise as a topic during interactions as a result of the person with MND raising it or in response to clinical assessment indicating a need. Following discussion with my PPI group, I decided that it could cause people with MND anxiety and distress if they were informed in advance (i.e. by sending them study information about the aim of the study) that their healthcare professional intended to discuss gastrostomy. A published review described scenarios where not gaining full informed consent may be legitimate including where the consent process itself could lead to increased stress or anxiety (Rebers *et al.,* 2016). Observations of interactions between people with MND and healthcare professionals at different stages of decision-making were deemed an important perspective to integrate into the case study data set. A decision was made, with support from my PPI group, to not include in the participant information that the study focused on decision-making about gastrostomy but instead only disclosed that it was exploring how healthcare professionals communicate with people with MND. To ensure consent was provided for any relevant observation to be included in the data set, further consent was sought following the interaction where all information about the study was disclosed and reasons given for the nature of consent used prior to the interaction. All people with MND pwMND recruited understood and accepted the rationale given. Unfortunately, I was not given NHS REC permission to observe consultations where people with MND were given their diagnosis despite these interactions being identified as times where gastrostomy can first be discussed. Future research should explore with PPI and NHS ethics panel's how these consultations could still be included as they likely represent a particularly unique context in which the option is discussed.

A further ethical consideration in relation to the ethics of consent related to the observation of MDT meetings. During these meetings healthcare professionals would discuss a range of patient cases, which could be pre-planned or ad-hoc. To optimise the likelihood of observing any discussion I needed to observe the entire meeting including discussions not relevant to the research. A decision was made not to seek consent from people with MND to observe the discussions healthcare professionals have about them in the MDT meetings. The ethical justification for this was it had the potential to cause upset if informed their healthcare professionals were considering offering gastrostomy especially if this had yet to be discussed directly with them. An argument was made that including these observations had a public interest benefit through improving the quality of the data collected. However, the ethical concern was that a researcher external to the healthcare team was gaining access to patient identifiable information and to discussions about their care without their explicit consent. To limit exposure to patient identifiable information, the observations of MDT meetings were not audio recorded and only field notes of discussions about gastrostomy were included in the study data set. This decision underwent significant scrutiny during PPI meetings and from the NHS REC panel and required approval from the confidentiality advisory group (CAG), with the approach presented being given ethical approval.

Interfering with real-world practice during observations

During the observations I sought to minimise the impact I was having on the natural clinical environment through carefully positioning myself out of a participant's eye line, turning off my computer camera and microphone during online meetings and avoiding any conversation where possible. Through these precautions I aimed to observe discussions, actions and practices as they would occur if I had not been present, accepting that my presence inevitably would impact on the interaction. Healthcare professionals were repeatedly reminded prior to any interaction observed to continue with their normal practice and not discuss gastrostomy just because they were being observed. While for most observations I was able to remain uninvolved during the interactions there were a number of occasions where this was not possible. For example, participants being observed would on occasion invite me into conversation during a consultation. When this occurred, without being rude I would seek to disengage as soon as possible to allow the consultation to continue. Another situation involved a neurologist showing the person with MND a film from the mytube website on their computer which included myself talking about the gastrostomy decision. This incident revealed that I was a healthcare professional and one with knowledge about gastrostomy placement in MND care, something that had not been disclosed to the person with MND previous to the observation. Finally, a person with MND and caregiver, had researched me online prior to attending a planned clinic observation and found out that I was a researcher and healthcare professional interested in MND and enteral nutrition. The same person with MND and caregiver, questioned during the interview whether the neurologist had only discussed gastrostomy because he was being observed for the purposes of my research.

The above scenarios capture the challenge associated with efforts to minimise the impact observations have on the natural settings under investigation. Participant knowledge about the research aim has the potential to change behaviour or their actions which represents a threat to the credibility of the research findings. From an ethical perspective, the healthcare patients are receiving could also be altered as a result of the observation. An alternative less obtrusive approach would be to audio record or video the consultations, to remove the researcher from the natural environment, though this would also likely come with practical challenges in terms of all healthcare professionals having recording devices available and relying on them to use the equipment correctly. Other than the incidents summarised above, I felt I was successful in minimising my impact on the environment in which the observations took place.

Anonymity

Case studies have been associated with a risk to revealing the identity of participants, and therefore caution should be taken about how participants or case sites are described (Crowe *et al.,* 2011). MND MDTs usually include a small number of healthcare professionals, with some professions represented by a single individual. Describing the demographics of the participants risks that healthcare professional participants could be identified in the reporting of the study. Identities were protected using pseudo-anonymised study identifiers and storing participants identifiable information in a separate protected folders to the data. Any information recorded in fieldnotes or transcriptions that may support identification of participants was deleted or anonymised including names, professional roles, geographical locations, and names of clinics or hospitals. When using any quotes to illustrate the findings, information about the participant source was limited to the Case and whether they were a person with MND, caregiver or healthcare professional.

3.10 Summary of the chapter

This chapter has defined the philosophical position from which I have designed and conducted this study, justifying the use of a mixed methods approach. I provided further justifications and information about the methods used, to add to the limitations that can be given in the paper format chapters. Information about the methods is also included in the papers presented in Chapters 4, 5, 6, and 7.

4. Qualitative evidence synthesis (Paper 1)

4.1 Chapter outline

The following chapter presents a qualitative evidence synthesis in publication format. The paper includes a short review of the literature to identify the research gap and development of the research question. A summary of the research methods is included with further information available in Chapter 3, Section 3.7, and the appendices. The thematic synthesis identified four analytical themes: ‘an emotional response to interventions’, sharing the decision with others’, ‘control’ and ‘tipping the balance’. Finally, the findings are compared to the existing literature with a focus on the emotional response, autonomy and timing of decisions.

4.2 Qualitative evidence synthesis research question

The research question the qualitative evidence synthesis was seeking to answer was:

> ‘What are the organisational, team and individual level factors that influence decision making by people with MND when making the decision to start an invasive healthcare intervention?’

4.3 Further information related to the study

Further information related to the development or conduct of the qualitative evidence synthesis can be read in the following appendices:

- Appendix 3 ENTREC statement
- Appendix 4 Selecting the qualitative evidence synthesis using the RETREAT framework
- Appendix 5 Search strategy described in terms of the components of the STARLITE framework
- Appendix 10 Full Medline on Ovid search strategy
- Appendix 11 Summary of supplementary search strategy.
- Appendix 12 License agreement to use publication in thesis from Health Expectations

4.4 Publication details

The qualitative evidence synthesis has been published in Health Expectations Journal. The full reference for the publication is:

> White S, O'Cathain A, Halliday V, Croot L, McDermott CJ. Factors influencing decisions people with motor neuron disease make about gastrostomy placement and ventilation: A qualitative evidence synthesis. *Health Expect*. 2023; 26: 1418-1435. doi:10.1111/hex.13786

The full paper is presented below in section 4.5. See Appendix 12 for permissions to include the Paper verbatim in this thesis.

4.5 Paper 1: Factors influencing decisions people with motor neuron disease (MND) make about gastrostomy placement and ventilation: a qualitative evidence synthesis

Abstract

Background: People with motor neuron disease (MND) (pwMND) are routinely offered gastrostomy feeding tube placement and (non-invasive and invasive) ventilation to manage the functional decline associated with the disease. This study aimed to synthesise the findings from the qualitative literature to understand how individual, clinical team and organisational factors influence pwMND decisions about these interventions.

Methods: The study design was guided by the ENTREC statement. The search of 5 bibliography databases and an extensive supplementary search strategy identified 27 papers that included qualitative accounts of pwMND, caregivers and healthcare professionals' (HCPs) experiences of making decisions about gastrostomy and ventilation. The findings from each study were included in a thematic synthesis.

Findings: Making a decision about interventions is an emotional rather than simply a functional issue for pwMND. The interventions can signal the end to normality, and increasing dependence, where pwMND consider the balance between quality of life and extending survival. Interactions with multiple HCPs and caregivers can influence the process of decision-making and the decisions made. These interactions contribute to the autonomy pwMND are able to exert during decision-making. HCPs can both promote and threaten pwMND perceived agency over decisions through how they approach discussions about these interventions. Though there is uncertainty over timing of interventions, pwMND who agree to interventions report reaching a tipping point where they accept the need for change.

Conclusion: Discussion about gastrostomy and ventilation options generate an emotional response in pwMND. Decisions are the consequence of interactions with multiple external agents including HCPs treading a complex ethical path when trying to improve health outcomes while respecting pwMND right to autonomy. Future decision support interventions that address the emotional response and seek to support autonomy have the potential to enable pwMND to make informed and timely decisions about gastrostomy placement and ventilation.

Patient or public contribution

The lead author collaborated with several patient and participant involvement (PPI) groups with regards the conceptualisation and design of this project. Decisions that have been influenced by discussions with the multiple PPI panels include widening the scope to decisions about ventilation in addition to gastrostomy placement and the perceptions of all stakeholders involved (i.e. pwMND, caregivers and HCPs).

KEY WORDS

Motor neuron disease; Amyotrophic lateral sclerosis; decision-making; gastrostomy; ventilation; qualitative

1 INTRODUCTION

Motor neuron disease (MND), also known as amyotrophic lateral sclerosis (ALS), is a progressive neurological condition with a global incidence of 1.59 per 100,000 person-years and prevalence of 4.42 per 100,000 population.[1] MND is associated with a 2-5 year survival after symptom onset with a lack of therapeutic options that can delay disease progression.[2,3] The clinical management of MND focuses on compensating for the progressive loss of vital physiological functions including respiratory failure and swallowing difficulties.[4]

People with MND (pwMND) are routinely presented with options to start interventions aimed at improving quality of life and/or extending survival.[3] Interventions offered include gastrostomy placement providing an alternative route for administering nutrition and hydration, and ventilation (non-invasive and invasive) for respiratory support.[5,6] Clinical guidance recommends discussing such interventions early in the disease course and the timely initiation of interventions to reduce procedural complications and promote better outcomes.[7-9] Though these interventions have the potential to extend survival[10-13], they can also address the negative impact that functional losses have on pwMND quality of life.[3] In addition to the proposed benefits, the risks and burdens should also be considered during decision-making.[14,15]

pwMND make decisions in the context of an often rapidly progressive disease with no hope of a cure. The risks and benefits associated with interventions are continually changing as the health threats presented by symptom progression escalate. King *et al.,* (2009) developed a cyclical model of decision-making describing how pwMND repeatedly respond and adapt to the relentless step-changes in their condition.[16] There are a range of contextual and relational factors that influence how, what and when, pwMND make decisions about their care.[17,18] Decisions are the result of interactions with a range of external agents including caregivers and the multidisciplinary team (MDT) supporting them.[18-20] In addition, the cognitive and behavioural changes prevalent in MND impact on the abilities of the individual to make decisions and the support they need to do so, adding to the complex context in which pwMND make decisions about their care.[21,22] There are a growing number of qualitative studies that have captured the perspectives of pwMND, caregivers and HCPs during decision-making about gastrostomy and ventilation. This synthesis and further conceptualisation of decision-making about these interventions in MND care from multiple perspectives, will inform the future development of contextually sensitive decision support strategies.

The aim of this qualitative evidence synthesis is to understand how individual, clinical team and organisational factors influence the decisions that pwMND make about gastrostomy and ventilation (for the purpose of this paper 'gastrostomy placement and ventilation (non-invasive and invasive)' will be referred to as 'interventions' from this point onwards).

2. METHODS

2.1 Design

A qualitative evidence synthesis was selected to systematically identify and synthesise the findings from the published qualitative literature, to gain a broad and rich understanding of the factors that influence decision-making about interventions in MND care.[23] The study design was guided by the enhancing transparency in reporting the synthesis of qualitative research (ENTREC) statement (Appendix A (Appendix 3 in this thesis).[24] The protocol was registered on PROSPERO on 14th October 2021 (CRD42021283314).

2.2 Search strategy

The search strategy was informed by a scoping search and in consultation with subject experts, academic information specialist, patient and public involvement groups and the research team. All searches took place during September and October 2021. Free text terms and subject headings related to the concepts ‘motor neuron disease’, ‘decision-making’, ‘gastrostomy OR ventilation’ and ‘qualitative study’ were combined using the Boolean term ‘AND’. The search strategies were adapted for searches on five bibliographic databases: Medline, Embase, CINAHL, PsycINFO and Cochrane Library database (see Appendix B for the full Medline search terms). An extensive supplementary search strategy was conducted to identify any references not captured by the bibliographic database searches (Appendix C (Appendix 11 in this thesis)). The reference lists of included studies, relevant guidelines and reviews, and the previous three years' contents of the ‘Amyotrophic Lateral sclerosis and Frontotemporal Degeneration’ journal and the International Symposium on ALS/MND conference proceedings were hand searched. A forward citation check of all the studies included in the review was performed using the Web of Science database. The authors of included studies and selected topic matter experts were contacted to identify further studies that may meet the inclusion criteria.

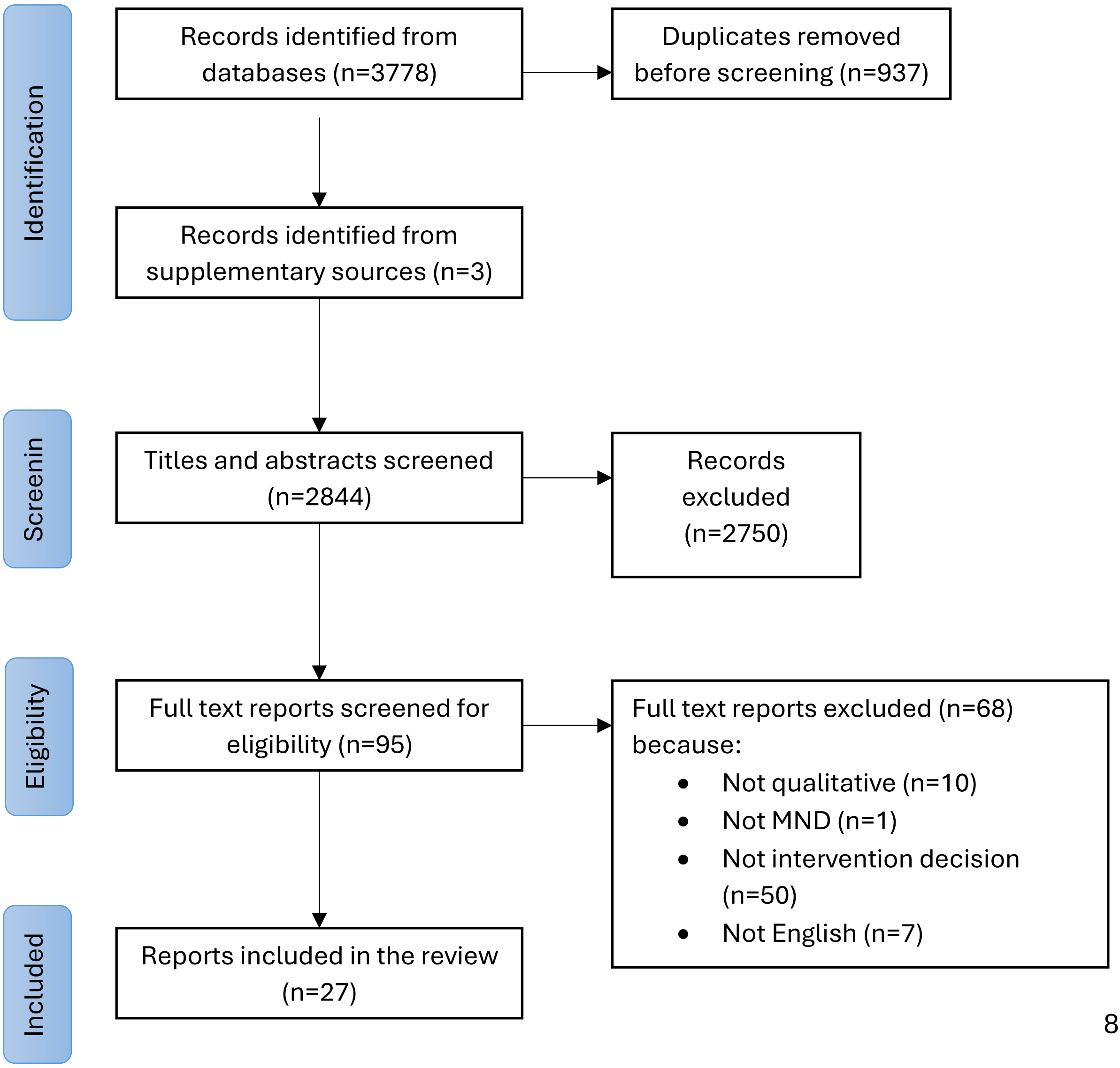
2.3 Inclusion criteria

Studies were included if they contained qualitative accounts of pwMND, caregivers or HCPs making decisions about interventions. Caregivers were defined as being unpaid people who support pwMND. Only studies in the English language were included and no date restrictions were set.

2.4 Study screening and selection

A total of 3781 references identified by the database and supplementary searches were imported into EndNote 20 reference manager. Following the removal of 937 duplicates, the title and abstracts of 2844 references were screened for inclusion by SW. A total of 95 full text papers were screened for inclusion in the review by SW. LC screened 10% of the references at both the title/abstract and full text screening stages. Any disagreements were discussed and resolved between SW and LC, with further discussions with CM, AC and VH when required. Finally, 26 papers met the inclusion criteria to be included in the review. The supplementary searches identified one further paper for inclusion.[25] See figure 1 for full results of the study screening and selection process.

FIGURE 1 Results of the study screening and selection process



2.5 Quality assessment

The CASP qualitative checklist was used to assess the quality and rigour of the included papers (Table 1).[26] The quality assessment did not guide study selection due to the documented concerns relating to the consistency and subjectivity of such appraisals.[27-29] Two studies scored poorly across most domains of the CASP checklist. Leslie (2008) did not include any methods, analysis or discussion, but the transcripts from the interviews provided interesting insights that were relevant to the aims of this review.[30] Versalovic and Klein (2020) paper lacked information about the study design and analysis, but the richness of the accounts addressed the review aim.[31] Only six studies discussed the researchers' relationship (i.e. reflexivity) with the participants.[32-37]

TABLE 1 Quality assessment of the included papers using the CASP checklist.[26]

Study Year	1	2	3	4	5	6	7	8	9	10
Ando *et al.,* (2015)[32]	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Chapman *et al.,* (2021)[25]	Y	Y	Y	Y	Y	N	Y	Y	Y	Y
Clarke *et al.,* (2018)[53]	Y	Y	Y	Y	Y	N	Y	Y	Y	C
Foley *et al.,* (2014a)[38]	Y	Y	Y	Y	Y	N	Y	Y	Y	Y
Foley *et al.,* (2014b)[39]	Y	Y	Y	Y	Y	N	Y	Y	Y	Y
Foley *et al.,* (2016)[40]	Y	Y	Y	Y	Y	N	Y	Y	Y	Y
Gottberg *et al.,* (2021)[54]	Y	Y	Y	Y	Y	N	Y	Y	Y	Y
Greenaway *et al.,* (2015)[48]	Y	Y	Y	Y	Y	N	Y	Y	Y	Y
Hirano and Yamazaki (2010)[50]	Y	Y	Y	Y	Y	N	Y	Y	Y	Y

Hodgins *et al.,* 2020[55]	Y	Y	Y	Y	Y	N	Y	Y	Y	Y
Hogden *et al.,* (2012a)[17]	Y	Y	Y	Y	Y	N	Y	Y	Y	Y
Labra *et al.,* (2020)[33]	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Lemoignan and Ells (2010)[34]	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Leslie (2008)[30]	N	Y	N	N	N	N	N	N	N	C
Martin *et al.,* (2016)[46]	Y	Y	Y	Y	Y	N	Y	Y	Y	Y
Murray *et al.,* (2016)[49]	Y	Y	Y	Y	Y	N	C	Y	Y	Y
Nolan *et al.,* (2008)[57]	Y	Y	Y	Y	Y	N	C	Y	Y	Y
Paynter *et al.,* (2020)[35]	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Pols and Limburg (2016)[41]	Y	Y	Y	Y	Y	N	C	N	Y	Y
Sakellariou (2016)[47]	Y	Y	Y	C	Y	N	C	Y	Y	Y
Seeber *et al.,* (2019)[36]	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Stavroulakis *et al.,* (2014)[45]	Y	Y	Y	Y	Y	N	C	Y	Y	Y
Sundling *et al.,* (2009)[56]	Y	Y	C	Y	Y	C	Y	Y	Y	Y
Veronese *et al.,* (2014)[52]	Y	Y	Y	C	C	N	Y	Y	Y	Y
Versalovic and Klein (2020)[31]	C	C	C	N	N	N	N	N	C	Y
Whitehead *et al.,* (2011)[51]	Y	Y	Y	Y	Y	N	Y	Y	Y	Y
Young *et al.,* (1994)[37]	Y	Y	C	Y	Y	Y	N	C	Y	Y

Note: Results of the quality assessment using the CASP qualitative checklist.[26] Criteria labels: 1. Was there a clear statement of the aims of the research?; 2. Is a qualitative methodology appropriate?; 3. Was the research design appropriate to address the aims of the research?; 4. Was the recruitment strategy appropriate to the aims of the research?; 5. Was the data collected in a way that addressed the research issue?; 6. Has the relationship between the researcher and participants been adequately considered?; 7. Have the ethical issues been taken into consideration?; 8. Was the data analysis sufficiently rigorous?; 9. Is there a clear statement of findings?; 10. How valuable is the research? (note: Question 10 of the CASP tool does not seek a yes/no/can't tell answer; yes has been selected when the author does address the hints provided in the tool). Abbreviations used: Y=Yes; N=No; C=Can't tell

2.6 Study characteristics

This review includes a synthesis of the findings from 25 studies reported in 27 papers, (three papers included the qualitative analysis of the same participant cohort from one study[38-40]) Of the 430 participants in the 25 studies, 241 were pwMND (where stated: 118 male, 75 female), 103 were caregivers (where stated: 35 male, 52 female) and 85 were HCPs (genders not stated). One study did not include any information about participants.[31] All the studies used interviews, with two studies also including observations[36,41] and another group interviews[17]. The papers included qualitative accounts of making decisions about gastrostomy alone (N=12), ventilation (non-invasive and invasive) alone (N=8) and both gastrostomy and ventilation (N=7). The studies were conducted in a number of countries including: UK (N=8); Australia (N=5); Netherlands (N=2); Sweden (n=2); USA (N=2); Canada (N=2); Ireland (N=1); Japan (N=1); Italy (N=1); not available (N=1). See table 2 for the study characteristics.

TABLE 2 Characteristics of the papers included in the review.

Author (year)	Country	Study aim	Perspective	Intervention	Study design / methodology	Sample size: total and per participant type; (gender)
Ando *et al.,* (2015)[32]	UK	Explore why pwMND declined or withdrew NIV, to understand patient experience of being offered NIV	Patients	NIV	Semi-structured interviews	9 (M=7; F=2)
Chapman *et al.,* (2021)[25]	Australia	Explore communication between pwMND and their clinicians about NIV and gastrostomy	Patients; HCP; caregivers	Gastrostomy and NIV	Semi-structured interviews	26; pwMND=1; Caregivers=6; HCP=19; (gender N/A)
Clarke *et al.,* (2018)[53]	UK	What are the experiences and views of pwMND and their families on decision-making concerning their care, with a focus on problems with eating and drinking	Patients; caregivers	Gastrostomy	Qualitative interviews	7; 4 pwMND (gender N/A), 3 caregivers (M=1; F=2)

Foley *et al.,* (2014a)[38]	Ireland	Identify processes that underpin how and why people with ALS engage with health care services.	Patients	Gastrostomy	Qualitative interviews	34 (M=17; F=17)
Foley *et al.,* (2014b)[39]	Ireland	Identify key psycho-social processes that underpin how people with MND engage with their services.	Patients	Gastrostomy and NIV	Qualitative interviews	34 (M=17; F=17)
Foley *et al.,* (2016)[40]	Ireland	Exploring pwMND experiences of receiving care from family members and formal service providers	Patients	NIV	Qualitative interviews	34 (M=17; F=17)
Gottberg *et al.,* (2021)[54]	Sweden	Investigate the experience of caregivers for pwMND on invasive ventilation via tracheostomy.	Caregivers	Mechanical ventilation via tracheostomy	Semi-structured interviews	8 (M=2; F=6)
Greenaway *et al.,* (2015)[48]	UK	To identify factors that affect pwMND accepting or declining NIV and gastrostomy.	Patients	Gastrostomy and NIV	Semi-structured interviews	21 (M=13; F=8)
Hirano and Yamazaki (2010)[50]	Japan	Investigating factors affect pwMND decision-making about invasive mechanical ventilation	Patients	Invasive mechanical ventilation	Semi-structured interviews	50 (M=34; F=16)

Hodgins *et al.,* 2020[55]	UK	Evaluate the impact of the Edinburgh Cognitive and Behavioural ALS Screen (ECAS) on pwMND, caregivers and HCPs.	Patients, caregivers and HCPs	Gastrostomy	Semi-structured interviews	21 (gender N/A)
Hogden *et al.,* (2012a)[17]	Australia	To explore patient decision-making for symptom management from the experience of health professionals and to identify factors influencing decision-making in specialised multidisciplinary ALS care	HCPs	Gastrostomy and ventilation	In-depth interviews and group interviews	32 (gender N/A)
Labra *et al.,* (2020)[33]	Australia	What are the factors that impact on pwMND uptaking gastrostomy	Patients	Gastrostomy	Mixed methods including standardised assessments, nutrition survey and semi-structured interview	10 (M=6; F=4)
Lemoignan and Ells (2010)[34]	Canada	To explore the experience of decision-making about assisted ventilation for pwMND.	Patients	NIV and mechanical ventilation	Semi-structured interviews	9 (M=6; F=3)

Leslie (2008)[30]	USA	To explore patients' thoughts on information, values, outside pressure, support and their reflections back on the process of making decisions about PEG.	Patients	Gastrostomy	Semi-structured interview	2 (F=2)
Martin *et al.,* (2016)[46]	UK	To investigate factors affecting decision-making about gastrostomy and NIV by people with ALS from the viewpoint of the HCPs supporting them through their decision-making.	HCPs	Gastrostomy and NIV	In-depth qualitative interviews	19 (gender N/A)
Murray *et al.,* (2016)[49]	Australia	To elicit the experiences of bereaved caregivers of MND patients who had or had not completed the letter of future care	caregivers	Gastrostomy	Semi-structured interviews	18 (M=5; F=13)
Nolan *et al.,* (2008)[57]	USA	Compare the preferences of patients with ALS for involving family in health care decisions at the end of life with the actual involvement reported by the family after death.	Caregivers	Gastrostomy	In-depth qualitative interviews	16 (M=8; F=8)

Paynter *et al.,* (2020)[35]	Australia	To explore involvement and engagement in decision-making, and how this was affected by communication or cognitive impairments.	Patients and caregivers	Gastrostomy and NIV	Semi-structured interviews	35; 19 pwMND (M=10; F=9), 15 caregivers (M=5; F=10)
Pols and Limburg (2016)[41]	Netherlands	To learn more about what the meaning of the term quality of life means when it is studied in daily life, in reference to gastrostomy feeding in ALS care.	Patients; caregivers and HCPs	Gastrostomy	Non-participant observation (N=28) and semi-structured interviews (N=11)	39; 11 pwMND interviewed (gender N/A); 28 pwMND observed (gender N/A)
Sakellariou (2016)[47]	UK	How do people involved in relationships of care enact subjectivity	Patients and caregivers	Gastrostomy	Interviews	2; 1 pwMND (F=1), 1 caregiver (M=1)
Seeber *et al.,* (2019)[36]	Netherlands	Evaluate timing and content of discussions about treatments and end of life care	Patients	Gastrostomy and NIV	Non-participant observation and in-depth interviews	21; (M13; F=8)
Stavroulakis *et al.,* (2014)[45]	UK	To explore the decision-making process in relation to timing of gastrostomy insertion from the perspective of the patients and their informal carers.	Patients and caregivers	Gastrostomy	Semi-structured interviews	18; 10 pwMND (M=7; F=3), 8 caregivers (gender N/A)

Sundling *et al.*, (2009)[56]	Sweden	To describe the experiences of patients with ALS as well as their caregivers, of non-invasive positive pressure ventilation	Patient and caregiver	NIV	Interviews	15; 7 pwMND (M=5; F=2), 8 caregivers (M=2; F=6)
Veronese *et al.*, (2014)[52]	Italy	To identify how the decision of a tracheostomy was taken by the patients	Caregivers	Tracheostomy	Semi-structured interviews	19; spouses=11, children=7, paid carer=1 (gender N/A)
Versalovic and Klein (2020)[31]	N/A	To elucidate the ways patients make sense of who they are and who they will be at later stages of illness through their conversations with and considerations of the others around them	Patients	Gastrostomy	Interviews	NK
Whitehead *et al.*, (2011)[51]	UK	To explore the experiences of people with Motor Neurone Disease (MND), current and bereaved carers in the final stages of the disease and bereavement period	Caregivers	NIV	Narrative interviews	18 (M=11; F=7)

Young *et al.,* (1994)[37]	Canada	To identify the factors involved when pwMND are making a decision to start mechanical ventilation	Patients	Mechanical ventilation	Semi-structured interviews	13 (gender N/A)

Abbreviations: HCP: healthcare professional; pwMND: person with MND; NIV: non-invasive ventilation; M: male; F: female; N/A: not available.

2.7 Data synthesis

A scoping search of the current evidence and the resources available informed the decision to perform a thematic synthesis.[42] Thematic synthesis allows for the generation of analytical findings that conceptualise how decisions are made in the context of MND care. The methods described by Thomas and Harden (2008), were followed including: 1. Line-by-line coding of text; 2. Developing descriptive themes; 3. Developing overarching analytical themes.[43]

Papers were imported into QSR NVivo (Version 13) for coding and analysis by SW. Only the text present in the findings section of the papers were extracted for the analysis.[43,44] All of the papers were read and re-read to increase familiarity with the data prior to coding. Codes were organised into descriptive themes that remained close to the original data. The final phase involved moving beyond the categorisation of descriptive themes and onto the development of analytical themes.

3 FINDINGS

Four analytical themes were developed describing the factors impacting on pwMND decisions about gastrostomy and ventilation: ‘An emotional response to interventions’; ‘Sharing the decision with others’; ‘Control’; and ‘Tipping the balance’ (figure 2). The analytical themes describe the intrinsic, contextual and relational factors that influence how and when pwMND make decisions about interventions. Verbatim quotes, punctuated with single punctuation marks, taken from the original papers are included to support the analytical findings, with the source of quotes indicated by pwMND (P), caregiver (C), HCP (H) or researcher (R). Table 3 describes the contribution each paper made to the analytical themes.

FIGURE 2 A summary of the four analytical themes (in capitals and bold) and subthemes (boxes with dashed lines) developed as a result of the thematic synthesis.

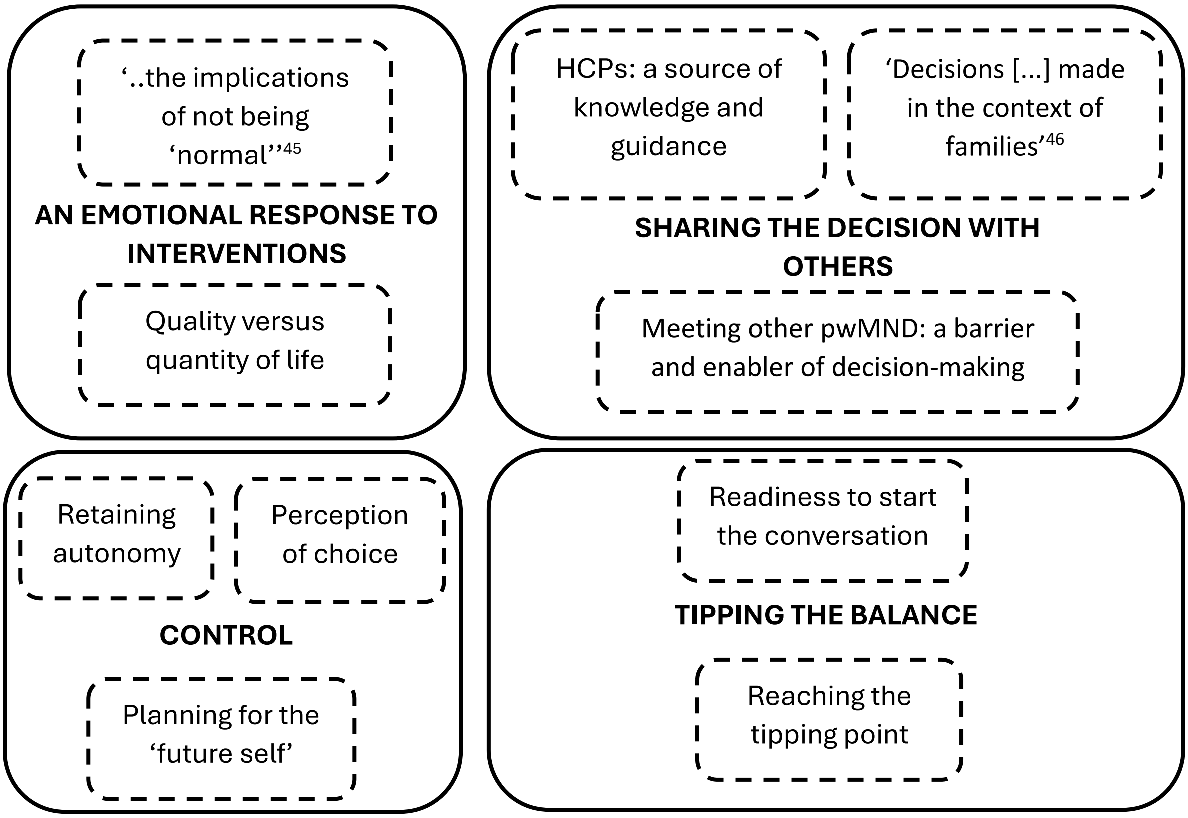


TABLE 3 The contribution made by each paper to the themes.

Study	An emotional response to interventions		Sharing the decision with others			Control			Tipping the balance	
	'...the implications of not being 'normal''[45]	Quality versus quantity of life	HCPs: a source of knowledge and guidance	'Decisions [...] made in the context of families'[46]	Meeting other pwMND: a barrier and enabler of decision-making	Retaining autonomy over decisions	When the perception of choice runs out	Planning for the 'future self'	Readiness to start the conversation	Reaching the tipping point
Ando *et al.,* (2015)[32]	✓	✓	✓		✓	✓				✓

Chapman *et al.*, (2021)[25]			✓					✓		✓
Clarke *et al.*, (2018)[53]				✓				✓		
Foley *et al.*, (2014a)[38]						✓		✓	✓	✓
Foley *et al.*, (2014b)[39]		✓				✓		✓		
Foley *et al.*, (2016)[40]		✓		✓						

Gottberg *et al.*, (2021)[54]				✓		✓	✓			✓
Greenaway *et al.*, (2015)[48]	✓	✓	✓	✓		✓	✓	✓	✓	✓
Hirano and Yamazaki (2010)[50]	✓	✓	✓		✓		✓		✓	✓
Hodgins *et al.*, 2020[55]						✓		✓		
Hogden *et al.*, (2012a)[17]			✓						✓	
Labra *et al.*, (2020)[33]	✓	✓						✓		✓

Lemoignan and Ells (2010)[34]	✓	✓	✓	✓		✓	✓	✓	✓	✓
Leslie (2008)[30]	✓	✓	✓	✓		✓			✓	✓
Martin *et al.,* (2016)[46]	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
Murray *et al.,* (2016)[49]	✓				✓	✓		✓		
Nolan *et al.,* (2008)[57]								✓		
Paynter *et al.,* (2020)[35]	✓	✓	✓				✓		✓	✓

Pols and Limburg (2016)[41]	✓	✓	✓					✓	✓	✓
Sakellariou (2016)[47]	✓					✓	✓			
Seeber *et al.,* (2019)[36]			✓						✓	✓
Stavroulakis *et al.,* (2014)[45]	✓		✓		✓	✓	✓	✓	✓	✓
Sundling *et al.,* (2009)[56]						✓				
Veronese *et al.,* (2014)[52]		✓				✓	✓			✓

Versalovic and Klein (2020)[31]								✓		
Whitehead *et al.,* (2011)[51]	✓			✓	✓	✓	✓	✓		
Young *et al.,* (1994)[37]	✓	✓				✓		✓		

3.1 An emotional response to interventions

The prospect of starting an intervention represented a threat to pwMND sense of normality including the implications for their level of independence, freedom, and reliance on others. Additionally, pwMND perceptions were influenced by how they value an interventions' potential to prolong life and the quality of that remaining life.

3.1.1 '...the implications of not being 'normal''[45]

pwMND deliberations about whether to commence interventions extend beyond the clinical problems they are proposed to ameliorate. Decision-making is influenced by a range of emotional responses with regards to what the interventions signify and how daily life with them will compare to the current status quo.

Discussions about interventions acted as an acute reminder for some pwMND of disease progression.[32,34,35,41,46,47] Acceptance of the need for interventions was associated with giving in to the disease[46] and signified a stage of the disease associated with a poor quality of life or nearing the end of life.[32,34,46]

> (H) *'...A very obvious sign of being ill...a very concrete representation that he's seriously ill.'*[46]

An emotional response was generated when learning about the procedures involved[30,35,41,45,46,48], a requirement to come into hospital[32,33,46] and threats to pwMND physical integrity[30,32,41,48,49]. For some pwMND their response to the prospect of interventions was influenced by previous negative healthcare experiences.[32,46,48]

Additionally, pwMND were concerned about how interventions may impact on their current sense of normality. pwMND reported contrasting views about how an intervention may enable or hinder their ability to engage in social activities[30,32,37,46] or alter their reliance on care from others[33,34,37,46,50,51]. While some pwMND were able to visualise the gains interventions could facilitate in terms of freeing time or reducing burden on caregivers[33,41,45,46], most studies reported how they represented changes to 'normal life' that they were unwilling to accept easily.

> (R) '*All participants talked about the affect of QOL (including the ability to communicate, eat, move around, and be surrounded by loved ones) on decisions about assisted ventilation.*'[34]

3.1.2 Quality versus quantity of life

Existential views on the potential for interventions to prolong life were integrated into pwMND decision-making. A number of studies reported pwMND preference to prioritise quality of life over prolonging a life that lacked value to them.[32-34,39,46,48] Knowledge about the terminal phases of MND could even bring about a preference to actively shorten life through refusing an intervention.[33,37]

> (R) '*Most participants suggested that losing independence rendered life less meaningful and that life-sustaining interventions had the potential to prolong 'suffering'.*'[39]

Reassurance from others and an overriding belief that quality of life will be maintained or improved, promoted a positive perception of interventions.[30,33-35,41,46,48,50] While some HCPs discussed interventions in relation to the impact on quality of life[46], there were also examples of HCPs goals focusing on clinical outcomes.

> (R) '*...the sensual qualities of the body surprised the gastroenterologist, who saw maintaining physical fitness and longer survival as the ultimate goals, and the feeding tube as a means towards this end…*'[41]

pwMND reported considering how any extension to their lives may impact on their caregivers. Interventions were perceived to lengthen the time they are a burden on caregivers[33] or, in contrast, facilitated them to be with or support family for longer[41,46,48,50]. For some pwMND extending life was an overriding goal, particularly when faced with a direct threat to life such as in crisis situations.[34,35,37,39,46,48,52]

> (P) '*Any living person's will to survive is primordial and outweighs many other concerns or reasons.*'[34]

pwMND perceptions of interventions are both intrinsically generated and the consequence of interactions with multiple external agents. The following theme captures how HCPs, caregivers and other pwMND can influence the decision-making process.

3.2 Sharing the decision with others

pwMND decision-making involves interactions with a range of external agents. HCPs share information and attempt to guide pwMND through the decision; caregivers share some of the burden of decision-making with pwMND; and knowledge of other pwMND can help clarify or challenge perceptions and values.

3.2.1 HCPs: a source of knowledge and guidance

The style, amount, and pace of information shared by HCPs influenced pwMND experience of the decision-making process. HCPs focusing on delivering comprehensive information about interventions, may prevent pwMND from having the opportunity to understand and explore the issues most important to them.[25,48] Some HCPs reported following a structured approach to supporting pwMND decision-making through seeking permission to initiate discussions, presenting options available, exploring preferences and distributing discussions over multiple interactions.[30,34,41] HCPs using a respectful, personable communication style including empathy and reassurance was valued by pwMND.[48]

> (C) '*So, you know, some health professionals – I think periods of silence to listen to the client are needed without the health professional thinking, "I've got to tell them X, Y, Z…"'*[25]

HCPs were a trusted source of information and pwMND expected them to use their expertise to guide decision-making.[30,45,46,48,50] However, credibility of healthcare services was threatened by a perceived lack of HCP knowledge[48], poor experiences of healthcare services[32], or a lack of clear guidance[48]. A breakdown in trust in HCPs or healthcare services may lead to disengagement of pwMND from the decision-making process.

pwMND reported having multiple interactions with different HCP roles when considering interventions.[25,30,36,45,46,48,50] HCPs expressed challenges with the MDT coordinating decision support including pwMND being given contradictory information from different team members[25,46,48] and poor communication between services[17,25,35].

> (H) '*I think my…fear is you can get so many teams of people involved. [...] the possibilities of confusion and misinformation are enormous...I was going to say warfare [can occur].*'[25]

3.2.2 ‘Decisions [...] made in the context of families’[46]

Caregivers were reported to play a supportive role, through seeking information, expressing opinions and deliberating about options.[30,34,40,48] Through these interactions caregivers *‘influenced not only the decision but also the process of decision-making*’.[46] Caregivers required information early in the disease process to facilitate their role supporting pwMND decision-making.[34,53] While caregivers recognised the pwMND right to self-determination, their contributions to the decision-making process were not always described as being neutral and were informed by their own preferences and acceptance of the diagnosis.[46,48,54] pwMND preferences for or against interventions were challenged when caregivers preferences were not aligned with those of the pwMND.[40,46,48,51]

> (R) ‘*Some patients experienced their families more negatively, with the family members’ emotional needs taking precedence over those of the patient.*’[48]

3.2.3 Meeting other pwMND: a barrier and enabler of decision-making

Understanding experiences of other pwMND facilitated some pwMND to clarify their own preferences, including learning about pwMND reflections of delaying decision-making, reassurance that they could adapt to life with an intervention and informing values with regards to prolonging life.[32,45,46,49,50]

> (P) ‘*The decisive factor was meeting ALS patients already using it. These people with the same disease had a positive outlook on life, and this gave me the will to live as well. My uncertainty disappeared.*’[32]

However, others reported meeting other pwMND would have been a barrier to decision-making, reminding them about the future symptom burden they would be living with and may wish to avoid.[50,51]

The involvement of multiple external agents raises the question of who is responsible for making decisions and how interactions with others may influence the loci of control.

3.3 Control

pwMND wanted to be in control of the decisions they made about interventions; a preference that could be facilitated or threatened by interactions with others.

3.3.1 Retaining autonomy over decisions

pwMND consistently reported a preference to remain in control over decisions about interventions.[30,32,34,37,38,46,48,49] In line with pwMND preferences, HCPs and caregivers sought to protect pwMND right to self-determination and empower pwMND to make their own decisions.[30,45,46,55]

> (P) '*John [dietician] came back out again and said as you get nearer for the PEG, don't let anybody bully you into it.*'[30]

This quote points to the possibility that external agents may influence pwMND ability to exert autonomy during decision-making, including convincing pwMND to make decisions that are not aligned with their preferences; a scenario that was described in a number of papers.[32,34,45-49,51,54,56] pwMND reported the perception of feeling pressured to accept interventions by HCPs was accentuated when approaches were repeated and from multiple different sources.[32,39,48,49,56]

> (P) '*I mean the speech therapist came round, she said 'well something you've got to start thinking about is this pipe' and [MND Specialist Nurse] came round and she said about it and that other woman said something about it an' all. It felt like a lot of pressure was being put on me*'[48]

In some studies, there was a sense of 'side taking', with HCPs and caregivers joining forces to project their own preferences for interventions.[46,52] Some HCPs reported finding it challenging to present neutral information, feeling a responsibility to advocate for the timely uptake of interventions.[46]

3.3.2 When the perception of choice runs out

pwMND perception of having choice about interventions was narrowed or removed by HCPs limiting the options presented or the significance of the health threat presented by the disease. While HCPs reported presenting options to commence or decline interventions, there were also examples where HCPs withheld options[46,50,51] or framed alternatives in a way to make them not even appear to be a choice[47,52].

> (H) *'I haven't discussed noninvasive ventilation because...how would he ever cope? So, I've made, on best interests, not to start discussing those issues....His anxiety, it's just going to raise his anxieties.'*[46]

pwMND were able to enact agency over decisions while they were coping with symptoms and choosing to delay or decline interventions. However, when symptoms presented a significant threat to pwMND health or life, interventions were often perceived to be the only choice available to prevent serious consequences.[34,35,45,46,48,50,52,54]

3.3.3 Planning for the 'future self'

Decision-making often involved pwMND attempting to visualise how their future self may value an intervention. pwMND reported struggling to imagine how they would feel about an intervention when living with increased disease burden.[31,34,41,45] Acceptance of disease progression was associated with pwMND planning ahead through either commencing interventions prior to them being needed or making advanced decisions to decline interventions altogether.[33,34,38,45,46,48,49,53,57] Planning ahead could allow pwMND to remain in control of their destiny, including those at risk of cognitive decline.[25,37,46,55] However, other pwMND feared that interventions would be commenced or continued despite them experiencing a poor quality of life.[37,39,46,51]

pwMND wanted to remain in control of decision-making. Interactions with HCPs and caregivers influenced pwMND perception choice and agency over their decisions. The timing of interventions is a challenging issue for all involved.

3.4 Tipping the balance

The timing of initiating discussions and commencing interventions was a source of conflict for all stakeholders. Commencing interventions often required pwMND to reach a subjective tipping point informed by disease progression, acceptance of need and recommendations of HCPs.

3.4.1 Readiness to start the conversation

While HCPs advocated for an individualised patient-centred approach, HCPs supported introducing intervention options early during the disease course.[17,46] In addition to referring to signs of disease progression, HCPs reported making subjective assessments of pwMND psychological readiness to discuss interventions.[36,46] Some pwMND reached crisis point without having the opportunity to understand their options.[50] While early discussions about interventions allowed more time to consider the options[30,41,45,46,48], for some pwMND having these discussions conflicted with a personal preference for focusing on the present day challenges of the disease[30,34,35,38,41,45,48].

> (P) '*No, no, no. I'd rather not know until there's a reason to know. I don't want to know all the nasty possibilities that might be in front of me. You know, I just don't want to know the detail, until there's a reason to know it*'[48]

3.4.2 Reaching the tipping point

With regards to the timing of interventions, HCP preferences for 'earlier rather than later' were informed by previous clinical experience of poor outcomes associated with late intervention.[31,33,35,41,45,46,48] pwMND perception of intervention need can deviate from that of HCPs, preferring to continue to cope with symptoms rather than introduce an intervention into their lives.[32,36,38,48] The tipping point was defined by acceptance of disease progression, experience of adverse consequences related to the health threat or recognition that the benefits of an intervention now outweigh the risks.[25,30,31,33,34,41,45,46,48,50,52,54]

> (P) '*It was a gradual change in my decision. As swallowing got worse I couldn't drink water and thickened stuff was not pleasant. So I thought to be hydrated in summer was a good idea and also taking medications would be easier through the tube.*'[33]

pwMND who perceived that their symptoms were not causing significant problems could continue to defer decisions about interventions, even when presented with objective measures of functional decline or the direct observations of others.[32,35,46] The uncertain rate of disease progression made it difficult for pwMND to identify the 'right time' to start an intervention and could limit the time available to make decisions especially when there was a serious health threat.[35,36,45,46,50,52,54]

> (C) '*You don't know how it's going to proceed…whether it's going to proceed quickly…the uncertainty makes it far more difficult to make decisions because you don't know what tomorrow's going to bring. I mean I'm sure [patient] would find it*

> *easier, if she knew how it was going to progress and if we had some idea of timescale.'*[45]

HCP guidance and recommendations were reported to influence the timing of interventions.[30,33,35,45,46,48] Some pwMND found information about timing of interventions inadequate and expected HCPs to use their experience to provide more concrete guidance.[45,48] However, as described earlier, the approach taken by HCPs when re-visiting decisions about interventions can result in pwMND feeling pressurised to commence interventions or change previous decisions.

> (C) '*They say make sure it's done sooner rather than later but what is sooner rather than later? They don't say we're talking next month, no definite time, it's down to you*'[48]

4 DISCUSSION

4.1 Summary of findings

The review findings describe how decisions about interventions generate an emotional response that extends beyond consideration of the functional issues and are the consequence of interactions with a range of external agents. pwMND reflect on how life with an intervention will alter their independence, freedom and survival compared to continuing with the status quo, including concern for the impact on their caregivers. HCPs and caregivers played an integral role during decision-making, through supporting pwMND to understand their options, retain control and make decisions aligned with their values. The sense of choice and agency experienced by pwMND during decision-making was mediated by disease progression and the actions of others. The timing of interventions is a source of uncertainty, with decisions being driven by symptom severity, HCP guidance and pwMND acceptance of need for intervention.

4.2 The emotional response to interventions

Most studies reported pwMND associating interventions with a life with increased restrictions, burden and reliance on others. These findings are consistent with Foley *et al.,*'s (2007) exploration of the 'meaning of quality of life' for pwMND, where participants reported an emotional response to the losses experienced due to the disease and fought to maintain normality and their own identity.[58] Discussions about interventions signified a step-change in disease progression requiring pwMND to undertake existential deliberations about the value of artificially prolonging their life. Though pwMND report prolonging life to be a driver for accepting interventions, many base decisions on the acceptability of the predicted quality of that life, reflecting on how interventions will impact on their lives and those of their caregivers in the present and the future. Aligned with previous research and theory of human behaviour, emotional responses (e.g. fear and anxiety) to the implications of an intervention may result in disengagement from decision-making discussions or deferring the making of a decision.[18,59] These findings highlight the importance of actively exploring pwMND representations of interventions. Engagement in decision-making discussions may be facilitated through psychological interventions that identify and address the emotional response to interventions.[59-62]

4.3 Intervention timing

Following the decision about 'if' they should accept intervention, pwMND are faced with the uncertainty about 'when' to commence the intervention. The review findings describe how decisions about timing of interventions are influenced by pwMND acceptance or understanding of need, and their response to HCP guidance. Decisions about the timing of interventions are important, with delays being associated with poorer outcomes, particularly in relation to gastrostomy placement.[11,63-65] Such studies underpin the rationale for discussing and commencing interventions earlier in the disease course.[7-9,66] However, pwMND decisions are not informed solely by deteriorating clinical markers but also how aligned outcomes of an intervention are with pwMND values and priorities including the perceived impact on caregivers.

The perceptions of the disease and interventions are informed by an iterative updating of illness cognitions in response to pwMND evolving acceptance and adaptation to the relentless changes in their condition.[16,61,67,68] The review confirms that pwMND need support to understand how they will perceive living with the increased symptom burden associated with disease progression. Such support may enable pwMND to accept the need for interventions earlier or, conversely, facilitate informed decisions about delaying or declining interventions.

4.4 Autonomy and control

The review captured how external agents can facilitate individual autonomy[69,70], an ethical principle valued by pwMND, through protecting the right to self-determination and provision of information. Caregivers play a multi-layered role during decision-making. Consistent with a critique of autonomy in end-of-life care, pwMND made decisions in collaboration with and through concern for their caregivers.[71] When pwMND decisions are based on concern for caregiver burden or prioritisation of caregiver preferences, an individualistic conceptualisation of autonomy could be perceived to be threatened. The social and interactional context in which pwMND make decisions in collaboration with caregivers may be better aligned with a relational conceptualisation of autonomy and is consistent with the principles of shared decision-making.[71,72] However, a line is crossed when dialogue with external agents disrupts the patient's preference to make decisions based on their own values.[72] This was evident in the review with pwMND sometimes feeling pressurised to accept interventions by caregivers and HCPs.

HCPs have an ethical and professional obligation to support pwMND to make autonomous informed values-based decisions about interventions.[69,73,74] The contrasting preferences of pwMND for, and emotional responses to, disease and intervention related information present a challenge for HCPs attempting to balance the ethical principles of beneficence and autonomy.[69,75] Despite being motivated by a responsibility to promote positive health outcomes, multiple HCPs repeatedly revisiting discussions placed some pwMND under pressure to accept interventions. Conversely, pwMND can expect HCPs to use their expertise to provide direction about if, and when, to start an intervention. While taking what may be considered a more paternalistic approach risks paying less attention to pwMND values[76], HCP recommendations can support patients to navigate the uncertainty present during decision-making[77-79]. HCPs could positively contribute to relational autonomy by ensuring recommendations are grounded in a knowledge of pwMND preferences, goals and values.[75]

Consistent with a previous review[21] only one study[55] referred to the impact cognitive impairment may have on pwMND decision-making. Cognitive impairment, which is common in MND[80,81], has been identified as a barrier to autonomous decision-making[19], and associated with reduced acceptance[82] or delayed starting[17] of interventions. Further research is required to explore how pwMND with mild-moderate cognitive deficits can be supported to engage in decision-making, and elicit pwMND preferences (i.e. advanced decisions) in the context of fluctuating and deteriorating mental capacity to make their own decisions.

4.5 Strengths and limitations

A limitation of this review was the absence of a second reviewer to shortlist the references for inclusion, extract data and independently code the data which may limit the credibility of the study. However, this is mitigated to a degree by some of the methodological strengths of the review such as the systematic approach to the study design, including a comprehensive bibliographic and supplementary search strategy, a second reviewer screening 10% of references at the abstract and full text stages and discussions with supervisory team when developing the analytical themes.

A further limitation relates to the review not making any distinctions between decisions made about gastrostomy, non-invasive ventilation and invasive ventilation. These are interventions, with differing clinical indications, outcomes and implications for pwMND and their caregivers. There may have been valuable insights gained from exploring the consistencies and differences of the decision-making process for the different interventions. A review of the data did not reveal enough rich data to provide a credible account of any such differences. Future qualitative research may benefit from comparing the contextual factors that influence decisions about different interventions to generate deeper analytical insights.

4.6 Reflexivity

SW is a dietitian with experience of supporting pwMND to make decisions about gastrostomy placement. Prior experience and opinions of a researcher have the potential to affect how data are analysed and interpreted.[83] Any such influence was mediated through staying close to the primary studies, maintaining a clear audit trail of the analysis process, and regular discussions with the research team about the decisions made during the screening process and analysis.

4.7 Implications for practice

The emotional response to interventions highlights the importance of exploring the value pwMND place on the implications and outcomes of an intervention beyond the concern for clinical outcomes. Decision-making should address the issues that matter most to pwMND including the existential debate pwMND have about an intervention's potential to affect their quality and length of life. While HCPs have an ethical responsibility to communicate with pwMND about the predicted disease course and timely consideration of interventions caution should be taken to respect individuals' preferences for information and control. Acknowledging early in the decision-making process, the contribution pwMND, caregivers and HCPs make towards a relational conceptualisation of autonomy may facilitate a shared understanding of each other's role and boundaries, and promote meaningful engagement in decision -making discussions.[71]

5 CONCLUSION

Addressing the emotional response pwMND have to the prospect of interventions could improve engagement in decision-making discussions. HCPs and caregivers have a complex ethical path to navigate when seeking to protect pwMND autonomy while simultaneously attempting to optimise health outcomes. Decision support strategies that mediate the emotional response and promote autonomy have the potential to enable pwMND to make timely, values-based decisions about interventions.

ACKNOWLEDGEMENTS

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CONFLICTS OF INTEREST

The authors have no conflicts of interest to declare relating to this study.

STATEMENT OF AUTHORS' CONTRIBUTIONS

Removed to maintain anonymity. Added to title page.

DATA AVAILABILITY STATEMENT

No primary data was generated, analysed or stored as part of this study.

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4.5 Summary of the chapter

This chapter has presented the design, conduct and findings of a qualitative evidence synthesis answering the research question:

> ‘What are the organisational, team and individual level factors that influence decision-making by people with MND when making the decision to start an invasive healthcare intervention?’

The findings are summarised below:

- Decisions about interventions are associated with an emotional response by people with MND.
- The prospect of interventions signals an end to normality and increasing dependence on others.
- People with MND undertake an existential debate about the value and consequences of extending their lives through engagement with life-prolonging interventions.
- Responsibility for decision-making is shared with caregivers and healthcare professionals.
- Caregivers and healthcare professionals can promote and threaten control and autonomy people with MND have over their decisions.
- Having many healthcare professionals involved in supporting people with MND decision making can cause confusion.
- The timing of interventions is associated with uncertainty.
- Symptom burden and healthcare professional guidance are drivers for people with MND making decisions.

5 HCP survey (Paper 2)

5.1 Chapter outline

The following chapter presents the publication of a survey that aimed to understand healthcare professionals practice and beliefs in relation to supporting people with MND making decisions about gastrostomy placement. A summary of the methods used are described with further details and justifications included in Chapter 3, section 3.8. The key findings from the survey are presented.

5.2 HCP survey research question

The survey answered the following research question:

> ‘How do healthcare professionals support people with MND to make decisions about gastrostomy placement, as individuals and as a collective unit of MDT decision support?’

5.3 Further information related to the study

Further information relating to the design and conduct of this component of the study is included in the following appendices:

- Appendix 6. Conceptualisation of the survey research questions
- Appendix 13. The full survey data set
- Appendix 14. Survey proforma
- Appendix 15. CHERRIES checklist
- Appendix 16. List of gatekeepers who agreed to distribute the survey link
- Appendix 17. Characteristics of the survey participants
- Appendix 18. Formation of new categories about the timing of gastrostomy placement in relation to the severity of dysphagia and respiratory failure
- Appendix 19 The topics discussed by the different professions in relation to the placement of a gastrostomy and life on enteral nutrition

5.4 Publication details

The paper has been published in Amyotrophic Lateral Sclerosis and Frontotemporal Degeneration journal under the Creative Commons Attribution License (CC BY). As such I, as the author, have the right to copy and redistribute the manuscript in any medium or format for any purpose, including for inclusion in a thesis. The citation for the published paper is:

> White, S., O'Cathain, A., Halliday, V., Bradburn, M., & McDermott, C. J. (2024). Supporting people with Motor Neuron Disease (MND) to make decisions about gastrostomy feeding tube placement: a survey of UK healthcare professionals' practice and beliefs. *Amyotrophic Lateral Sclerosis and Frontotemporal Degeneration*, *25*(3–4), 290–298. https://doi.org/10.1080/21678421.2024.2314061

5.5 Paper 2: Supporting people with Motor Neuron Disease (MND) to make decisions about gastrostomy feeding tube placement: a survey of UK healthcare professionals' practice and beliefs

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Abstract

Objective: Understand the practice and beliefs of healthcare professionals (HCPs) supporting the decision-making of people with MND (pwMND) about gastrostomy placement, including identifying differences between professions.

Methods: An online cross-sectional survey disseminated to HCPs who support the decision-making of pwMND about gastrostomy placement.

Results: A total of 139 participants completed the survey including representation from a range of healthcare professions. A third (36/101, 36%) initiated discussions about gastrostomy later in practice than they believed was ideal. In relation to the outcome of declining compared to accepting gastrostomy, participants were more likely to discuss aspiration (80% vs 68%), choking (76% vs 58%) and prognosis (36% vs 22%). Participants believed gastrostomies should be placed after a mean 8.1% weight loss since symptom-onset. More participants favoured gastrostomy placement before pwMND presented with respiratory symptoms (45%) compared to onset of dysphagia (11%). Half believed pwMND placed gastrostomies too late. Participants were more likely to 'often'/'always' recommend pwMND to have a gastrostomy (23%) than continue without (7%) or decline (4%) gastrostomy, when believing these were the best option for pwMND. Nurses and dietitians discussed the broadest range of information, while doctors were more likely to discuss mortality risk and prognosis.

Conclusion: There is variation in HCPs practice and beliefs about initiating discussions, the sharing of information and recommendations, and timing, about gastrostomy placement. The information shared varies by profession and there is evidence of sub-optimal communication between HCPs. Further research is required to understand how these findings may impact on the decision-making of pwMND about gastrostomy.

Keywords: motor neuron disease, amyotrophic lateral sclerosis, gastrostomy, decision-making, survey, multidisciplinary team

Introduction

People living with motor neuron disease (pwMND) face making many decisions throughout their disease course. These decisions are made within the context of experiencing a progressive loss of function, limited treatment options and an average survival of 2-4 years. In the absence of a cure, therapeutic options focus on compensating for functional losses, including dysphagia and respiratory failure (1,2,3). Between 44-85% of pwMND experience dysphagia one year after diagnosis, contributing to a high risk of malnutrition and aspiration (4,5). Malnutrition is an independent prognostic indicator (6,7) and aspiration pneumonia was the reported cause of death in 19% of pwMND (8). Although evidence for improved quality of life or survival remains equivocal (9), gastrostomy placement is routinely offered to pwMND to address the risk of malnutrition or aspiration (10,11,12).

Guidelines recommend that healthcare professionals (HCPs) collaborate with patients to develop a shared understanding of the benefits and risks of intervention options available and make decisions that are consistent with the patient's values and preferences (13,14). An emerging qualitative evidence base has captured the contextual factors that influence how, when and why pwMND make decisions about gastrostomy placement (15,16). HCPs are a valued source of information about the disease and interventions for pwMND and can be expected to use their experience and knowledge to guide decision-making (17). Decisions about gastrostomy placement are rarely made between a single HCP and person with MND, in a single interaction. More often, decision-making is distributed over time including interactions with multiple HCPs, caregivers and other sources of information (18,19,20). A distributed process of decision-making challenges the multidisciplinary team (MDT) to deliver consistent decision support (21,22,23,24). There is a paucity of research focusing on the practice and beliefs of different healthcare professions, across key stages of decision-making.

The objectives of this study were to:

(1) Understand UK HCPs' practice and beliefs in relation to supporting pwMND at key stages of decision-making including when discussions about gastrostomy are first initiated, the sharing of information and when decisions about gastrostomy are made.

(2) Identify any differences in the practice and beliefs of different professions involved in supporting the decision-making of pwMND.

Methods

Study design

A cross-sectional online survey, enabling a broad reach to the different healthcare professions involved in supporting the decision-making of pwMND about gastrostomy placement was chosen as an appropriate study design (25,26,27). The study report has been guided by the Checklist for Reporting Results of Internet E-Surveys (CHERRIES) for reporting internet e-surveys (28) (see supplementary information 1 (Appendix 15 in this thesis).

Sample and sampling

Four healthcare professions are known to be routinely involved in discussions with pwMND about gastrostomy placement: doctors, dietitians, speech and language therapists (SLTs) and nurses. A snowball sampling strategy was used to recruit a convenience sample, representative of those professions involved in these discussions (29).

Survey development

The questionnaire was developed using a sequential approach (27,30) including using the findings from a qualitative evidence synthesis (16) and discussion with the research team (AO, VH, CMJ), topic experts and a patient panel. A draft survey was refined in response to feedback from 9 HCPs who participated in a pilot to produce a 56-item questionnaire hosted on Qualtrics online survey platform (Qualtrics, Provo, UT) (see supplementary information 2 for the full questionnaire (appendix 14 in this thesis).

Data Collection

Key gatekeepers, including the MND Association and relevant HCP organisations, agreed to distribute the questionnaire (see supplementary information 3 (appendix 16 in this thesis) for a list of gatekeepers) through their communication channels. Reminders were sent every two weeks between 13/6/2022 and 30/8/2022. Participants were asked to read a participant information sheet, answer screening questions, and complete a consent form embedded in the online survey, before proceeding to complete the questionnaire.

Analysis

Descriptive statistics were used to analyse the data set using IBM SPSS Statistics (Version: 28.0.0.0 (190)). For some findings, response categories have been combined to allow comparisons between related questions. Cross-tabulations were undertaken to identify differences between the responses from doctors, dietitians, SLTs and nurses. The Chi squared test was applied to identify differences in responses for binary outcomes. McNemar's paired test of symmetry (for binary outcomes) or marginal homogeneity (for ordinal outcomes) were applied to identify differences between the paired responses for related questions.

Ethics

The study received approval from the University of Sheffield ethics panel (reference: 210151905) and Health Research Authority (HRA) (reference: 308744).

Results

Participant characteristics

Of the 212 HCPs confirming they met the inclusion criteria, 139 participants completed the survey including 73 dietitians (53%), 23 nurses (17%), 19 SLTs (14%), 17 doctors (12%), 4 physiotherapists (3%) and 3 occupational therapists (2%). The participants had a mean 11.1 years (SD 8.3, range 1-39) experience of caring for pwMND and the majority were female (123/139, 89%). Participants most frequently (86/138, 62%) responded that <20% of their caseload were pwMND. Discussion about gastrostomy most commonly took place in the homes of pwMND (92/139, 66%), telephone calls (69/139, 50%) and hospital outpatient clinics (60/139, 43%). Participant characteristics are summarised in table 1 (see supplementary information 4 (appendix 17 in this thesis) for a full description of participant characteristics). The following findings present the participant beliefs and practice in relation to supporting pwMND to make decisions about gastrostomy placement (see supplementary information 5 for the full findings(Appendix 13 in this thesis)).

Characteristic	n (%)
Gender (N=139)	
Female	123 (89)
Male	14 (10)
Prefer not to say	2 (1)
Healthcare profession (N=139)	
Dietitian	73 (53)
Nurse	23 (17)
Speech and language therapist	19 (14)
Doctor	17 (12)
Physiotherapist	4 (3)
Occupational therapist	3 (2)
Number of pwMND on caseload (N=138)	
0-10	61 (44)
11-20	30 (22)
21-30	16 (12)
31-100	18 (13)
>100	13 (9)
Percentage of clinical caseload that includes pwMND (N=138)	
0-19%	86 (62)
20-39%	22 (16)
40-99%	16 (12)

100%	14 (10)
Number of years experience caring for pwMND (N=83)	Mean 11.1 years (SD 8.3; Range 1-39)
Setting in which discussions about gastrostomy placement occur (N=139)	
Domiciliary visit	92 (66)
Telephone calls	69 (50)
Hospital based out-patient clinic	60 (43)
Hospital ward	59 (42)
Video call	39 (28)
Community based out-patient clinic	36 (26)
Hospice based out-patient clinic	20 (14)
Hospice in-patient	3 (2)
Other	2 (1)

Table 1. Participant characteristics

Stage 1: Initiating discussions about gastrostomy placement with pwMND

Of the participants (115/139, 83%) stating they initiated discussions about gastrostomy placement with pwMND, most were prompted by pwMND presenting with swallowing difficulties (114/115, 99%) and weight loss (105/115, 91%). A similar proportion of participants initiated discussions about gastrostomy placement prior to (42/115, 37%), or following (51/115, 44%) the first presentation of any clinical indications (e.g. dysphagia, weight loss). A third (36/101, 36%) of participants believed they initiated discussions later in practice than was ideal.

Stage 2: Sharing information with pwMND

Outcomes of gastrostomy placement

There were differences with regards to the potential outcomes participants would address with pwMND, when discussing either the option to **accept** or **decline** gastrostomy placement (see table 2). When discussing the potential outcomes of **declining** gastrostomy placement, participants were more likely to report they discuss the risk of aspiration (80% vs 68%; p= 0.018), choking (76% vs 58%; p=0.0002) and prognosis (36% vs 22%; p=0.0002). When discussing the potential outcomes of **accepting** gastrostomy placement, participants were more likely to report they discuss quality of life (83% vs 75%; p=0.041) and impact on caregivers (63% vs 50%; p=0.007).

Outcome of gastrostomy placement discussed	**Discussed in relation to accepting gastrostomy (n (%)) N=139**	**Discussed in relation to declining gastrostomy (n (%)) N=139**	**p value**
Impact on the person's weight	111 (80%)	109 (78%)	0.706
Risk of aspiration	95 (68%)	111 (80%)	**0.018**
Risk of choking	80 (58%)	106 (76%)	**0.0002**
Impact on quality of life	115 (83%)	104 (75%)	**0.041**
Time taken to finish meals	98 (71%)	87 (63%)	0.071
Estimated length of life remaining (prognosis)	30 (22%)	50 (36%)	**0.0002**

Impact on caregivers (e.g. family)	87 (63%)	70 (50%)	**0.007**

Table 2. The outcomes discussed with pwMND when discussing accepting or declining gastrostomy placement. Significant differences between the proportion of participants who would discuss outcome in relation to accepting and declining gastrostomy indicated in **bold** (McNemar's paired test of symmetry $p \leqslant 0.05$).

Information about gastrostomy placement and life on enteral feeding

When discussing gastrostomy placement, three quarters of participants included details about the procedure (108/139, 77%) and expected length of hospital stay (103/139, 74%). The risk of not surviving the procedure was reported to be discussed by the lowest proportion of participants (54/139, 39%). In relation to life on enteral feeding, enteral feeding methods (119/139, 86%) and the support pwMND will receive from HCPs (118/139, 85%) were discussed by the most participants. Gastrointestinal side-effects were reported as the least discussed (60/139, 43%). There was a varied response in relation to how frequently participants would discuss the option to withdraw enteral feeding in the future at the time of decision-making (never/rarely: 53/139, 38%; sometimes: 43/139, 31%; often/always: 43/139, 31%).

Stage 3: Making the decision about gastrostomy placement

HCP recommendations about gastrostomy placement

Participants reported a varied belief and practice in relation to giving recommendations to pwMND about whether or not to have a gastrostomy placed:

- Half of participants (72/139, 52%) stated they believed HCPs have a responsibility to give pwMND recommendations about whether or not to have a gastrostomy placed.
- Half of participants (71/139, 51%) stated they 'never' or 'rarely' give pwMND such recommendations in practice.

Participants were asked how frequently they would give pwMND a recommendation to: 1. have a gastrostomy placed; 2. continue without a gastrostomy or; 3. never have a gastrostomy; **when** the participant believed one of these was the best option for a pwMND to take. Participants most commonly responded that they ‘never’ or ‘rarely’ made recommendations but were significantly more likely to state they ‘often’ or ‘always’ recommend pwMND to have a gastrostomy (31/136, 23%) than to continue without (10/137, 7%) or to never have a gastrostomy placed (6/135, 4%) (figure 1); p<0.001 for all pairwise comparisons.

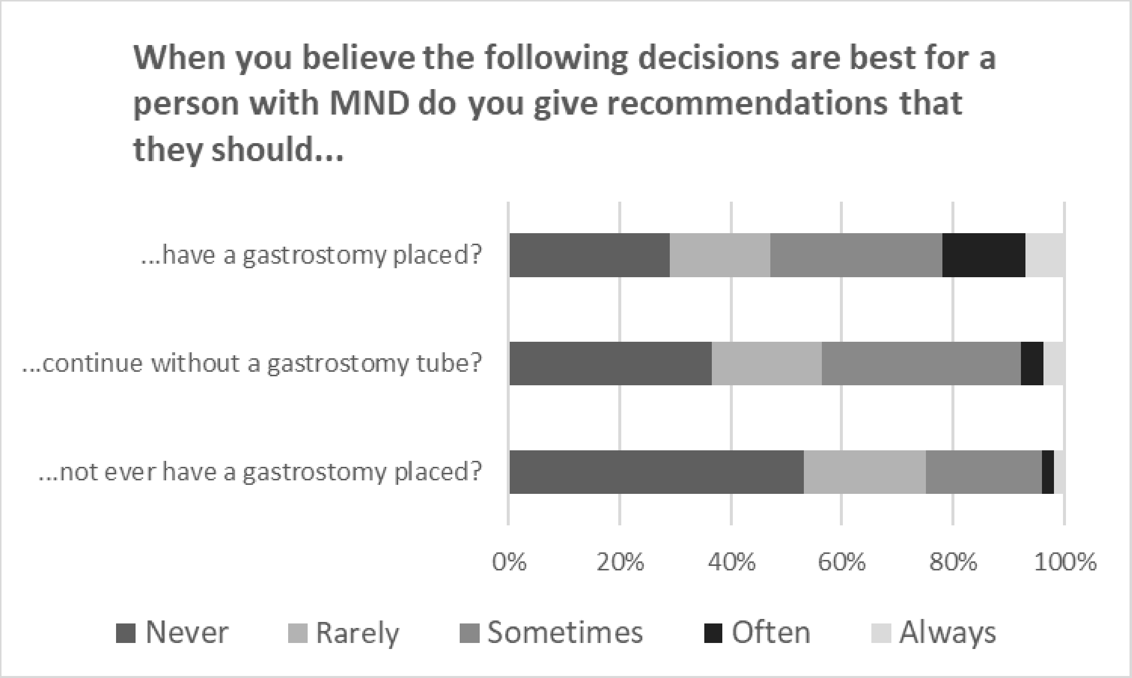

Figure 1. The frequency with which participants would give pwMND recommendations

Timing of gastrostomy placement

A similar proportion of participants believed that pwMND should have a gastrostomy placed by the time they have lost 5% (53/124, 43%) or 10% (56/124, 45%) weight compared to their weight at symptom onset (mean 8.1% (SD 3.8; range 1-25%)). Over half (64/124, 52%) reported gastrostomy should be placed when pwMND have lost ≥10% weight since symptom onset.

Participants were asked when they believed pwMND should have a gastrostomy placed in relation to the presentation and severity of respiratory failure and swallowing difficulties. Significantly more participants (p<0.001) believed gastrostomy tubes should be placed earlier in relation to the severity of respiratory symptoms (53/137, 39%; indicated by the cells below the shaded cells in table 3) than in relation to the severity of dysphagia symptoms (4/137, 3%) (see supplementary information 6 for further information about how categories were combined). A similar proportion of participants believed that pwMND had gastrostomy tubes placed ‘about the right time’ (71/138, 52%) or ‘too late’ (66/138, 48%).

	When participants believed gastrostomy tubes should be placed in relation to respiratory function (n (%))				
When participants believed gastrostomy tubes should be placed in relation to swallowing function (n(%))	Prior to any respiratory symptoms	Compensating for early respiratory symptoms i.e. pre-NIV	Experiencing significant consequences i.e. on NIV	Never refer to respiratory failure	Total
Prior to any swallowing problems	13 (10)	1 (1)	0 (0)	1 (1)	15 (11)
Compensating for impact of dysphagia e.g. through texture modified diet	48 (35)	40 (29)	3 (2)	20 (15)	111 (81)
Experiencing significant consequences e.g. chest infections, choking episodes	1 (1)	4 (3)	0 (0)	4 (3)	9 (7)

Never refer to dysphagia	0 (0)	0 (0)	0 (0)	2 (2)	2 (2)
Total	62 (45)	45 (33)	3 (2)	27 (20)	137

Table 3. Participants responses to questions about when they believe pwMND should have a gastrostomy placed in relation to their respiratory and swallowing function.

Multidisciplinary decision support

There were differences in the topics discussed with pwMND by different professions. Nurses or dietitians were significantly more likely ($p \leqslant 0.05$) to state they would discuss 17 of the 19 (89%) decision-related information topics, and 12 of the 14 (86%) potential outcomes associated with accepting or declining gastrostomy placement presented as question response options in the survey. In contrast, doctors were significantly more likely ($p \leqslant 0.05$) to state they discuss the impact of respiratory failure on procedural risks (16/17, 94%) and the risks of not surviving the procedure (10/17, 59%), and discuss prognosis in relation to accepting (8/17, 47%) or declining gastrostomy placement (12/17, 71%). See supplementary information 7 (Appendix 19 for this thesis) for the responses to these questions by each profession.

Most participants (60/138, 44%) believed that information given to pwMND by different members of the MDT was ‘moderately consistent’ and half (72/139, 52%) believed that communication between local HCPs was ‘very effective’. A third (45/138, 33%) encountered challenges communicating with HCP colleagues. Neurologists (20/40, 50%), gastroenterologists (12/40, 30%) and respiratory doctors (10/40, 25%) were the HCPs that most participants reported a challenge communicating with.

There was a significant difference ($p=0.009$) between the beliefs of different professions about whether HCPs have a responsibility to give recommendations to pwMND about whether or not to have a gastrostomy, with 74% of nurses (17/23), 65% of doctors (11/17), 45% of dietitians (33/73) and 26% of SLTs (5/19) stating that HCPs should give such recommendations. Significant differences ($p=0.008$) were similarly identified in relation to reported practice, with 71% of doctors (12/17), 70% of nurses (16/23), 44% of dietitians

(32/73) and 21% of SLTs (4/19) ‘sometimes’, ‘often’ or ‘always’ stating they give pwMND recommendations about whether or not to have a gastrostomy placed.

Discussion

Summary of findings

The findings represent a credible account of HCPs’ beliefs and practice in relation to supporting pwMND making decisions about gastrostomy placement. The study identified differences in relation to when discussions about gastrostomy are initiated with pwMND, the information that HCPs share with pwMND and the timing of gastrostomy placement. Nurses and dietitians were more likely to state they address the broadest range of information, except for prognosis and mortality which is more likely to be discussed by doctors. While half of participants believed communication between MDT members was ‘very effective’, some challenges were identified in relation to communicating with medical professionals.

Timing of gastrostomy placement

Half of the participants believed that pwMND have gastrostomy tubes placed too late and varied beliefs were expressed about when gastrostomy tubes should be placed in relation to the key indicators of weight loss, dysphagia and respiratory failure. These findings reflect the contention that exists in the literature about the optimal timing of gastrostomy placement (10,11,12,31,32).

The finding that half of participants believed that gastrostomy tubes should be placed when pwMND have lost >10% weight since symptom onset is consistent with previous case note reviews (33,34). Weight loss after diagnosis and prior to gastrostomy placement is associated with shorter survival informing a recommendation that gastrostomy tubes should be placed prior to pwMND losing 5% weight from diagnosis (7,35). Interestingly, significantly more participants believed pwMND should have a gastrostomy placed earlier in relation to the severity of respiratory failure compared to the severity of dysphagia pwMND are experiencing. While respiratory failure may increase the procedural risks (10,36,37), improving nutritional status and aspiration management are ultimately the primary goals of commencing enteral feeding. The finding that a third of HCPs believe the discussion about gastrostomy is started later than ideal and an acceptance of increased levels of weight loss and dysphagia by the MND MDT may contribute to the delayed placement of a gastrostomy tube and limit the potential for enteral feeding to positively affect the outcomes of pwMND.

There is a need to develop predictive tools that can inform HCPs and pwMND about the outcome of gastrostomy placement in relation to differing presentations of nutritional status, swallowing function and respiratory function. Such decision-support tools could facilitate timely decisions that prevent the development of malnutrition rather than react to it (5,38,39).

HCP recommendations

Participants' expressed divided opinions about whether HCPs have a responsibility to give pwMND recommendations about whether or not to have a gastrostomy; a belief reflected in participants' reported varied use of recommendations in practice. HCPs offering recommendations (40,41) could meet the preferences of some pwMND for HCPs to guide them through the uncertainty of timing of gastrostomy placement (16,17). HCP expertise and opinions are valued by pwMND (17,42,43) and HCP recommendations are often actioned by patients (44). Despite being cited as an essential component of shared decision making (13) concerns remain that decisions informed by HCP recommendations may not be aligned with the values of the patient (45).

A shared understanding of HCPs reasoning (46) for or against gastrostomy placement could help pwMND clarify their own preferences for treatment options available. Interestingly, participants were less likely to state they give pwMND recommendations to continue without or never have a gastrostomy, even when they believed these options were the best for pwMND. These findings suggest an imbalance in how HCPs choose to share their reasoning with pwMND about the best course of action to take. If HCP recommendations are to be used to inform pwMND decision-making, there should be equity in how they are used to aid the understanding of pwMND about all options available including the option to delay or decline gastrostomy placement.

MND MDT decision support

The survey confirmed the multidisciplinary nature of decision support reported in previous studies (18,19,31,47,48). Significant differences were identified between professions' responses including nurses and dietitians being more likely to share information about a wider range of issues relevant to the options available, while doctors were more likely to address sensitive issues such as mortality risk or prognosis. Additionally, nurses and doctors were more likely to give pwMND recommendations about whether to have a gastrostomy placed.

These findings suggest that different professions may take on specific responsibilities during the decision-making process (49) and have contrasting views on how to support the decisional needs of pwMND. The reports in qualitative studies, of conflicting information being shared by HCPs (17,22,23,24) is reflected by 44% of participants believing that their local MND MDT only gives moderately consistent information to pwMND about gastrostomy placement. Inconsistencies in the information shared by different HCPs may lead to decisional conflict and delay the decisions of pwMND about gastrostomy placement, which could, subsequently, impact on patient outcomes.

Strengths and limitations

The absence of a defined sampling frame limits the generalisability of the findings due to not being able to estimate how representative the sample is of the total population. However, validity is strengthened by the comprehensive sampling strategy employed, using a broad range of gatekeepers to reach HCPs known to be involved in discussions about gastrostomy placement with pwMND. With over half of participants being dietitians (73/139, 53%), there may be some bias towards the dietetic perspective which could impact on the generalisability of the findings.

Further strengths of this study include the rigour with which the survey tool was developed including the conceptualisation and design of the survey being informed by the findings of a qualitative evidence synthesis (16,50), collaboration with topic matter experts, and a completion of a pilot study.

Implications for practice

With a third of participants believing they initiate discussions about gastrostomy later in practice than is ideal, MND MDTs should aim to identify and address any barriers to introducing the intervention to pwMND. Professional guidance recommending gastrostomy placement is discussed ‘early’ is vague and lacks concrete guidance about how and when to open these sensitive conversations. Providing psychological support for pwMND to engage in discussions may allow pwMND more time to deliberate about their options and ultimately lead to more timely commencement of the intervention. HCP recommendations may help pwMND navigate the uncertainty in relation to the timing of gastrostomy placement. HCPs should carefully consider how they communicate their preferences for starting gastrostomy feeding, to allow pwMND to make informed decisions that remain aligned with their own values.

Conclusion

The findings of this survey have highlighted differences in the beliefs and practice of the different HCPs’ involved in discussions with pwMND about gastrostomy placement. Any delay in initiating the discussion and beliefs about how progression of indicators inform need for gastrostomy, may contribute to the perceived late placement of gastrostomy tubes. MND services should seek to develop decision-support care pathways, including the range of HCPs and teams involved in these discussions with pwMND, that aim to improve lines of communication and enable a consistent approach to supporting the decision-making of pwMND. Such MDT decision-support frameworks should account for the variation in information and professional recommendations shared with pwMND by individual HCPs. Further research is required to understand the HCP, pwMND and organisational barriers to initiating earlier discussions and to the timely placement of gastrostomy tubes in those pwMND who wish to proceed with the intervention.

Funding

This work was supported by SW’s Clinical Doctoral Research Fellowship (NIHR301592) funded by the Health Education England (HEE) / National Institute for Health and Care Research (NIHR). The views expressed in this publication are those of the author(s) and not necessarily those of the NIHR, NHS or the UK Department of Health and Social Care. CJM is supported by NIHR BRC Sheffield and a NIHR research Professorship award.

Disclosure statement

The authors report there are no competing interests to declare.

Acknowledgements

The authors wish to thank the members of SW fellowship topic expert and patient panels, for their support in the development and write up of this study.

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Data availability statement

The authors confirm that the data supporting the findings of this study are available within the article and its supplementary materials.

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5.6 Summary of the chapter

This chapter has presented the design and conduct of a cross-sectional online survey that has answered the research question:

> ‘How do healthcare professionals support people with MND to make decisions about gastrostomy placement, as individuals and as a collective unit of MDT decision support?’

The findings from the paper are summarised below:

- A third of healthcare professionals initiate discussions about gastrostomy later than they believe is ideal.
- The information healthcare professional share with people with MND about the outcomes of gastrostomy placement differs depending on whether discussing accepting or declining the intervention.
- There is a lack of consensus about whether healthcare professionals should share recommendations with people with MND about gastrostomy placement. This is reflected in varied practice.
- Healthcare professionals are more likely to give a recommendation for people with MND to accept gastrostomy than to recommend people with MND delay or decline gastrostomy, when these are the healthcare professionals preference.
- Respiratory failure informs healthcare professionals reasoning about gastrostomy timing more than dysphagia severity.
- Half of healthcare professionals believe people with MND agree to gastrostomy too late.
- There is only moderate satisfaction with communication and consistency between the MDT, with most concern expressed about the communication of doctors.
- Professional disciplines vary in the information they share with people with MND and in relation to the giving of recommendations with doctors and nurses are more likely to give people with MND recommendations than dietitians or SLT.

The survey findings were used to develop the research plan for the multiple case study to support a contextual understanding of why healthcare professionals may have these beliefs and implement this practice.

6 Multiple case study: timing of discussions and decisions (Paper 3)

6.1 Chapter outline

The following chapter focuses on how discussions about gastrostomy are initiated and on the timing of decisions. Background information relating to the initiation of discussions and timing of decision is followed by a summary of the methods used. Further detail and justifications about the methods can be found in Chapter 3, Section 3.9. A rich and in-depth report of the findings is provided followed by a comparison of key findings with the current literature.

6.2 Multiple case study research question

The research question for the multiple case study was:

> ‘How do healthcare professionals, operating within MDTs, support people with MND to make decisions about gastrostomy placement?’

6.3 Further information related to the study

The following additional information is included as appendices that is related to the development and conduct of the multiple case study (these materials apply to Paper 4 presented in Chapter 7 as they were applied across all three cases):

- Appendix 7 Multiple case study protocol
- Appendix 8 Interview topic guide for people with MND
- Appendix 9 Documentation review proforma
- Appendix 20 Medical note review proforma
- Appendix 21 Scoping email sent to potential cases
- Appendix 22 Screening email sent to interested cases
- Appendix 23 Screening questionnaire sent to interested cases
- Appendix 24 Characteristics of the MND services on Cases 1, 2 and 3
- Appendix 25 Characteristics of people with MND participants

6.4 Publication plans

The paper presented below is currently being prepared for submission for publication in a peer reviewed journal. Ideally, I would like to publish in a MND or neurology focused journal though these often prioritise quantitative research or the restricted word counts would not allow a rich and in-depth report to be included. Therefore, I am considering journals that publish qualitative healthcare research including Health Expectations, PLOS ONE, Qualitative Health Research.

6.5 Paper 3: Timing of discussions and decisions about gastrostomy in Motor Neuron Disease care: a qualitative multiple case study

White S, O'Cathain, A, Halliday V, Birchall S, Bekker H, McDermott C.

Abstract

Decision-making about gastrostomy placement in motor neuron disease (MND) care is associated with a range of clinical, emotional and organisational factors. However, little is known about how these factors influence the engagement of people with MND in discussions about their options and the timing of decisions. The aim of this research was to explore how healthcare professionals support people with MND to engage in timely discussions and decision-making about gastrostomy.

Three UK MND teams participated in a qualitative multiple case study. Purposive sampling was used to recruit to observations of 42 healthcare professional discussions with people with MND and 18 multidisciplinary team (MDT) meetings, interviews with 12 people with MND, 9 caregivers and 18 healthcare professionals and 3 mixed discipline healthcare professional focus groups. Reviews of the medical notes of people with MND and other professional documentation were integrated into the framework analysis.

The timing of discussions and decisions about gastrostomy were influenced by a misalignment in the priorities and preferences of people with MND and healthcare professionals. Healthcare professionals, informed by their knowledge of the disease and consequences of late decisions, preferred being proactive including promoting earlier discussions. However, healthcare professionals were aware of the emotional cost of asking

people with MND to consider disease progression and could choose to delay discussions about gastrostomy if judged people with MND to not be ready. Such a concern is valid, information about disease progression and the gastrostomy option disrupting the avoidant coping strategies people with MND used to help live with the disease. Healthcare professionals’ preference for early intervention sought to protect people with MND from the procedural risks associated with the late placement of gastrostomy but they were sometimes unable to provide concrete information about the timing of decisions. A lack of concrete guidance in addition to the continued avoidant behaviours of people with MND, contributed to delays in decision-making.

The preferences and priorities of healthcare professionals and people with MND, in relation to engaging in discussions about gastrostomy and the timing of decisions, can be misaligned. The emotional response of people with MND to discussions can drive a preference to defer discussions which may delay the commencement of enteral nutrition. Decision-support interventions are needed to empower people with MND to engage in timely meaningful discussions about their options.

Introduction

People with MND are asked to make many complex care and intervention decisions with knowledge that they are living with a disease associated with an average survival of 2-4 years, a lack of effective treatment options and progressive loss of essential physiological function leaving them increasingly dependent on others for care (McDermott and Shaw, 2008). One such decision is the choice about whether to have a gastrostomy feeding tube placed (Stavroulakis *et al.,* 2013). Gastrostomy is routinely offered as an option to manage the risk of dysphagia which affects the majority of people with MND during the disease course and the consequential prevalence of malnutrition and aspiration in MND which are associated with increased morbidity and mortality (Genton *et al.,* 2011; Perry *et al.,* 2021). There are signals in the literature that a higher weight, and a high calorie diet, is associated with improved outcomes (Janse van Mantgem *et al.,* 2020; Ludolph *et al.,* 2020). Gastrostomy is a relatively low risk, surgical option that allows nutrition, hydration and medications to be administered via an alternative route when people with MND are no longer able to safely take adequate amounts orally (Stavroulakis *et al.,* 2013).

Several qualitative studies have identified how decision-making about gastrostomy is emotionally complex with deliberations extending beyond the clinical indications of dysphagia and malnutrition (Lisiecka *et al.,* 2021; White *et al.,* 2023). Firstly, when asked to consider gastrostomy the acceptance and adaptation of people with MND to the diagnosis or disease progression influences their response to intervention options (King *et al.,* 2009). Secondly, being presented with the gastrostomy option can trigger an emotional response associated with no longer be able to eat, signalling disease progression, increased dependence on others and concern about extending a poor quality of life (Martin *et al.,* 2016; White *et al.,* 2023). Thirdly, the decisions of people with MND are influenced by concern for the impact of accepting or continuing without a gastrostomy on their caregivers (Martin *et al.,* 2016). Previous studies have identified a preference of people with MND to focus on the present and avoid discussing future disease progression or intervention options (Greenaway *et al.,* 2015; Hogden *et al.,* 2012b). Such avoidant behaviours present a barrier to opening timely discussions about how people with MND may wish to manage the risks associated with dysphagia.

From a clinical perspective the decision is complicated due to the range of clinical and social factors that indicate a need to consider gastrostomy including dysphagia, reduced intake, weight loss, mealtime length or burden and respiratory failure (Van Eenennaam *et al.,* 2021). Possibly a more complex decision that people with MND face is not just '*if*' but '*when*' to have a gastrostomy placed. While placing a gastrostomy support people with MND to meet their nutritional requirements, there remains a lack of evidence for the effectiveness of the intervention in relation to survival or quality of life outcomes (Sulistyo *et al.,* 2023). However, a controlled trial evaluating the use of gastrostomy in MND would be ethically challenging with the intervention already being used routinely in practice and the challenges with recruiting a suitable control group (Pols and Limburg, 2016). Gastrostomy placement prior to presenting with high aspiration, malnutrition or respiratory risk is associated with improved outcomes (Kasarskis *et al.,* 1999; ProGas study 2015). A large observation study found that >10% weight loss at the time of gastrostomy placement was associated with significantly reduced survival (ProGAS study, 2015). This study informed the current recommendation for people with MND to have a gastrostomy placed before the loss of 5% weight compared to diagnosis (ProGas Study, 2015). Respiratory failure, which is common in MND, presents a less intuitive indicator for the timing of gastrostomy placement (Kasarskis *et al.,* 1999; Miller *et al.,* 2009). As respiratory function declines, the risk of gastrostomy procedural complications increases (Kasarskis *et al.,* 1999; Miller *et al.,* 2009). Respiratory failure has been reported to drive healthcare professionals' preferences for earlier gastrostomy placement (White *et al.,* 2024). The lack of guidance for healthcare professionals and people with MND about the optimal timing of gastrostomy placement contributes to the uncertainty associated with this decision (White *et al.,* 2023; White *et al.,* 2024).

A combination of the clinical and contextual complexities described above contribute to the uncertainty people with MND and healthcare professionals have to navigate when making decisions about the timing of gastrostomy placement. The aims of this research were to better understand how healthcare professionals support people with MND to:

1. Engage in initial discussions about gastrostomy placement.
2. Make decisions about the timing of gastrostomy placement.

Methods

Design

A qualitative multiple case study method following the approach described by Yin (2018) was undertaken. Using the case study method allowed for the process of decision-making about gastrostomy placement in MND care to be studied from several perspectives, using multiple qualitative methods within the real-world settings.

Settings and case definition

In the UK, multidisciplinary teams (MDTs) including a range of different professions working in both clinical and community settings provide care for people with MND (Hogden *et al.,* 2017; NICE, 2019). Three MND teams in the UK were purposively selected as cases including healthcare professionals working in any setting, who discuss gastrostomy with people with MND that attend a single MND clinic. The case was defined as 'the MND multidisciplinary team operating across the clinic and community setting that supported people with MND to make decisions about gastrostomy'.

Sampling and recruitment

Purposive sampling was used when recruiting cases, participants and in relation to selecting non-participant observations. A screening exercise guided the sampling of cases, ensuring variation in the location, MND caseload and model of MND care delivery. A description of the three cases can be found in Appendix 24. Healthcare professionals were sampled to ensure variation in professional discipline and setting in which they worked. People with MND were sampled to ensure variation in stage of decision-making (i.e. early or revisited discussions about gastrostomy, and having made decisions to accept, delay or decline gastrostomy). The non-participant observations were sampled to ensure variation in setting (i.e. home v clinic), the stage of decision-making of people with MND, and healthcare professional disciplines observed. The principal investigator (PI) within each case forwarded invites about the study to potential participants. All participants consented to taking part in each data collection method they participated in. Most of the people with MND, caregivers and healthcare professionals observed in the non-participant observations, were the same individuals that participated in the interviews and focus groups (healthcare professionals only). However, there were some participants who only participated in a single data collection method e.g. only participated in an interview.

Data collection

Fieldwork took place between March 2023 and January 2024. All data was collected by SW. Each case study began with the observation of interactions of healthcare professionals with their colleagues (e.g. MDT meetings) and people with MND (e.g. in clinic or home visits). During and following each observation the researcher recorded detailed field notes relevant to the research aims. Interview topic guides exploring participants' experience of decision-making about gastrostomy placement were iteratively updated to support exploration of emerging concepts. The interviews with people with MND and caregivers lasted 34-84 minutes and healthcare professional interviews 43-72 minutes. Any documentation relevant to discussions about gastrostomy placement in medical notes were extracted. A documentation review of patient information sheets, guidelines, protocols, policies or care pathways related to making decisions about gastrostomy placement was undertaken. Finally, a single mixed discipline healthcare professional focus group was conducted within each case exploring how the MDT supported people with MND to make decisions about gastrostomy placement. The focus groups lasted 78-83 minutes. All interviews and focus groups were audio recorded, except for the focus group on Case C where the recording failed and detailed field notes were recorded instead.

Ethics

The study received NHS research ethics committee approval from West of Scotland Research Ethics Service (REC reference: 22/WS/0171) on 14/12/2022.

Public and participant involvement

The researcher collaborated with several PPI groups, including a group of people with MND and past caregivers recruited to support this study. The individuals involved informed the conceptualisation, design and conduct of the research, including the decision to purposively sample people with MND at different stages in their decision-making journey.

Analysis

All the textual data was uploaded onto NVivo (version 14), in preparation for following the framework approach developed by Ritchie *et al.,* (2014). An *a priori* framework was developed informed by the findings of a qualitative evidence synthesis (White *et al.,* 2023) and online healthcare professional survey (White *et al.,* 2024), which included initiation of discussions, decision-making process, decision-making and the organisation of MDT decision support. The field notes from observations and transcripts from the interviews and focus groups were line-by-line coded to the framework with new descriptive themes and sub-themes created and applied to each case. Matrices including data summaries for each unit of analysis (i.e. individual observation, interview or focus group) under each sub-theme were developed in NVivo (Ritchie *et al.,* 2014). Indicators and dimensions were identified from the data summaries to inform the presentation of the findings in the themes below. A process of pattern matching was undertaken through interrogating the matrices across themes and between cases, including actively seeking for any alternative explanations (Yin, 2018)

Findings

Description of cases and participants

Over the three cases, 42 interactions between healthcare professionals and people with MND (35 clinic appointments and 7 home visits), and 18 MDT meetings were observed. In total, 18 healthcare professionals, 12 people with MND, and 9 caregivers were interviewed. Across the three cases 13 different healthcare professional disciplines were observed, 8 different disciplines interviewed, and 9 different disciplines participated in the focus groups. The characteristics of healthcare professional participants are not reported to protect anonymity. Observations took place in the clinic and community setting. See table 1 for a description of the characteristics of people with MND participating in interviews. The cases within which the people with MND were recruited is not reported to protect anonymity.

The cases were from different regions of the UK. The cases differed in their total MND caseload and the organisation of the MND MDT. Cases 1 and 2 included a central MND Care Centre clinic team with several supporting community teams surrounding it, and within Case 3 the same healthcare professionals supported people with MND in both the clinic and community setting. Each case varied with regards how the MDT was organised and the representation of different healthcare professional disciplines. (see appendix 24).

Table 1. Characteristics of the people with MND participating in interviews.

	P1	P2	P3	P4	P5	P6
Gender	Male	Male	Female	Male	Male	Male
Time since diagnosis	75 months	2 months	9 months	4 months	12 months	8 months
Speech	No speech problems	Significant dysarthria. Communicated by writing on a pad.	Unable to speak. Communicated using I-Pad.	No speech problems	No speech problems.	No speech problems
Observations involved in	Observed in clinic with neurologist and MND CNS.	Not observed.	Observed in clinic with neurologist and dietitian.	Observed discussing gastrostomy in home with dietitian and nurse	Not observed	Not observed
Healthcare professionals discussed gastrostomy with	Neurologist, MND CNS, palliative care consultant, respiratory CNS.	Ward doctor (not a neurologist), MND CNS, SLT, Dietitian	Neurologist, dietitian, respiratory centre,	Dietitian, community neurology nurse	Neurologist, respiratory consultant	MND CNS, dietitian, neurologist, respiratory consultant.
First discussion	~ 3½ years after diagnosis	At diagnosis	2 months after diagnosis.	3 months after diagnosis.	2-3 months after diagnosis.	MND nurses discussed gastrostomy 3 months after diagnosis.

Current decision	Declined gastrostomy.	Agreed to gastrostomy soon after diagnosis.	Agreed to gastrostomy 7 months after diagnosis.	Is open to gastrostomy placement when experiencing dysphagia.	Agreed to gastrostomy 10 months after diagnosis.	Agreed to gastrostomy 7 months after diagnosis.
Gastrostomy placed	No	Awaiting gastrostomy placement	Gastrostomy placed 3 weeks ago.	Deferred decision	Awaiting gastrostomy placement	Awaiting gastrostomy placement
Caregiver interviewed?	Yes (separately)	Yes (separately)	Yes (separately)	Yes (separately)	Yes (with person with MND)	Yes (with person with MND)

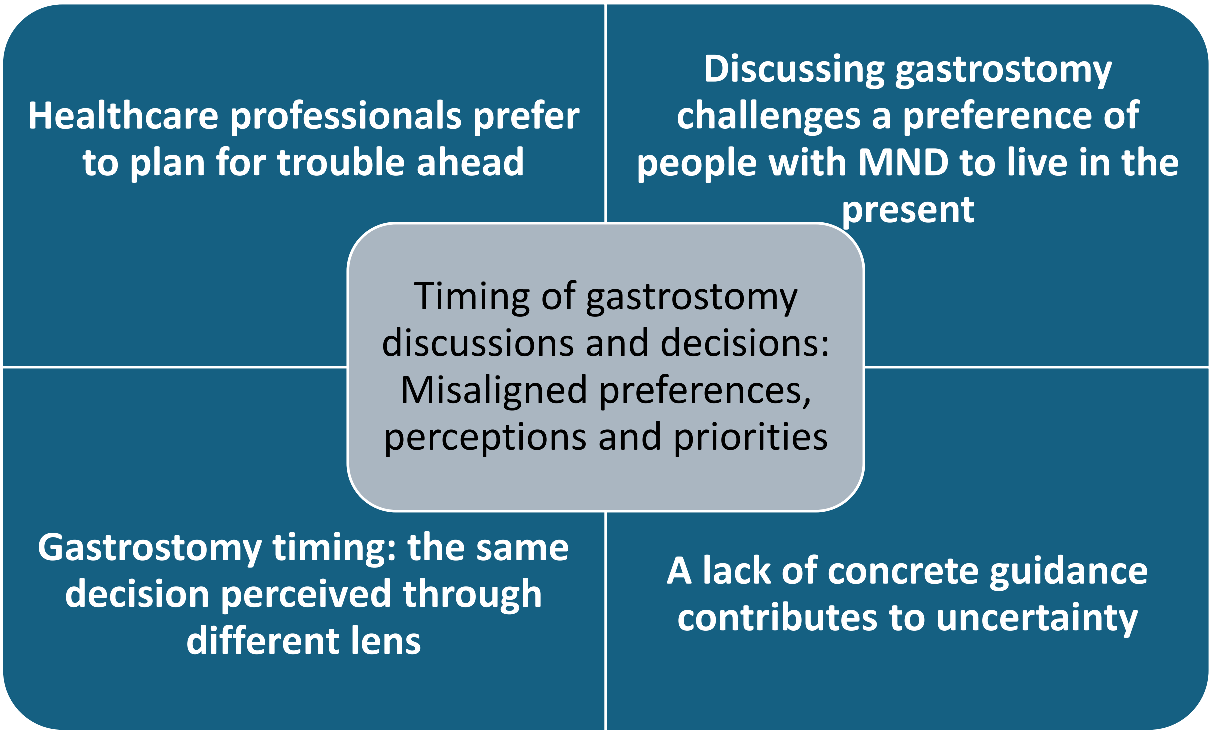
	P7	P8	P9	P10	P11	P12
Gender	Female	Male	Male	Male	Male	Male
Time since diagnosis	9 months	4 months	21 months	2 years	20 months	3 years
Speech	Unable to speak. Communicated through writing.	No speech problems	No speech problems	No speech problems	No speech problems	No speech problems
Observations involved in	Observed in clinic with neurologist and respiratory consultant	Not observed	Observed in clinic with neurologist, MND CNS, palliative care consultant and	Observed in clinic with respiratory consultant and physiotherapist.	Observed in clinic with dietitian, SLT, palliative care consultant, respiratory consultant, physiotherapist and nurse. Observed at home with dietitian.	Not observed

			respiratory consultant			
Healthcare professionals discussed gastrostomy with	Respiratory consultant, two neurologists, SLT, MND coordinator and MND CNS.	MND nurse, dietitian, neurologist.	Neurologist, MND CNS, respiratory consultant, palliative care consultant.	Dietitian.	Dietitian, SLT.	Dietitian, palliative care consultant, MND coordinator.
First discussion	Neurologist discussed gastrostomy 3 months after diagnosis.	Neurologist discussed gastrostomy at diagnosis	Early after diagnosis as part of advanced care planning.	Neurologist discussed gastrostomy at diagnosis.	Dietitian discussed gastrostomy 19 months after diagnosis.	Unsure. Thinks it was mentioned early by palliative care consultant.
Current decision	Uncertain	Agreed to gastrostomy shortly after being diagnosed	Declined gastrostomy	Agreed to gastrostomy 16 months after diagnosis.	Agreed to gastrostomy	Agreed to gastrostomy 2 years and 10 months after diagnosis.
Gastrostomy placed	No	Awaiting gastrostomy placement	No.	Yes	Awaiting gastrostomy placement.	Awaiting gastrostomy placement.
Caregiver interviewed?	Yes (with people with MND)	Yes (separately)	No caregiver	No (declined)	Yes (separately)	Yes (with person with MND)

Overview of themes

The themes presented below relate to the case study objective to understand the factors influencing the initiation of discussions about, and timing of, gastrostomy placement. An overarching theme was that of misaligned preferences, perceptions and priorities between healthcare professionals and people with MND about the initiation of discussions and timing of gastrostomy. Healthcare professionals' tacit knowledge of disease progression and consequences of delayed decisions about gastrostomy informed a preference to be proactive and act ahead of a clinical need presenting. In contrast, the preference of people with MND to extend a sense of normality, and uncertainty about when gastrostomy is indicated could result in discussions and decisions being avoided or delayed. How healthcare professionals described and framed the decision about timing of gastrostomy influenced people with MND perception of need and acceptance of referrals for placement.

Figure 1. Themes relating to the timing of discussions and decisions about gastrostomy



Healthcare professionals prefer to plan for trouble ahead

While the knowledge people with MND have of MND and the gastrostomy decision is often limited to the information shared at or after diagnosis, healthcare professionals enter the collaboration already in possession of a significant amount of tacit and clinical knowledge about the disease and future care options. Healthcare professionals’ knowledge is informed by their prior clinical experience of the consequences of dysphagia, the published guidance and research on the topic, the time it can take people with MND to make a decision, and the limitations of local services to place gastrostomy quickly.

Across all cases, healthcare professionals had a consistent clinical preference to be proactive in relation to having discussions about gastrostomy. While all participants recognised that people with MND should be supported to make autonomous decisions, the introduction of the gastrostomy option was more often under the control of the healthcare professional. Healthcare professionals aimed to introduce the gastrostomy option early in the disease course including prior to significant symptom burden, motivated by a desire to promote choice, improve clinical outcomes and avoid or reduce procedural and disease-related risks. Opening early conversations was perceived by healthcare professionals to give people with MND more time to understand and evaluate their options before communication or cognitive decline and to facilitate the making of timely decisions.

> ‘by the time there are, communication maybe slowing or they may be getting more fatigued. So, I think we're depriving them if we don't, depriving them of a really important opportunity to begin thinking about it[…]it's just got to be early.’ (Case 3 healthcare professional)

Healthcare professionals’ preference for being proactive sometimes needed to be adapted in response to the current or predicted clinical need of people with MND, or by an internal judgment about whether people with MND were emotionally ready to discuss gastrostomy. HCPs were aware that introducing the gastrostomy option could trigger an emotional response and would themselves sometimes delay initiating a discussion about gastrostomy through concern for the emotional wellbeing of people with MND or that the discussion could threaten their therapeutic relationship. However, healthcare professionals’ concern for the emotional wellbeing of people with MND were overridden when they presented with significant clinical indications for gastrostomy. For example, where people with MND presented at diagnosis with swallowing problems, healthcare professionals were compelled to discuss gastrostomy placement while they were still processing information about the diagnosis. Indeed, one Case 3 healthcare professional described having a ‘moral obligation’ to open such early discussions to avoid widening health inequalities if only those people with MND who were emotionally able to engage in these difficult conversations were given the opportunity to understand their options early.

> ‘…one of the things we see in MND is that there are people who are very, very well informed and there are others who are less so […] I think it's on us the responsibility to try to address some of that imbalance […] if we only do that for those who already know that there are ways for that voice to be heard, and don't do it for those, then we risk systematically widening the existing inequities’ (Case 3 healthcare professional)

Discussing gastrostomy challenges the preference of people with MND to focus on the present

In addition to the physical impact of the disease, the diagnosis has an emotional consequence for people with MND and their caregivers, in relation to the predicted short prognosis, progressive loss of vital functions and increasing dependence on others. Discussions about how to manage current or future dysphagia were one of many difficult conversations people with MND were observed to have with healthcare professionals about their disease course and care options, many of which can be taking place simultaneously, even within the same clinic appointment. This was the challenging context into which the option to have a gastrostomy is positioned and contributes to an emotional response to the option being presented.

> ‘Yes, back to that appointment, we went outside into the car park, and he was completely shocked about all the, all the things that was, he was being told then. He did get quite, I’m not going to say upset because he wasn't upset, but he was, you could see it in his face that he was very anxious about what these changes meant for him because it was all a big shock to him then really’ (Case 3 caregiver interview)

People with MND and their caregivers frequently reported or were observed to be upset or shocked by the gastrostomy option being raised including one Case 2 person with MND describing it as feeling similar to when she was given the diagnosis. People with MND could lack an understanding about why the placement of a gastrostomy was being discussed when they were not currently experiencing any physical symptoms that would intuitively indicate a need. The emotional response to the unexpected presentation of the gastrostomy option appeared to be compounded when people with MND were still questioning their diagnosis or lacked awareness of the predicted future changes to their swallow. People with MND experienced a further emotional response to what placement and feeding via a gastrostomy represented for them and their caregivers. The discussion challenged people with MND to consider a time when they would not be able to eat and represented a signal that their condition was progressing, or they were getting closer to the end of their lives.

> ‘it did make me realise the disease would progress in a way I had not thought of. It was not a good feeling. Almost as bad as first being told that I had MND’ (Case 2 interview with a person with MND)

Interestingly, the emotional responses of people with MND were not always overtly available to the healthcare professional during discussions about gastrostomy, as captured by the observation and interview of the same person with MND and caregiver below. The Case 1 person with MND was observed to be comfortably engaging in discussion with two healthcare professionals about gastrostomy, but subsequently reported during an interview at being surprised and unprepared for the conversation.

> ‘The patient did not appear to be upset by the discussion and both [pwMND] and [caregiver] engaged.’ (Observation of a home visit in Case 1 involving a person with MND, two caregivers and two healthcare professionals)

> ‘there was no warning, but you know the person I am its surprise rather than anger’ (Case 1 interview of the same person with MND observed above)

> ‘I was just a bit shocked that it was that day, because I wasn’t expecting it’ (Case 1 interview of caregiver present in observation above)

A number of people with MND were observed and reported minimising, delaying or avoiding discussions to allow them to prioritise focusing on what they could control in the present. People with MND frequently wanted to extend a sense of normality and expressed a preference to address physical challenges when they were perceived to be necessary. Though some people with MND believed such avoidant coping strategies were effective in helping them live with the disease they were also a barrier to healthcare professionals implementing their preference to be proactive and delayed an opportunity for people with MND to be informed about their options. A number of healthcare professionals made the observation that people with MND may not always be making informed choices to defer discussions, suggesting they should be supported to understand the potential consequences of delaying discussions.

> ‘So, in a way, deciding whether to talk about gastrostomy is very like deciding whether to have a gastrostomy. Before you can decide you need to know something, something, and at least a bit about the pros and cons of talking versus not talking. And that's on us.’ (Case 3 healthcare professional)

Gastrostomy timing: the same decision perceived through different lens

People with MND reported how initially they aimed to defer agreeing to gastrostomy until they perceived there was a physical need. However, the perceptions of people with MND and healthcare professionals with regards the need to make a decision were not always aligned or shared. Healthcare professionals’ preference for decisions about gastrostomy to be made before people with MND were experiencing significant symptom burden was largely driven by a motivation to reduce the disease-related and procedural risks associated with delayed gastrostomy placement. Healthcare professionals used their tacit knowledge of MND and observable markers of disease progression to predict the current and future clinical need for enteral nutrition. While healthcare professionals were observed to routinely describe what the intervention involved (i.e. how the tube was placed, used and cared for), and the risks associated with the procedure, there was less focus on the expected health outcomes of continuing with or without enteral nutrition. People with MND often reported to understand the advice that there were increasing procedural risks associated with worsening frailty and respiratory failure including the potential for the option to have a gastrostomy to be withdrawn. Long waiting lists to place gastrostomy also informed healthcare professionals’ preference for people with MND to accept referral for gastrostomy prior to there being a need for enteral nutrition. People with MND were advised to expect to wait up to 3 months for gastrostomy placement following having agreed to proceed, within which time healthcare professionals feared people with MND could reach disease-related crisis or be assessed as no longer being fit for the procedure.

> ‘[…]we do struggle sometimes with individuals not having been given the full facts and then they've been disappointed that they didn't realise there was a clinical cut-off as well as a decisional cut-off from their point of view’ (Case 1 healthcare professional interview)

Decisions to defer gastrostomy placement can be motivated by people with MND wanting to maintain a sense of normality, even when there was objective (e.g. deteriorating test results) or experiential signals (e.g. difficulty feeding self) of clinical need. Through using compensatory strategies (e.g. oral nutrition support or adapting the way they feed themself), people with MND were able to maintain a sense of normality which they often wished to extend for as long as they possibly could. When informed about, or even experiencing, changes in their swallow, weight or breathing, people with MND could continue to question whether the symptoms indicated a need for gastrostomy or the accuracy of test results. The procedure to place a gastrostomy, body image concerns and negative perceptions about being enterally fed, triggered a fear response which could be avoided by delaying decisions. Therefore, people with MND were often waiting to be convinced that gastrostomy placement was worth the burden they and their caregivers would have to take on to overcome their instinctive avoidant behaviours. While experience of symptoms was a driver for making a decision, healthcare professionals also played an important role in convincing people with MND about the timing of decisions about gastrostomy.

> ‘Because when I use the stairlift, I am closer to dying. And that is why I resist everything, and it wasn’t a case of [healthcare professionals] convincing me that I needed it. [HCP] needed to convince me that I needed it now.’ (Case 3 interview with a person with MND)

A lack of concrete guidance contributes to uncertainty

People with MND are broadly identified by healthcare professionals, caregivers and themselves as having responsibility for making the final decision about gastrostomy. However, some people with MND wanted healthcare professionals to use their previous experience of MND to guide when they should make the decision to have a gastrostomy. The decisions of people with MND to accept referral for gastrostomy were driven by not just their experience of symptom burden but also the guidance provided by healthcare professionals. Even in the presence of symptoms, some people with MND needed a healthcare professional to validate that their current presentation represented an appropriate time to accept gastrostomy or to overcome an instinctive preference for delay. Healthcare professional guidance appeared to be particularly relevant for scenarios when people with MND were asked to consider gastrostomy prior to experiencing significant physical symptoms, for example when the procedural risks associated with respiratory failure were the primary indicator. In such scenario's, healthcare professionals' reasoned arguments were required to convince people with MND to accept living with a gastrostomy without needing to use it, in exchange for experiencing the predicted long-term benefits for their future selves. People with MND described how a direct approach by the healthcare professional helped reduce their uncertainty, challenged avoidant behaviour and, supported them to make what they perceived as a well-timed decision.

> 'how the respiratory consultant spoken to me that was more powerful in making my decision. The other people they were saying, but they can't like they haven't explained well why I need it now and is there any need it or should I go for it or shouldn't, should I go for it like, they were leaving to me all the like to dig out and find out like information, they were willing to answer my own question, but not giving exact information why I needed that was the only like with my respiratory consultant, she gave me that with the timing.' (Case 1 interview with a person with MND)

> 'So, I have to, I'm going to do a small damage [now] to avoid big damage [in the future]' (Case 1 interview with a person with MND)

Healthcare professionals' messaging about the timing of gastrostomy was observed to lack concrete information to inform decisions, often relying on abstract descriptions or analogies. Healthcare professionals often used abstract terms such as 'earlier rather than later', 'window of opportunity' or compared the decision to paying taxes to capture the time-limited nature of the decision or the benefits of making a decision ahead of needing enteral nutrition. While such descriptions enabled people with MND to understand the time-limited nature of the decision, they were less helpful in identifying the criteria by which decisions about timing should be made upon. Healthcare professionals acknowledged that such abstract descriptions were open to interpretation. In contrast to some healthcare professionals not feeling it necessary to provide quantitative evidence to support the timing decision, a few people with MND expressed a wish for more concrete guidance and objective risk communication to help inform their timing decisions. Uncertainty about the rate of disease progression and the outcomes of gastrostomy placement often resulted in people with MND continuing to exert their preference for deferring the decision until they experienced a physical need for intervention.

> ‘‘I think it creates a time limit, but I can’t give them the timeframe’ (Case 2 healthcare professional interview)
>
> ‘I wasn’t sure what weight [person with MND] could go down to, and still kind of survive, but I thought we’re getting to the lower end of it’ (Case 1 caregiver interview)
>
> ‘I don’t know whether they could do, give a weight, do you know what I mean, and seeing the weight’s dropping, […] I had prostate cancer, they were looking at the figures, and they said, ‘when it gets to this figure we need to do something’, so that kind of thing.’ (Case 1 caregiver interview)

Preparing people with MND for decision-making

A number of preparatory strategies were observed that may address the predicted emotional response, supported engagement in discussions or helped people with MND and healthcare professionals to understand each other’s reasoning. As one Case 1 healthcare professional highlighted, people with MND had a predictable emotional response to being presented the gastrostomy option and therefore it could be identified, validated and addressed by healthcare professionals to facilitate meaningful discussions. However, it was also acknowledged that not all healthcare professionals have the communication skills or confidence to do this. Healthcare professionals were frequently observed to seek permission to discuss the future or present potential treatment options including checking in during discussions that people with MND were happy to continue, giving people with MND some control over what information was shared with them. Providing people with MND with a clear rationale for why the healthcare professional wanted to begin or continue discussing gastrostomy, particularly in the absence of symptoms indicating a need, facilitated engagement and helped address resistance. Palliative care healthcare professionals in particular referred to the importance of exploring the understanding, values, goals and preferences of people with MND when discussing gastrostomy placement to address misconceptions and align decisions with what is important to people with MND. Finally, post diagnosis group information sessions were proposed as a potential strategy by some healthcare professionals and people with MND to mediate the emotional response associated with discussing sensitive topics one-to-one with a healthcare professional.

> 'You also realised there were other people in the same boat and it became less emotional. It became less emotionally connected because we weren't one to one with a Medical Practitioner.' (Case 3 caregiver interview)

Discussion

Overview of findings

The timing of discussions and decisions about gastrostomy is emotionally, clinically and ethically complex for people with MND and healthcare professionals. Healthcare professionals and people with MND enter the collaborative process of decision making with different knowledge, perceptions and priorities. Healthcare professionals' preference to plan ahead is not always aligned with people with MND wishing to focus on the present and delay thinking about the future. Discussions about gastrostomy trigger an emotional response that is not always overtly expressed during consultations with their healthcare professionals. People with MND were instinctively waiting for physical evidence for a need to accept gastrostomy and often needed convincing by their healthcare professionals that they had reached this stage. There was evidence of strategies that could support engagement in discussions. The findings will now be discussed in relation to the current literature.

Engaging people with MND in discussions about the gastrostomy option

Discussions start with people with MND and healthcare professionals being worlds apart

Healthcare professionals and people with MND enter the initial discussions about gastrostomy with different tacit and clinical knowledge about the disease which influences their preferences, perceptions and priorities. In line with published research and guidance most healthcare professionals expressed a proactive preference for initiating discussions about gastrostomy prior to people with MND experiencing a significant decline in any indicators for the intervention (Preston *et al.,* 2012; ProGas Study, 2015; Martin *et al.,* 2016; Paynter *et al.,* 2019; Paynter *et al.,* 2020). Healthcare professionals' justification for earlier discussions included optimising the ability of people with MND to engage in discussions or express their preferences (i.e. prior to communication, cognition or general disease decline) and through concern for the negative consequences associated with delayed decisions (Harris *et al.,* 2023). Healthcare professionals' decisions about when to discuss gastrostomy were influenced by the outcome of clinical assessments, tacit knowledge of the disease course and the outcome of late decisions, and subjective judgements about emotional readiness (Hogden *et al.,* 2015; Benstead *et al.,* 2016; Martin *et al.,* 2016). Similar to a previous study, healthcare professionals use their tacit knowledge about MND to be 'one step ahead' of people with MND to predict symptom decline and the timely discussion of intervention options (McConigley *et al.,* 2014). While these are ethically and clinically reasonable rationales, the current study highlighted how people with MND may not always share the same preferences and priorities as healthcare professionals in relation to discussing future disease decline and the options available. A previous healthcare professional survey identified that a third of healthcare professionals believed they initiated discussions about gastrostomy later in practice than was ideal, suggesting there is a barrier to opening these conversations (White *et al.,* 2024). The current study was an opportunity to understand what the barriers were to early discussions in real-world practice.

Emotional response and coping strategies can delay engagement

Healthcare professionals concern for the emotional well-being of people with MND is justified. Consistent with previous research, learning about future disease progression and interventions are unwelcome conversations for many people with MND (O'Brien 2004; Greenaway *et al.,* 2015). People with MND frequently reported an emotional response to being presented the gastrostomy option, with some comparing it to being given the diagnosis, or signalling being a step closer to the end of their lives (Hogden, 2014). Interestingly, such emotional responses were not always overtly expressed during discussions and therefore not available for healthcare professionals to modify their approach to account for them. This finding highlights that healthcare professionals cannot solely rely on what they can see and therefore should prophylactically prepare all people with MND for the emotional cost of these discussions.

Consistent with the findings of previous research, some people with MND employed avoidant coping strategies that deferred needing to accept there may be a need to consider gastrostomy, including delayed engagement in discussions or questioning the outcome of test results (O'Brien, 2004; Gale, 2015; Paynter *et al.,* 2020; Pinto *et al.,* 2021; Connolly *et al.,* 2023). Previous studies have linked healthcare professional assessments of the emotional well-being of people with MND to the timing of discussions about gastrostomy (Barc *et al.,* 2022) and the coping strategies employed by people with MND, to gastrostomy uptake (Montel *et al.,* 2012; Pearlman and Thorsteinsson, 2019). Therefore, the response of people with MND to the discussion and the coping strategies they employ play an important role in relation to when people with MND enter the process of understanding their options. These findings support the concept that people with MND and healthcare professionals' priorities and perceptions are not aligned. Healthcare professionals are motivated by promoting positive clinical outcomes by planning ahead and people with MND prioritise strategies that support them to cope with the present including delaying discussions and decisions. Therefore, addressing the response of people with MND to discussing disease progression or care options should be a key attribute of strategies aimed at encouraging earlier engagement in decision-making discussions.

Despite the barrier to engaging in discussion they present, the psychological benefits of avoidant coping strategies for people with MND should not be disrupted without additional support, with individuals having different preferences for information or involvement in decision-making. As reported elsewhere, people with MND want to remain in control and may disengage with healthcare services if they perceive control is being taken from them (Foley *et al.,* 2014a; Genuis *et al.,* 2023). The current study confirmed that healthcare professionals largely control the timing of initial discussions about gastrostomy, a role often supported by people with MND. However, people with MND were able to exert control through their decisions about when they engaged in discussions or decisions about gastrostomy (Foley *et al.,* 2014a; Greenaway *et al.,* 2015). Empowering people with MND to be able to enter and remain engaged in discussions about their options should be a core aim of the decision-making process in MND care (Foley *et al.,* 2014b; Gale, 2015).

The ethical argument for preparing people with MND to engage in discussions

Decisions about when and how to open discussions about gastrostomy are associated with competing biomedical ethical principles: autonomy, beneficence, and nonmaleficence (Beauchamp and Childress, 2013). Healthcare professionals in the present study were motivated to initiate early discussions because they believed they were associated with better outcomes (i.e. beneficence) and reduced the risk of future risks or harm (i.e. nonmaleficence). Respecting a person with MNDs' preference to avoid discussion about gastrostomy could reasonably be viewed as protecting their autonomous rights, something most healthcare professionals identified as a priority within their approach to supporting decision-making. However, for decisions to be autonomous they should also be informed including knowledge of the risks and benefits of each option available. If people with MND lack understanding of the consequences of avoiding timely discussions, particularly when choices are driven by an emotional response, the principles of autonomy, beneficence and nonmaleficence are all under threat. A Case 3 healthcare professional proposed that healthcare professionals have a moral obligation to support all people with MND to engage in discussions to prevent widening health inequalities. If discussions are delayed in those avoidant people with MND, who are not emotionally 'ready' to learn about their options, they risk experiencing less choice and poorer outcomes than others who are capable of taking a more active problem-focused approach. This is consistent with the findings of a cohort study which found that people with MND with more active attitudes were more likely to make decisions about NIV or gastrostomy and the risk of some patient groups being denied the opportunity to engage in timely interventions (Martin *et al.,* 2014). There is an ethical argument for MND services to address barriers limiting the motivation or capability of people with MND, to promote engagement in timely decision-making discussions.

Supporting engagement in decision-making discussions

MND decision-making models have focused on informing people with MND about the health threat (the problem), the intervention and outcomes of the options (Hogden *et al.,* 2015). The current study findings argue for extending the process of decision to before information about options is shared and to include creating an environment which addresses the emotional representations and coping strategies to promote engagement in decision-making (Witt *et al.,* 2012). Strategies were observed that may prepare people with MND to engage in discussions included explaining the rationale for discussing options, signposting the discussion ahead of time, seeking permission and proactively addressing emotional responses. Following diagnosis or even within individual interactions there is a space for people with MND to be empowered with the knowledge, motivation and capability to engage in discussions, including promoting psychological flexibility (Pearlman and Thorsteinsson, 2019). There is increasing evidence for the role of the psychological interventions in MND care to support people with MND to adopt positive coping strategies for living with MND (Gould *et al.,* 2024). Psychological interventions such as acceptance and commitment therapy (ACT) which has recently been proven to improve the quality of life of people with MND, may also facilitate engagement in sensitive discussions which are emotionally challenging (Tramonti *et al.,* 2012; Gould *et al.,* 2024). If successful, this is half the battle won. Healthcare professionals will have people with MND engaged and open to learning about, understanding and evaluating their options. The second half of the battle is supporting people with MND to make informed values-based decisions about gastrostomy placement, including in relation to the timing of the intervention.

Informing the decisions people with MND make about the timing of gastrostomy placement

A complicated clinical decision for healthcare professionals to communicate and people with MND to understand

Similar to decisions about when to initiate discussions, discussions about the timing of gastrostomy were often associated with misaligned priorities, preferences and perceptions of need of people with MND and healthcare professionals. The timing of gastrostomy is complicated for several reasons. Consistent with previous studies, the clinical need for gastrostomy was discussed in response to several indicators including the escalating malnutrition and aspiration risks (Stavroulakis *et al.,* 2013; Van Eenennaam *et al.,* 2021). Maybe less intuitively, healthcare professionals also presented respiratory failure as a reason to consider gastrostomy in relation to increasing procedural risk or even a future contraindication to placing a gastrostomy. Uncertainty about the presentation and rate of progression of each indicator contributed to the decisional conflict about timing. A particularly difficult concept for people with MND to accept is agreeing to gastrostomy in the absence of any indications for enteral nutrition i.e. due to respiratory failure. A further organisational factor that all cases considered was the time taken for services to arrange gastrostomy placement, which could be up to 3 months. This is consistent with an Australian study, where people with MND waited 52 days (range 18-163 days) between referral and gastrostomy placement (Labra *et al.,* 2020). Only the Canadian ALS guidelines have included a recommendation for a minimum wait (i.e. 4 weeks) between gastrostomy being indicated and placement (Shoesmith *et al.,* 2020). Therefore, when considering the timing of their decision people with MND and healthcare professionals should acknowledge the further deterioration that could occur while waiting for gastrostomy placement. Available objective information about predicted disease progression and healthcare professionals sharing their experience of MND, could help inform the decisions people with MND make about the timing of decision making. Healthcare professionals have a complicated task of synthesising a range of clinical and organisational factors when counselling people with MND in relation to their decisions about timing of gastrostomy placement, in addition to considering how people with MND perceive and emotionally respond to the information.

Decision drivers – role of healthcare professional guidance and recommendations

The dialogue between healthcare professionals and people with MND influenced when decisions were made about gastrostomy. People with MND reported two main drivers for accepting gastrostomy referral: direct or adverse experience of symptom progression and the impact of healthcare professional guidance (Vesey *et al.,* 2008; Hogden *et al.,* 2012a; Martin *et al.,* 2016; Paynter *et al.,* 2020; White *et al.,* 2023). However, similar to previous research, experience of symptom burden or healthcare professionals presenting objective evidence of disease decline were not always enough alone to convince people with MND of a need for intervention (Greenaway *et al.,* 2015; Paynter *et al.,* 2020). People with MND often prioritised extending a sense of normality through the significant effort of compensatory action, to delay needing to accept that gastrostomy was indicated. A more direct approach by healthcare professionals including giving clinical recommendations and explicit explanations of the negative outcomes of deferring decisions was influential on decision-making (Genuis *et al.,* 2023). Healthcare professional guidance and recommendations appeared to be particularly effective when guiding decisions to have a gastrostomy prior to the presentation of any significant indications for enteral nutrition including when people with MND were struggling to accept there was a need to act. Some people with MND acknowledged that they required healthcare professionals to identify and challenge their resistance and validate that they needed to make a decision. These scenarios suggest that healthcare professionals are taking some responsibility for decisions, and aligned with previous research, people with MND are happy to release some control when it helps navigate the uncertainty associated with this time-limited decision (Foley *et al.,* 2014a; White *et al.,* 2023). Healthcare professional recommendations have the potential to inform and but also manipulate the decisions people with MND make about gastrostomy (Martin *et al.,* 2016). Therefore, healthcare professionals should consider carefully how recommendations are framed and informed, ideally ensuring they are embedded in a knowledge of the goals and values of the person with MND.

Reducing procedural risk or avoiding disease-related crisis was a high priority for healthcare professionals and featured prominently in their discussions with people with MND about the timing of gastrostomy. Healthcare professionals communicated, and people with MND often understood, one clear message: to place gastrostomy ‘earlier rather than later’. However, some healthcare professionals and people with MND struggled to define when ‘earlier’ was. While people with MND understood the potential for increasing procedural risks, healthcare professionals focused less on the outcomes of enteral nutrition during discussions about gastrostomy. This is consistent with a previous study where people with MND reported feeling uninformed about the potential benefits of gastrostomy placement (Stavroulakis *et al.,* 2014). Future decision support interventions should seek to inform people with MND about outcomes of gastrostomy placement AND of commencing enteral nutrition at any stage of disease progression, to allow them to compare this with the option of continuing without.

Aligned with previous research, people with MND found the use of abstract descriptions or analogies unhelpful such as ‘earlier rather than later’ or compared the decision to paying taxes or into an insurance scheme to guide timing of the decision (Greenaway *et al.,* 2015). Greenaway *et al.,* (2015) described how such abstract descriptions signified healthcare professionals uncertainty in relation to predictions of disease progression. People with MND described wanting more concrete information about likely progression, the outcomes of continuing without intervention and the risk of the option being removed. Despite on-going research to develop prediction models, decisions about the timing of gastrostomy will continue to be associated with a degree of uncertainty and require healthcare professionals to collaborate with people with MND to help weigh up their options and align them with their own goals and values.

Strengths and limitations

The rigour with which this research has been designed, conducted, analysed and reported is a strength of the multiple case study presented. Firstly, the study objectives and research design were informed by a systematic search of the published literature and in collaboration with PPI, academic and healthcare experts, providing clear direction to the fieldwork completed. The credibility of the study is further strengthened through the excellent access attained within each case to observe and purposive sample cases, settings, and participants. The rigorous and reflexive approach to undertaking the framework analysis and the rich and thick descriptions of the themes, contribute to the dependability of the findings and allow the reader to consider how transferable the findings are to other settings. Finally, while this study has focused on the specific decision people with MND make about gastrostomy placement, the findings are likely to be transferable to other decisions made in MND care, or other progressive neurological conditions.

However, there are limitations in relation to this study. Most of the data collected was cross-sectional and relied on participants retrospective reflections of their experiences. However, decision-making about gastrostomy is clearly a process that takes place over time, people and settings. Future research would benefit from studying how the perceptions of people with MND of the decision problem evolve over time to take into consideration the longitudinal and cyclical nature of decision-making (King *et al.,* 2009). The number of observations and interviews undertaken were limited by the resource and time available. Finally, this study focused on the interactions between healthcare professionals, people with MND and their caregivers. The decisions people with MND make are also likely to influenced by the information they access online or by discussions they have with other people with MND. Including such insights would have provided greater depth to the study findings and should be a focus of future research.

Implications for research and practice

This study has several implications for practice and further research or intervention development. First, promoting timely discussions that begin the process of developing an understanding of the problem and the options available are a clinical and ethical priority. There is a need to develop strategies and interventions that empower people with MND to make informed choices about engaging in meaningful discussions including addressing any emotional response. Second, the decision about timing of gastrostomy should be considered as a stand-alone decision (i.e. separate to the decision about if people with MND want to have a gastrostomy at all). While decision aids have been recently developed, they focus more on the decision about whether to have a gastrostomy, rather than the timing of decisions (Maunsell *et al.,* 2019). Evidence based decision support interventions are needed to support understanding the clinical complexities and uncertainty associated with the timing decision (Stacey *et al.,* 2024). Third, in relation to the organisational factor of delays placing gastrostomy once referred, a more efficient process would allow people with MND to make decisions closer to there being a clinical need. Finally, future research is required to develop and evaluate the effectiveness of different strategies or interventions that seek to improve engagement or the quality of the decision-making process.

Reflexivity

It was important to acknowledge and discuss how my previous experience through my clinical role and in my private life, may have influenced any decisions made during the study (May and Perry, 2017, p. 3). The impact my experience had on the design, conduct and analysis of the study was discussed with my supervisors, PPI panel and expert advisory group, including challenging any decisions or interpretations being made.

Conclusion

Decisions about the timing of gastrostomy placement are complex from a clinical, emotional and organisational perspective. The emotional response to initiating discussions and making decisions trigger avoidant coping strategies which oppose healthcare professionals’ preferences for being proactive. The time-limited nature of the decision highlights the need to develop interventions that empower people with MND to engage in discussions and provide enough information to make informed decisions about the timing of gastrostomy placement.

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6.6 Summary of the chapter

The chapter above has presented the findings from the multiple case study that have contributed to answering the research question:

> 'How do healthcare professionals, operating within MDTs, support people with MND to make decisions about gastrostomy placement?'.

The findings are summarised below:

- An overarching finding was that there was a misalignment between the preferences, perceptions and priorities of healthcare professionals and people with MND about the initiation of discussions and timing of decisions.
- Healthcare professionals' preference to be proactive was in contrast to the use of avoidant coping strategies by people with MND to help them live with the disease.
- Informed by their experience of MND, healthcare professionals' proactivity was motivated by a goal to reduce future risks and promote the autonomy of people with MND.
- Decision-making discussions disrupted the coping strategy people with MND employed to extend a sense of normality and avoid discussing future disease progression or care options.
- People with MND frequently experience an emotional response to discussing gastrostomy even though it was not always overtly evident during interactions with healthcare professionals.
- A lack of concrete guidance on the timing of gastrostomy in addition to the avoidant behaviours employed by people with MND, contributed to delays in decisions.
- Some people with MND require healthcare professionals to validate that they have reached the optimal time to accept a gastrostomy even in the presence of physical indicators for the intervention.
- The long waits for gastrostomy placement following referral informed healthcare professionals' motivation to discuss gastrostomy early and to encourage people with MND to make timely decisions.

7 Multiple case study: MDT supported decision support (Paper 4)

7.1 Chapter outline

The following chapter focuses how MND teams coordinate the decision support they deliver across the MDT. The relevant literature is reviewed, followed by a summary of the methods used. Recognising the intention to submit at least two papers based on the same multiple case study, the methods between Papers 3 and 4 have been summarised differently to avoid self-plagiarising. Further detail and justifications about the methods can be found in section 3.9. A rich and in-depth report of the findings is provided followed by a comparison of key findings with the current literature.

7.2 Multiple case study research question

> 'How do healthcare professionals, operating within MDTs, support people with MND to make decisions about gastrostomy placement?'

7.3 Further information related to the study

The following additional information is included as appendices that is related to the development and conduct of the multiple case study (these materials apply to Paper 4 presented in Chapter 7 as they were applied across all three cases):

- Appendix 7 Multiple case study protocol
- Appendix 8 Interview topic guide for people with MND
- Appendix 9 Documentation review proforma
- Appendix 20 Medical note review proforma
- Appendix 21 Scoping email sent to potential cases
- Appendix 22 Screening email sent to interested cases
- Appendix 23 Screening questionnaire sent to interested cases
- Appendix 24 Characteristics of the MND services on Cases 1, 2 and 3
- Appendix 25 Characteristics of people with MND interviewed

7.4 Publication plans

The paper presented below is currently being prepared for submission for publication in peer reviewed journal. Ideally, I would like to publish in a MND or neurology focused journal though these often prioritise quantitative research or the restricted word counts would not allow a rich and in-depth report to be included. Therefore, I am considering journals that publish qualitative healthcare research including Health Expectations, PLOS ONE, Qualitative Health Research.

7.5 Paper 4: Coordination of decision support about gastrostomy placement across the motor neuron disease multidisciplinary team: a multiple case study

Abstract

In MND care, several healthcare professionals from different disciplines can be involved in supporting people with MND to make decisions about gastrostomy. Little is known about how decision support is coordinated across the MND multidisciplinary team (MDT) care model which is distributed across multiple healthcare professionals, settings and organisations. The aim of this study was to understand how healthcare professionals coordinate decision support across the MDT.

Three UK MND clinics and the associated community teams were selected as cases to take part in a qualitative multiple case study. Data collection on each case included 60 non-participant observations of healthcare professionals and people with MND, 39 interviews of healthcare professionals, people with MND and caregivers, medical note review, documentation review and 3 mixed profession focus groups. Framework approach was used to analyse the data.

The influence of two contrasting models of MND care on the coordination of decision support delivered about gastrostomy was explored. Within Cases 1 and 2 most healthcare professionals were restricted to either the clinic or community setting and responsibility for supporting decisions about gastrostomy were distributed over several healthcare professionals. In contrast within Case 3, the same healthcare professionals covered all settings, and most responsibility for decision support about gastrostomy was taken on by a single HCP. Cases 1 and 2 experienced challenges coordinating decision support in relation to the quality and accessibility of communication between healthcare professionals, limiting awareness of the current indication for gastrostomy or of any previous progress made with decision-making. Assigning responsibility to a single healthcare professional reduced the requirement for complex flows of information between healthcare professionals and across settings. There was a lack of an MDT strategy within all cases in relation to the organisation of gastrostomy decision support. There was evidence that the purposeful use of the home setting to discuss gastrostomy where people with MND were more comfortable and could be supported by family, allowed a focused discussion.

Several healthcare professionals separated by geography, profession and organisation are involved in supporting people with MND to make decisions about gastrostomy. Challenges with interprofessional communication and a lack of MDT cross-setting strategy disrupted the coordination of MDT decision support. MND teams should develop a strategic approach, grounded on strong lines of interprofessional communication to coordinate the decision support they deliver to people with MND.

Introduction

Following diagnosis, people with Motor Neuron Disease (MND) are supported by a range of healthcare professional disciplines working across several settings including out-patient clinics, hospices, hospital wards and in the homes of people with MND, organised into a multidisciplinary team (MDT) (OPM, 2016). Most specialist MND care is delivered by a clinician led MND clinic, with support from a range of other clinical specialities, nurses and therapists working in the clinic and community setting (OPM, 2016; Hogden *et al*., 2017). The healthcare professionals involved in the care of people with MND often have their own area of clinical expertise such as speech and language therapists (SLT) focusing on speech or swallowing problems (Rosen *et al.,* 2018). The combination of each professions knowledge and skills enables the MDT to identify, assess and address the range of physical, social and psychological consequences of MND, aiming to improve clinical and quality of life outcomes (Al-Chalabi, 2007). Attendance at a specialist MND MDT clinic has been found to confer a survival benefit and improved quality of life for people with MND compared to other models of MND care (Van den Berg 2005; Aridegbe *et* al., 2013; de Almeida *et al.,* 2021). However, MDT clinics were not necessarily introduced to improve survival, but to allow people with MND access to MND expertise to 'tackle the moving goalposts this disease presents' (Al-Chalabi, 2007). The mechanisms behind the survival benefit of MDT care for people with MND remains uncertain, with earlier access to life-prolonging interventions such as non-invasive ventilation (NIV) or gastrostomy feeding tube being one possible mechanism (Miller *et al.,* 2009a; Berlowitz *et al.,* 2023). People with MND value the information and support provided by healthcare professionals when making decisions about interventions including gastrostomy (White *et al.,* 2023). Therefore, the timing of interventions could reasonably be hypothesised to be influenced by when and how healthcare professionals support people with MND to make decisions.

The loss of multiple functions associated with MND, including upper limb weakness and dysphagia, explain the high risk of malnutrition and aspiration in MND (Genton *et al.,* 2011; Perry *et al.,* 2021). Malnutrition and aspiration are both associated with negative survival and quality of life outcomes for people with MND (Burkhardt *et al.,* 2017; Ning *et al.,* 2019; Janse van Mantgem *et al.,* 2020). It is routine practice for people with MND to be offered gastrostomy to provide a route for nutrition, hydration and medications, when unable to safely take adequate amounts via the oral route (NICE, 2019).

The decisions about gastrostomy placement in MND care are complex for several clinical and contextual reasons (Stavroulakis *et al.,* 2014). First, the intervention is associated with a time when people with MND will no longer be able to eat and drink; a basic human function that has significant social and psychological value attached to it (Zarotti *et al.,* 2019; Coates *et al.,* 2023). Second, a range of factors indicate a need to consider gastrostomy including dysphagia, weight loss, and respiratory failure each of which have uncertain times of presentation and rates of progression (Van Eenennaam *et al.,* 2021). Third, gastrostomy is a time limited decision with escalating procedural risks associated with increasing respiratory failure and frailty, and declining benefits of enteral nutrition (Kasarskis *et al.,* 1999; ProGas Study, 2015). Professional guidance recommends healthcare professionals to start discussions with people with MND about gastrostomy early in the disease course and support a clinical preference for placement of gastrostomy prior to significant swallowing, nutritional or respiratory decline (Miller *et al.,* 2009b; NICE, 2019). With people with MND valuing the MND expertise of healthcare professionals, the MND team play an important role in guiding decisions about not just whether to have a gastrostomy placed, but just as importantly, when to have a gastrostomy place (Stavroulakis *et al.,* 2014; Greenaway *et al.,* 2015; Martin *et al.,* 2016; White *et al.,* 2023). Finally, being asked to consider gastrostomy can evoke an emotional response in people with MND that can present a barrier to engagement in discussions or delays making decisions (White *et al.,* 2024; see also Paper 3 in Chapter 6).

The coordination of decision support across MDTs is influenced by the quality of inter-professional care, including the communication between healthcare professionals (Légaré *et* al., 2011; Brogan *et al.,* 2019). Surveys of healthcare professionals in the UK, Netherlands, and Canada have identified that a range of professional disciplines support people with MND making decisions about gastrostomy including doctors, nurses, dietitians and speech and language therapists (Rio and Cawadias, 2007; Ruffell *et al*., 2013; Van Eenennaam *et al.,* 2021; White *et al.,* 2024). Challenges have been identified in qualitative literature in relation to the coordination, consistency and accuracy of information shared between HCPs (Hogden *et al.,* 2012; Chapman *et al.,* 2021; White *et al.,* 2023). A survey of UK healthcare professionals revealed a lack of consensus about how people with MND are informed about their options, concerns about consistency of information shared by the MDT and challenges associated with inter-professional communication (White *et al.,* 2024).

Further research is required to better understand how MND teams organise and deliver decision support to people with MND considering gastrostomy placement, in the real-world setting. The aim of this study was to understand how healthcare professionals coordinate the decision support about gastrostomy across settings.

Methods

Multiple case study design

A qualitative multiple case study was chosen as a study design to allow the study of MDT supported decision making about gastrostomy placement within the context of UK MND MDT care delivery (Yin, 2018). Multiple qualitative methods were used to explore how the decision support delivered to people with MND was coordinated across the MDT from the perspectives of the different people involved (Crispin *et al.,* 2017). The inclusion of multiple cases, and comparisons between them, allowed for theoretical generalisation in relation to how the conceptual findings may apply to settings wider than those studied (Ritchie *et al*., 2014, p. 352-354).

Case definition and boundaries

A case was defined as the ‘the MND multidisciplinary team operating across the clinic and community setting that supported people with MND to make decisions about gastrostomy’. The boundaries of each case were restricted to the actions, perceptions, and interactions, of people with MND, caregivers, and healthcare professionals making decisions about gastrostomy placement.

Case selection

Access to observe the phenomenon of interest is a primary objective of case study research (Stake 2006, p. 24-27). All MND Care Centres and Networks listed on the MND Association website were approached to express an interest in being case study site for the proposed research. Using purposive sampling, three MND teams were selected based on their responses to a screening questionnaire about their MND care model, MND caseload and location.

Participant sampling

Purposive sampling was used to recruit participants to each data collection method and when selecting the healthcare professionals’ discussions with people with MND, caregivers and other healthcare professionals, to observe. Non-participant observations of healthcare professionals and people with MND were sampled to ensure variation in location of discussion, stage of decision making (first discussion, follow-up discussion, decision made) and healthcare profession involved. Non-participant observations of MDT meetings were sampled by setting, seeking to observe interactions in the clinic and community setting. People with MND were sampled to be interviewed by current decision (accepted, declined or not made decision). Healthcare professionals recruited to interviews and focus groups were sampled by profession, seeking to recruit professional disciplines identified within each case as being involved in supporting people with MND to make decisions about gastrostomy.

Recruitment

The principal investigator (PI) within each case was crucial for gaining access to the study site and for successful recruitment. The PI invited healthcare professionals and people with MND, verbally or through sending invitational materials, to express an interest in being involved in the study. Those interested in participating contacted the PI or researcher to learn more about the study and to provide consent to taking part.

Inclusion criteria

The inclusion criteria for people with MND involvement in the study included a confirmed diagnosis with MND, attending the MND clinic within the case and if already had gastrostomy, it was placed less than a month ago. The inclusion criteria for caregivers included being a non-paid informal caregiver of a person with MND recruited to the study. The inclusion criteria for healthcare professional participants included that they supported people with MND attending the case MND clinic to make decisions about gastrostomy and were a qualified healthcare professional.

Consent

All participants read a participant information sheet and given the opportunity to discuss the study with the researcher, before signing a consent form.

Data collection

A sequential approach was taken to data collection beginning with non-participant observation, followed by semi-structured interviews with people with MND, caregivers and healthcare professionals, and ending with a mixed profession focus group (see figure 1). Most participants (people with MND and healthcare professionals) observed were often also interviewed or took part in the focus group (healthcare professionals only), while others only participated in a single data collection method. Any documentation relating to the discussions about gastrostomy recorded in the medical records of those people with MND interviewed were extracted. Healthcare professional participants were asked to share any documents that may relate to how people with MND are supported to make decisions about gastrostomy. Table 1 describes the aim of including each data collection method in the case study.

Figure 1. Sequential approach to recruitment and data collection within each case.

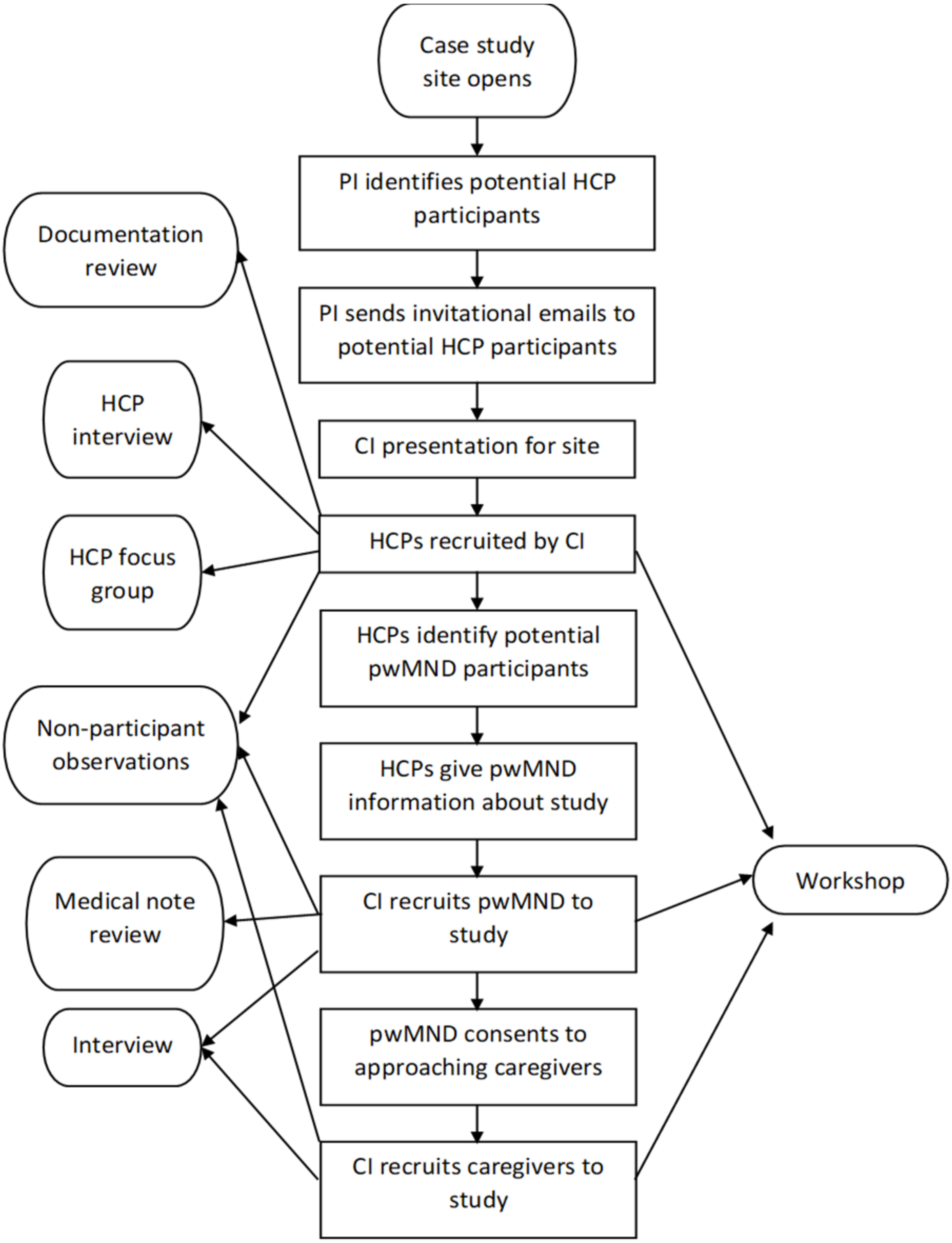


Table 1. Summary of case study data collection methods. *the audio recording of the focus group on Case C failed, so detailed field notes were recorded immediately afterwards and included in the analysis.

Data collection method	**Aim of method**	**Data collected**
Non-participant observation of discussions between healthcare professionals and people with MND, their caregivers and other healthcare professionals (e.g. MDT meetings)	Understand how healthcare professionals, people with MND and caregivers interact when discussing the option to have a gastrostomy.	Field notes recorded by the researcher taken during and after each observation
Healthcare professional interviews	Understand healthcare professionals' experience of supporting people with MND considering gastrostomy.	Professional transcriptions of the audio recordings of each interview.
Interviews of people with MND and caregivers	Understand the experience of people with MND and caregivers of being supported by the MND MDT to make decisions about gastrostomy.	Professional transcriptions of the audio recordings of each interview.
Medical note review	Understand healthcare professionals documented version of supporting people with MND making decisions about gastrostomy.	Extracts taken from medical notes by researcher.
Healthcare professional focus group	Understand how the MND MDT support people with	Professional transcriptions of the audio recordings of each focus group.*

	MND making decisions about gastrostomy.	
Documentation review	Understand what documentation guides healthcare professional practice	Electronic copies of documentation.

Ethics

The study was granted ethical approval by West of Scotland Research Ethics Service (REC reference: 22/WS/0171) on 14/12/2022.

Analysis

The textual data collected was managed in NVivo CAQDAS software. The framework approach developed by Ritchie *et al.,* (2014) was used to analyse the data. An a priori framework was structured around key stages of decision making developed from findings of a previous qualitative evidence synthesis and healthcare professional survey (White *et al.,* 2023; White *et al.,* 2024) including 1) initiation of discussions; 2) process of decision making; 3) making of decisions; 4) organisation of MDT decision support. The data collected within each case was coded using the framework with new themes and sub-themes being inductively developed as indicated. Data summaries for each sub-theme were added to matrices in NVivo for each unit of analysis (i.e. each observation, interview, focus group). The data summaries were used to identify key indicators and dimensions associated with the themes. Finally, an iterative revisiting of the matrices and original data, informed a process of pattern matching to develop the themes presented in the findings below.

Findings

Across the three cases, 42 interactions between healthcare professionals and people with MND and their caregivers were observed (35 in clinic and 7 in the home of people with MND) and 18 MDT meetings were observed, 12 people with MND were interviewed and had their medical notes reviewed, 9 caregivers were interviewed (3 with the person with MND), 17 healthcare professionals (including 8 different disciplines) were interviewed, 20 healthcare professionals (including 9 different professional disciplines) participated in 3 focus groups and 21 documents were reviewed. The characteristics of the people with MND interviewed are included in appendix 25. Field work took place between March 2023 and January 2024.

The characteristics of each case are summarised in table 2. The cases were from different regions of the UK (details not reported to protect the anonymity of the services involved and participants recruited). All cases provided MND specialist MDT care across the clinic and community setting. They varied slightly in relation to the size of their caseloads (n=130-232) and number of new referrals each year (n=37-100). A key difference between the cases was that people with MND attending MND Clinic within Cases 1 and 2 were supported by multiple community teams in the region. In contrast, within Case 3 people with MND were supported by broadly the same group of healthcare professionals in the clinic and community setting.

Table 2. Characteristics of the MND teams on Cases 1, 2 and 3.

Characteristic	Case 1	Case 2	Case 3
MND caseload	255	130	171
New MND diagnoses per year (approximate)	100	37	70
MND Clinic	Weekly MND clinics hosted in a large teaching hospital out-patient department.	Monthly MND clinics hosted in each of two different hospital out-patient departments in the region.	Monthly MND clinics hosted on two sites in the region (one hospital outpatients and another in an old GP surgery).
Professional disciplines present in MND clinic	Neurologists, MND CNS and respiratory technician. Bimonthly MDT clinics are also	Neurologist, MND nurses, MND coordinator, respiratory clinicians,	Neurologist, respiratory consultant, respiratory nurse,

	attended by dietitian, SLT, physio, OT and social worker. Respiratory consultant and nurse/physiotherapist attends clinic monthly. Palliative Care Consultant attends clinic once a month.	respiratory nurse and palliative care consultant.	palliative care consultant, dietitian, SLT, respiratory physiotherapist, research nurse and MND coordinator
MDT meetings	Bi-monthly hospital-based MDT meeting attended by the care centre MDT. Monthly MDT meeting in respiratory centre attended by ventilation CNS, neurologists, palliative care consultant and MND CNS. Monthly MDT meetings in each community locality attended by range of community healthcare professionals and MND CNS from MND centre.	Monthly MDT meetings attended by MND clinic team and HCPs from the acute service. Periodic (usually monthly) MND MDT meetings in each community locality attended by range of community healthcare professionals and MND CNS from MND centre.	Face to face site-based MDT meeting before and after each MND clinic. 3 monthly regional MDT meeting attended by the team.

Community services	Fourteen community localities across the region belonging to different NHS Trusts include cross-section of allied healthcare professionals (AHP) and nursing roles.	A large number of community localities across the region belonging to different NHS Trusts and including variety of nursing and AHP roles.	Most healthcare professionals in clinic visited people with MND at home between clinic appointments. There were no separate community therapy teams except for community palliative care team and occupational therapy (see below)
Respiratory services	Respiratory service hosted in different hospital and NHS Trust. Respiratory consultant attend monthly MND clinics in the MND centre. Respiratory centre runs its own respiratory out-patient clinics. Respiratory CNS team provide out-patient clinic and community out-reach services.	Respiratory consultants and nurses core part of MND MDT, attending all MND clinics and MDT meetings.	Respiratory consultants and nurses core part of MND MDT, attending all MND clinics and MDT meetings.
Palliative care services	Palliative care consultant attends a monthly MND centre	Palliative care consultant attends MND clinics. People	Palliative care consultant core member of MND

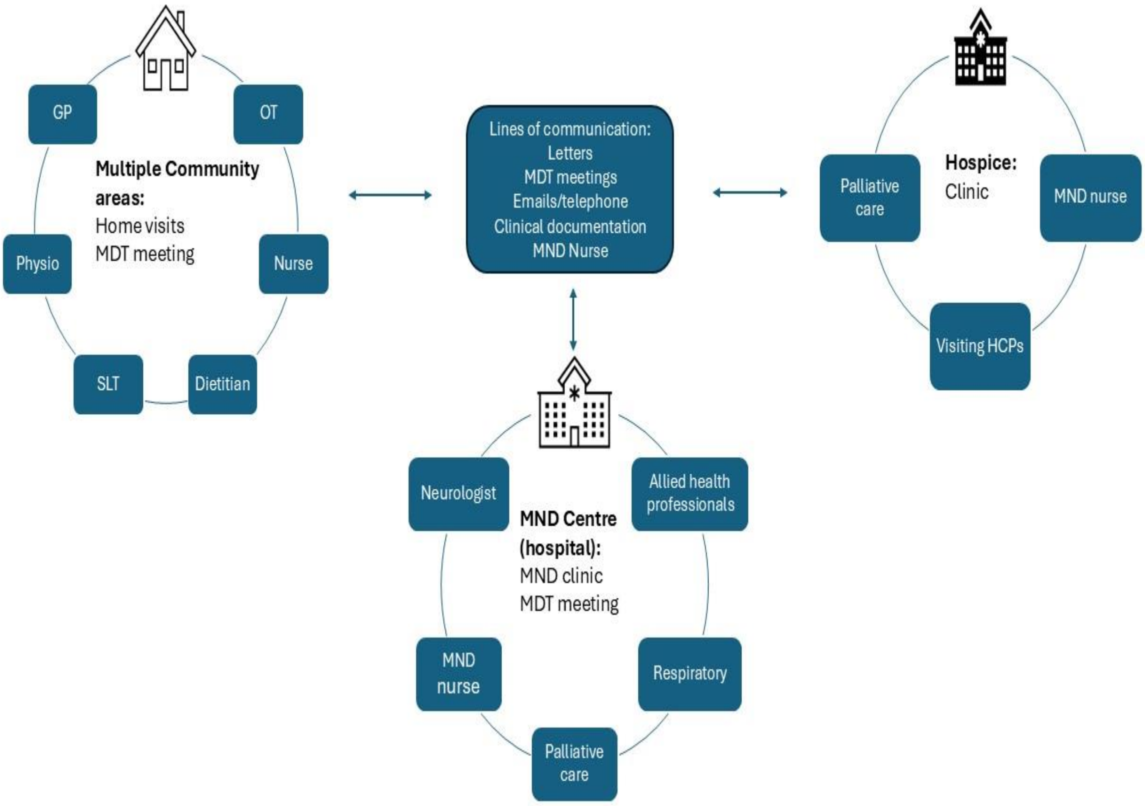
	clinic. MND clinics also run in two regional hospices, attended by palliative care, neurologist and MND CNS.	with MND can also be referred to their local hospice or community palliative care teams.	clinic MDT, seeing all people with MND from diagnosis. Can also refer into the community palliative care team.

The organisation of services providing healthcare to people with MND, including those involved in discussions about gastrostomy was complex and varied within each case. Figure 2 captures a generic schematic of how MND services were organised and the lines of communication between them. Each case though differed in their organisation and lines of communication. Key differences between the cases were:

- Within Case A:
    - the respiratory services were hosted within a different hospital (and NHS Trust).
    - Joint clinics (including palliative care, neurologist and MND nurse) were held in two local hospices.
    - Only the respiratory nurses reviewed people with MND in the clinic and community setting.
    - MND Centre, community and respiratory team services often belonged to different organisations.
- Within Case B:
    - Only the MND nurses reviewed people with MND in the clinic and community setting.
    - Did not have any Allied Health Professionals e.g. dietitians, physiotherapists, occupational therapists, represented in the MND clinic.
- Within Case C:
    - There was no MND nurse in the MDT.
    - Most healthcare professionals were able to review people with MND in clinic and home setting.
    - All healthcare professionals belonged to the same healthcare organisation.

- In contrast to Cases B and C there were not multiple community teams involved.

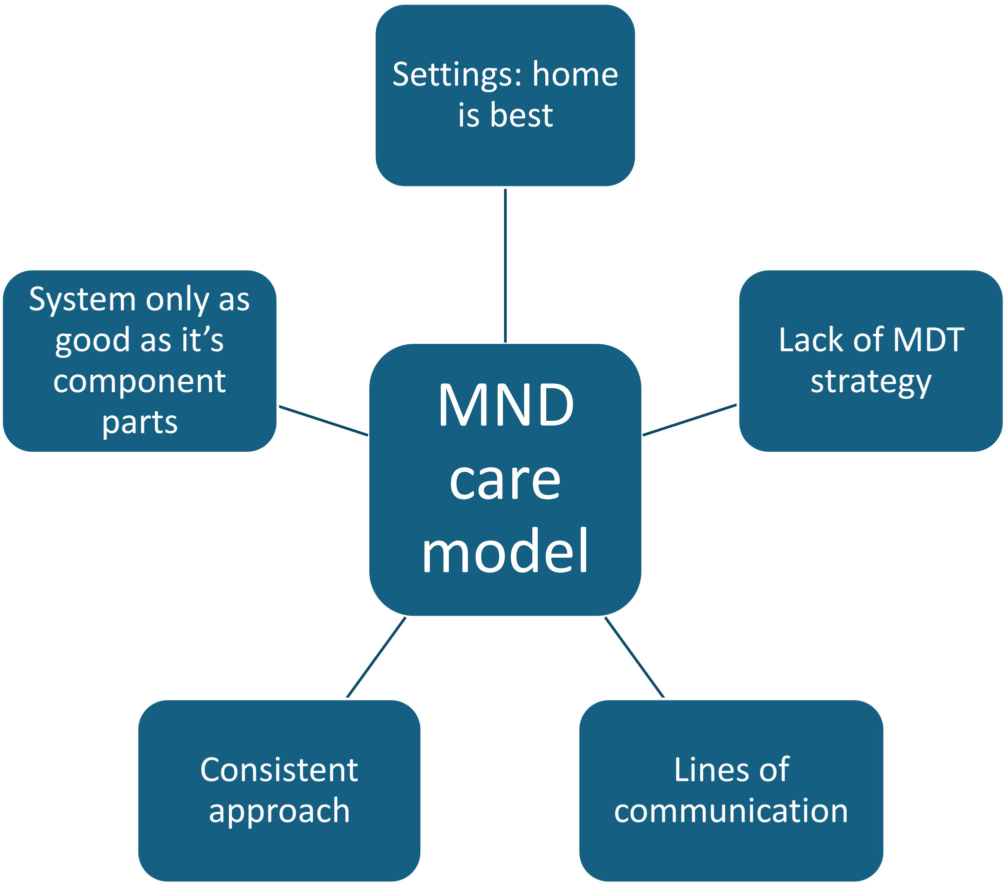
Figure 2. Schematic of MND services and the lines of communication between them.



Overview of MDT decision support for gastrostomy

People with MND receive information and support from several healthcare professionals in a variety of settings when considering their option to have a gastrostomy. The analysis identified an overarching theme of how the model of MND care and the organisation of the HCPs within it, shapes how decision support can be delivered and coordinated. Six themes were developed that relate to the model of care, settings, strategic approach, lines of communication, consistency between HCPs, and the knowledge and skills of the HCPs operating within each case. The themes are summarised in figure 2.

Figure 3. Themes relating to the coordination of MDT decision support about gastrostomy.



Model of MND multidisciplinary care shapes how decision support can be delivered

The model of MND multidisciplinary care delivery within each case shaped how local MDT supported people with MND considering the gastrostomy option. Discussions with people with MND about gastrostomy were initiated and continued in a range of settings, by a variety of healthcare professionals distributed over large geographical regions. People with MND all returned to a central MND clinic every 3 months. While healthcare professionals were organised into one or more distinct MDT's, the professional roles, responsibilities, knowledge, boundaries and connections between them varied considerably.

Two models of MND care were purposively selected as cases. Cases 1 and 2 included several community teams spread across the region who cared for people with MND attending a tertiary MND clinic within the MND centre. Community services included a mix of nursing and allied healthcare professionals (AHPs) and often lacked medical representation. The clinics were medically lead (i.e. by neurology or palliative care consultants), supported by

MND CNS, palliative care, respiratory team and in Case 1 only, a range of AHPs (see table 2). While the MND CNS or coordinators were the only professions observed to be working across settings, only the respiratory CNS in Case 1 and MND CNS in Case 2 were able to visit people with MND both in clinic and their own home. The organisation of MND care presented healthcare professionals within Cases 1 and 2 with challenges delivering consistent and coordinated decision support across settings which will be described in the following themes.

In contrast, Case 3 involved a more streamlined model of MND care reported to have been purposely designed to have the same group of healthcare professionals reviewing people with MND in the clinic and community setting. It was evident from observations and interviews that a single healthcare professional took on most of the responsibility for monitoring the need for, initiating and continuing discussions about gastrostomy. While other team members contributed to discussions, one healthcare professional took a lead in supporting the decision-making of people with MND about gastrostomy. The smaller number of healthcare professionals reviewing people with MND across settings and a clarity of roles, avoided the need for multiple lines of communication and the increased effort required to coordinate decision support.

> ‘it would be really difficult to have that carryover between if there was a clinic team and then a community team. Because I think you get so much information…most people, like they might pop in and see people between the clinics and then it’s just information that then they can share. It’s not like if I was solely a clinic therapist that then I just saw them every three months. I wouldn’t really feel like I knew them as well as being able to do them both.’ (Case 3 healthcare professional interview).

Settings for discussing gastrostomy: home is best

Conversations about gastrostomy occurred in the clinic, hospice, ward and home setting, with benefits and challenges associated with each. While people with MND value the expert and compassionate support they experienced during clinic visits, they may not always be the most appropriate setting in which to have the complex and emotional discussions about gastrostomy. Clinic visits are typically lengthy, involve long journeys or waits for hospital transport, and include multiple discussions about disease progression and the need to consider interventions. As a result, people with MND can be left emotionally and physically fatigued, overwhelmed by the information they have been given and not always able to accept or engage in meaningful discussions about gastrostomy. People with MND and healthcare professionals consistently identified the home as the most appropriate setting in which to have discussions about gastrostomy. Benefits included people with MND being in a familiar environment, easily allowing family to be present, less time pressure and enabling a focus on the gastrostomy decision. Particularly within Case 1, some HCPs identified the inability of MND specialist healthcare professionals to visit people with MND at home as a limitation in terms of being able to adequately support their decision-making. The clinic setting was often observed to be used to 'plant the seed' about the option, while planning to visit at home shortly after to have a more detailed discussion. Such a planned approach to stagger the discussion was appreciated by people with MND and caregivers, as an effective way to gently introduce the option as it allowed their initial response to be tested while being reassured that further detail would be shared soon after in the home setting.

> 'Sometimes, with all the seeing all the doctors at once, it's a bit too much to take in, and it can be quite a heavy day. But then when they come individually then, and they sit down and have a chat with you in the front here [the home of the person with MND], then you feel a bit more focused and it's easier to make a decision, than rushing into a decision there…' (Case 3 interview with person with MND)

Lack of a cross-setting MDT strategic approach to decision support

Absence of decision support care pathways

In all cases there did not appear to be a strategic plan for how decision support would be coordinated across the wider MDT. While Case 3 had developed guidance identifying when healthcare professionals should discuss gastrostomy and Case 1 were undertaking a system wide review of their gastrostomy pathway, the focus was more on the indications for enteral nutrition and organisation of the procedure, rather than how the MDT support people with MND to make decisions. There was an absence of MDT consensus about when discussions should start, the responsibilities of healthcare professionals, the information that should be shared and how to support decisions about timing of gastrostomy placement. In all cases, discussions tended to start and continue organically, in response to a range of factors to be reported elsewhere (See Chapter 6, Paper 3).

Role and responsibility for decision support

Roles and responsibilities observed taken on by healthcare professionals included assessing need for discussing gastrostomy, advance care planning, introducing the option, informing about what the intervention involves, discussing the risks and benefits, and guiding decisions about the timing of gastrostomy placement. In contrast to Case 3 where one healthcare professional took on most of the responsibility for discussing gastrostomy, in Cases 1 and 2 many professions across the MDT independently took responsibility for one or more of these roles. None of the teams responsible for placing gastrostomy tubes, i.e. gastroenterology or radiology, were observed supporting the decision-making of people with MND about gastrostomy in any of the cases and often only got involved following a decision having been made. The lack of involvement of gastrostomy placement teams was a concern for one Case 3 caregiver, who expected those placing the tube to be involved.

> ‘[caregiver] stated he was very upset by this and explained that he needed this information about the risks involved so that they could make a decision. [caregiver] stated that she needed to be assessed by radiology and that he wished to discuss this with a doctor. [caregiver] questioned how the decision can be made to not have a gastrostomy without there being an assessment performed by radiology.’ (Fieldnotes from an observation of an interaction between a person with MND, caregiver and healthcare professional within Case 3)

Within cases where the responsibility for discussing gastrostomy was distributed more widely across the MDT (i.e. Cases 1 and 2) healthcare professional interviews and observations revealed some repetition of assessments or discussions, and a risk of inconsistent or contradictory messaging, including within a single clinic visit.

> ‘Healthcare professional: I had him [person with MND] in here with the family, the palliative consultant and the respiratory doctor and their thoughts were it was best to leave it alone and I was like that’s what they think, that’s their decision, that’s great and then [neurologist] had felt differently, so he was talking about it again, but maybe that was our miscommunication.

> Researcher: In the actual, in one clinic was that?
>
> Healthcare professional: Yes.’ (Case 2 healthcare professional interview)

Revisiting the decision

Revisiting the decision for those people with MND choosing to continue without or declining gastrostomy also appeared to lack a strategic approach. Coordinating why, when and who, revisited the decision presented an organisational challenge particularly when the responsibility for subsequent discussions was to be delegated to another healthcare professional. For example, within Case 1, healthcare professionals reported uncertainty about who would follow up a discussions and concern about whether colleagues had the knowledge and skills to discuss the option appropriately with people with MND.

> ‘we do feel it sort of goes out and we're not entirely sure exactly what has followed up and with who and what exact conversations take place. So that's the bit of the pathway that I think we really need some further work on.’ (Case 1 healthcare professional interview)

Some healthcare professionals reported having good relationships with specific MDT colleagues and used a ‘tag team’ approach to prompt further assessments or discussions. Less rigorous approaches to planning further discussions included asking the person with MND to initiate contact with their local healthcare professional, assumptions that such discussions would take place and inclusion in clinic letters which were open to interpretation. Healthcare professionals and caregivers acknowledged the risks associated with relying on people with MND to prompt discussions, with people with MND not always being aware or accepting of the significance of functional changes necessitating HCP interpretation and explanation.

> ‘I think if you put ‘refused’ then that might just if a colleague is reading that they may look at it and think oh well, they’ve made their mind up, but they may not have made their mind up.’ (Case 1 Healthcare professional interview)
>
> ‘Patients don't know what they don't know about MND, which is where it's really difficult when somebody says give us a call if you need me. Because they don’t

> always know when they need you, do they.' (Case 1 Healthcare professional interview)

Lines of communication required to keep everyone on the same page

A lack of sharing or access to information about the current or future indication for gastrostomy of people with MND and what progress had previously been made with decision-making impacted on the coordination of MDT decision support.

Modes of communication

All modes of communication between healthcare professionals, including meetings, clinical notes, letters, phone calls and emails, were documented in an electronic patient record (EPR), providing a repository of information about assessments, what discussions had taken place and plans for future action; for those who could access it. However, when healthcare professionals belonged to different organisations (i.e. Cases 1 and 2), they often could not access each other's EPRs. Even within the same organisation, some healthcare professionals' records were not accessible to other members of their MDT, limiting their effectiveness as a method of communication. The utility of letters as a method of communicating with the MDT was limited by which healthcare professionals were copied in or would have access the EPR they were saved on.

> '[Healthcare professional] advised [person with MND] that the respiratory services are from a different hospital and that this can impact on the communication between services.' (Field notes from the observation of a Case 1 MND clinic appointment)

MDT meetings presented an effective forum for healthcare professionals to share information, clinical reasoning and to discuss patient cases; for those healthcare professionals who were present. During MDT meetings, mutual professional respect was observed between those healthcare professionals present, with all present contributing to the discussions and little evidence of hierarchies or power imbalances. Representation at MDT meetings was often limited to healthcare professionals working in the setting in which they took place with usually only the MND CNS or coordinator attending meetings in all settings. The MND CNS acted as an important communication link between the clinic and community teams in relation to indications to discuss or place gastrostomy, the discussions having taken place and any decisions made. Healthcare professionals not present at MDT meetings did not benefit from an awareness of the added contextual information shared that may not always be included in written documentation. Having one MND MDT on Case 3, meant most healthcare professionals were present in all MDT meetings. Healthcare professionals within Case 3 met as a team before, during and after clinic, in a central meeting room allowing information to be shared about the discussions had in the community since the previous clinic, prepare for and feedback about discussions in clinic, and plan for discussions required after clinic. Healthcare professionals on Cases 1 and 2 reported challenges having discussions with colleagues during clinic due to their busy nature and acknowledged the benefit of having pre or post clinic MDT meetings though they were not able to implement these currently. While MDT meetings provided an opportunity for case discussions, they were time-limited, covered many different clinical priorities, needed to allow for many healthcare professionals to speak and did not include decision-making about gastrostomy as a fixed agenda item. Healthcare professionals on all cases identified the lack of broad professional discipline representation, time and attention on gastrostomy decision-making as a limitation of MDT meetings as a source of effective communication and for agreeing a team approach.

Awareness of previous discussions

Healthcare professionals were frequently observed on Cases 1 and 2 to lack an awareness of the previous assessments and discussions with people with MND in relation to gastrostomy or what the outcome of any discussions were. A lack of detail shared in some professional communications presented a barrier to healthcare professionals' awareness of the previous discussions had with people with MND. Often documentation included only brief accounts of discussions, lacking nuance about the information shared, the response of people with MND, preferences or reasoning, or any concrete plans for follow up. There was evidence of some very detailed summaries of discussions, though these were more frequently observed in clinical notes rather than letters. Some healthcare professionals reported being selective about when they felt it necessary to share information with the MDT, while others did not feel a lot of detail was required. Healthcare professionals on all cases trusted their MDT colleagues to be having the 'right conversations' or made assumptions these discussions were occurring elsewhere without necessarily having any evidence for it. Healthcare professionals often relied on people with MND and/or caregivers to provide subjective updates about their current status or previous discussions. However, people with MND were described by healthcare professionals and caregivers as not always being the most reliable source of information due to being poor historians or through not wanting to openly acknowledge a decline in their function.

> ‘there's other teams that have got really useful information and you're relying heavily on the patient to tell you what they said and what they found. Which is sometimes not as accurate.’ (Case 1 healthcare professional interview)

Healthcare professionals routinely reviewed previous documentation when preparing for a consultation. However, within no cases was there a single documentary source that synthesised the information required to understand the current need to discuss or place a gastrostomy. Healthcare professionals had to search through many documentation entries to find the information needed which was a source of frustration and risked relevant information being missed. The consequences of a lack of awareness included healthcare professionals not being in possession of the relevant information needed to support the decision-making of people with MND and the risk of repetition or contradicting information already shared previously by the MDT.

> ‘…on these electronic systems, there's so much input on them, it's easy to miss that little conversation that says oh, I'm not sure I want this right now or, there should be, yes, planning and letting, communications really important.’ (Case 1 healthcare professional interview)

Strengthening the lines of communication

A number of other strategies were observed that promoted sharing of information between the MDT. Healthcare professionals on all cases performed joint visits or co-populated clinic letters (cases 1 and 3 only) supporting their awareness of others assessments and discussions. The MND team within Case 1 were developing a new IT platform which would present a range of relevant information in one source and had potential to inform healthcare professionals’ discussions about gastrostomy. Structured consultations, databases or clinic proformas were used to prompt discussions and to keep healthcare professionals updated about the indication of people with MND for gastrostomy.

Delivering a consistent approach across the MDT

Most people with MND and caregivers interviewed reported that the information shared by different healthcare professionals was relatively consistent. However, there was evidence of differences in how healthcare professionals framed the decision. Some healthcare professionals were reported to vary in relation to how direct they were about the outcomes of delaying decisions or could challenge decisions made in previous discussions. For example, within Case 1 there was a difference reported between how the healthcare professionals in the MND and respiratory centres framed the information they shared with people with MND about the timing of gastrostomy.

> ‘I think from dealing with the MND team, I think they're very kind and very hand holdy…And sometimes I'm a bit surprised that they don't have information that I would expect them to have had from their diagnosing team’ (Case 1 healthcare professional interview)

> ‘how the respiratory consultant spoken to me that was more powerful in making my decision. The other people they were saying, but they can't like they haven't explained well why I need it now and is there any need it or should I go for it or shouldn’t...’ (Case 1 interview with a person with MND)

Most healthcare professionals reported in interviews that the information shared by their MDT was consistent and proposed advantages of MDT involvement in supporting the decision-making of people with MND about gastrostomy. Such advantages included people with MND having access to different perspectives and expertise, or in relation to certain healthcare professionals having better therapeutic relationships with individual people with MND. It was clear from observations there was a uniform clinical preference for gastrostomy to be placed ‘earlier rather than later’. However, some healthcare professionals expressed concerns about having different services involved.

> ‘I guess that maybe a negative and a challenge is it sometimes doesn't always feel particularly joined up. So, we might not be fully aware of what's happening and

> when and I think that's around like the importance of the MDTs that we have and copies of the letters going to relevant people.’ (Case 1 healthcare professional focus group)

> ‘I think there, there's a real disconnect between sort of tertiary services and then the community services and getting that information in.’ (Case 1 healthcare professional interview)

Particularly among Case 1 healthcare professionals there were differences in views of the respiratory and MND services in relation to each other’s approach to informing people with MND about the gastrostomy option. The MND centre were perceived to miss opportunities to encourage earlier decision-making, and the respiratory centre were considered too procedurally focused. In these scenarios a lack of consensus and communication between healthcare professionals contributed to a less a consistent approach. While the healthcare professionals’ professional opinions may have been valid these examples point towards an inconsistency in the reasoning of different professionals within the same MND MDT based on their individual clinical priorities which had the potential to cause decisional conflict if communicated to a person with MND.

> ‘I have felt I have to admit, sometimes very frustrated by conflicting conversations.’ (Case 1 healthcare professional interview)

> ‘So, people can have more, have harmonised conversations. Because the patients are stuck in the middle of this, you know, and most of the time they listen to healthcare professionals. So, you have to be careful.’ (Case 1 healthcare professional interview)

> ‘I think sometimes there's this kind of feeble discussion, that's very gentle and wonderful and lovely. It's not actually actioned anything or that there's no urgency to it. And then of course there is.’ (Case 1 healthcare professional interview)

The system is only as good as it’s component parts

Healthcare professional MND knowledge and expertise

While organisational factors influenced the coordination of decision support across the cases, the function of the MDT as an effective unit of decision support relied on the knowledge and expertise of the individual healthcare professionals involved. Healthcare professionals within the MND clinic team expressed concerns about the MND knowledge of some non-MND specialists (e.g. ward or community HCPs) and their ability to appropriately support the decision-making of people with MND . Within Cases 1 and 2, there was a postcode lottery in terms of the community support available in different regions, and the MND knowledge of those teams that did exist. A lack of dietetic expertise in clinic (Case 2) and in community settings (Cases 1 and 2) was viewed as a gap in relation to supporting gastrostomy decision making.

> ‘we've got 13 community localities within [local area] and I think it was felt amongst different members of the MDT it was quite evident there were varying dietetics services in terms of funding, staffing, expertise’ (Case 1 healthcare professional interview)

Healthcare professionals on all cases referred to how previous MND clinical experience, published research and guidance, and learning from MDT colleagues contributed to their knowledge and the information they shared with people with MND. However, those healthcare professionals not present at MDT meetings did not benefit from this source of learning. Some MND centres had acknowledged their responsibility for improving the knowledge of the wider MDT through organising formal study events, in their correspondence or by educating during interprofessional interactions.

A lack of healthcare professional knowledge had the potential to disrupt the decision-making of people with MND through them not being aware of likely disease progression or the time taken to organise gastrostomy placement. Even within the MND specialist team, healthcare professionals reported lacking knowledge or confidence in discussing certain aspects of the decision including the relevance of respiratory decline, outcomes of declining tube placement, and the technical aspects of the intervention. For example, a caregiver on Case 3 was observed challenging a healthcare professional about whether they were the right person to be assessing the person with MNDs’ fitness to have a gastrostomy. A Case 3 MDT colleague acknowledged the difficult position the healthcare professional was placed in when expected to have discussions about clinical issues that were not a core remit of their role.

> ‘quite a difficult role for her because she’s got to have that – like, she’s got to have that understanding actually why but, actually, her role is reviewing the needs of the nutrition…And so in those situations where it’s been really difficult, if they haven’t got, like, overt dysphagia or anything and it is from respiratory decline, I think it’s been really difficult because, yes, I think she’s had to lead those conversations. And it’s hard.’ (Case 3 healthcare professional interview)

Healthcare professional communication skills

The need for advanced communication skills when discussing gastrostomy was identified by people with MND, caregivers and healthcare professionals. People with MND and caregivers valued when healthcare professionals spoke to them with compassion, empathy and good listening skills. Healthcare professionals acknowledged that not all every professional naturally have these skills and, in some cases, could result in missed opportunities to engage in discussions about gastrostomy. Healthcare professionals on all cases were supportive of being involved from diagnosis, before their clinical input was required, to develop the trust and rapport required to engage in sensitive discussions about disease progression and interventions. Some healthcare professionals believed active listening was essential to understand the goals of people with MND and manage the emotional response to discussing disease progression or need for gastrostomy.

> ‘People need to feel heard, and people's emotions need to be respected and acknowledged. I think that's the first thing we need to do as clinicians and that opens up all sorts of avenues to have very deep, meaningful conversations to fix problems and understand what their wishes are…’ (Case 1 healthcare professional interview)

Drawing on the expertise of specific professional roles

There was evidence of the expertise of certain professions being purposely utilised within cases. For example, people with MND who were delaying or declining gastrostomy were often referred to palliative care teams to discuss the outcomes of these choices and how they will be managed. Indeed, palliative care consultants reported taking a more goals and values-based approach to discussing gastrostomy than other healthcare professionals who may focus more on the clinical indications for the intervention. People with MND who were reluctant to engage in discussion were reported to be referred to medical colleagues, particularly in the presence of a significant need to consider interventions, with a perception that patients may listen more to a doctor. Nurses, dietitians, SLT and doctors were all observed to describe what the intervention involved including demonstrating the equipment to people with MND. So, while these roles or responsibilities were not formally documented, there was evidence of some strategic use of healthcare professionals differing knowledge or skill sets during the decision-making process.

Discussion

MND service design influences coordination of MDT decision support

The coordination of MDT decision support was influenced by the model of MND care employed within each case. In line with professional recommendations and previous research, on all three cases people with MND were supported by a range of professional disciplines across the MND clinic and community setting, many of whom were involved in supporting decision-making about gastrostomy (Miller *et al.,* 2009a; NICE 2019; Van Eenennaam *et* al., 2021; White *et al*., 2024). However, there were two key differences between the MND care models which influenced how healthcare professionals were able to coordinate decision support about gastrostomy across the MDT. First, within Cases 1 and 2 people with MND attended a tertiary MND clinic and were supported by one of several community teams depending on where they lived in the region. In contrast, within Case 3, one single team of healthcare professionals delivered care across the clinic and community setting to people with MND living in the region. Second, in Cases 1 and 2 responsibility for supporting the decisions of people with MND about gastrostomy was widely distributed over several HCPs operating in the clinic and community setting. Within Case 3 most of the responsibility for supporting decision-making about gastrostomy was assigned to a single healthcare professional. The performance of any healthcare team is reliant on agreed responsibilities, effective inter-professional communication, organisation, continuity and coordination (Rydenfalt *et al.,* 2017; Rosen *et al.,* 2018; Soukup *et al.,* 2018). These are qualities identified as important in the current study, supporting the concept that that inter-professional decision support is associated with the performance of the MND MDT (Legare *et al.,* 2011).

Assigning responsibilities for decision support

In contrast to previous research where doctors or the MND Clinic were reported to lead on supporting the decision-making of people with MND , the responsibility for decision support about gastrostomy was often shared by doctors, nurses and AHPs operating in all settings including monitoring indications, introducing and discussing options, and implementing decisions (Hogden *et al.,* 2015; Sohi *et al.,* 2015). Only within Case 3 was a single healthcare professional identified by the MDT as having the majority of responsibility for leading on supporting the decisions of people with MND about gastrostomy. The benefits of the approach in Case 3 were that people with MND and the MDT were clear about the healthcare professional's role, and it reduced the need for several lines of communication between multiple healthcare professionals. Therefore, in Case 3 there was less evidence of the challenges reported in previous qualitative studies of breakdowns in communication and cohesiveness across the MDT (Hogden *et* al., 2012a; Chapman *et al*., 2021). However, it is crucial that the healthcare professionals have the right knowledge and skills to inform people with MND about all aspects of the decision, and where this is lacking there is the need to utilise the wider MDT to ensure access to this information.

Home v Clinic setting

The setting in which discussions about gastrostomy took place was largely dictated by the organisation of the MND service including where the healthcare professionals taking responsibility for supporting the decision-making of people with MND were employed. Healthcare professionals and people with MND identified both benefits and burdens of having such discussions in the home or clinic. There was general support for the home being the most appropriate setting to have the lengthy, complex and sensitive discussions about gastrostomy. MND MDT clinics were valued by people with MND as they allowed efficient access to specialist MND care from a range of professional disciplines, including the doctor responsible for their care, with most medical professionals only being accessible in clinic (Hogden *et al.,* 2012b; Schellenberg and Hansen, 2018; Kierkegaard *et al.,* 2021). However, the study findings highlighted challenges in relation to having quality discussions about gastrostomy placement during busy clinic visits. Consistent with previous research, clinic visits were emotionally and physically fatiguing for people with MND due to travel times, their length, attending multiple appointments and having to process information about the diagnosis, disease progression and future care options (Paganoni *et al.,* 2017; Schellenberg and Hansen 2018; Paynter *et al.,* 2020; Genuis *et al.,* 2023). Therefore, people with MND may lack the emotional, mental or physical energy required to engage in meaningful discussions about their options. Some MND clinics also lacked representation from all the disciplines who may play a role in informing the decisions of people with MND e.g. dietitian and SLT within Case 2.

The home setting was broadly supported by people with MND, caregivers and healthcare professionals as the optimal environment to have discussions about gastrostomy. Benefits of the home setting included people with MND being in a comfortable environment with their family, avoidance of the fatigue associated with the time spent travelling or sitting in clinic and allowing a focus on the gastrostomy decision. This is consistent previous studies reporting that the home setting promoted intimacy and supported having sensitive discussions (Vierhout *et al.,* 2019; Bublitz *et al.,* 2024). Where healthcare professionals had the flexibility within their role, there was evidence of a strategic approach being used by restricting the clinic to present the option (i.e. 'sow the seed') and purposively planning the detailed discussion to take place in the home of the person with MND. Such a staggered approach was appreciated by people with MND as it allowed time to process the information and for any initial emotional response to settle (White *et al*., 2023). Consistent with previous research, healthcare professionals acknowledged the benefits of working across settings including improved consistency, responsiveness, and closer monitoring of disease progression, with frustration expressed by some when MND Centres lacked an out-reach service (Hughes *et al.,* 2005). Therefore, the development of MDT decision support strategies should seek to facilitate discussions to take place in settings that will enable people with MND to make the best quality decisions.

Inter-professional communication is key to coordinated MDT decision support

Healthcare professionals supporting were often separated by geography, team and organisation. Therefore, the accessibility and quality of communication between healthcare professionals was key to coordinating the decision support delivered across the MDT (Paganoni *et al.,* 2017). Communication challenges included a lack of easy access to sources of information about the indication for discussing or deciding about gastrostomy and with regards progress already made with decision-making. Similar to previous research, even where documentation was available, the level of detail was often brief, lacked valuable contextual information and could be left open to interpretation (or misinterpretation) (Hogden *et al*., 2012a). The observation that healthcare professionals often lacked awareness of the current indication for gastrostomy or progress people with MND had already making a decision, may be explained by the communication barriers described above. The findings of this study are consistent with a report which identified how a lack of shared records and patient updates was a barrier to the coordination of MND care (OPM, 2016). The current study also provides a contextual understanding of the concerns about the quality of MDT communication expressed by healthcare professionals responding to a recent survey (White *et al.,* 2024).

Coordination requires continuity of care

Continuity of care requires ‘informational continuity’ allowing healthcare professionals understanding of the patient’s condition and their preferences and values (Haggerty, 2003). In addition to understanding the indication for, and people with MND views about gastrostomy, improved connectivity between healthcare professionals can lead to shared norms and therefore consistency in relation to how care is delivered (Alexander *et al.,* 2005). Professional guidance recommends healthcare professionals share with the MDT any decisions made with people with MND (NICE, 2019). With decision-making taking place over time and people, there is a need to ensure the MDT are up-to-date not just with what decisions have been made, but also the progress made and any challenges experienced with decision-making. People with MND were generally satisfied with their care and the consistency of information shared with them. However, there was evidence of how healthcare professionals lacking awareness of the current indication for gastrostomy or about previous discussions, could disrupt the decision-making process (Foley *et al.,* 2012, Hogden and Crook, 2017). Examples included when healthcare professionals’ preferences or practice differed in relation to when options are introduced, how the information is framed and the current need for a decision. Previous research in Australia, where care is distributed over much larger distances than in the UK, reported how poor interdisciplinary communication can impact on coordination of decision making across the MDT: ‘one hand doesn’t know what the other hand’s doing’ (Hogden *et al.,* 2012a; McConigley *et al.,* 2014). The current study demonstrated that communication barriers are not only due to geography but also because of organisational, strategic, and practice factors and these can contribute to a lack of continuity of care.

Organisation of MDT decision support

The findings of this study highlight the need for a cross-setting strategic approach to how people with MND are supported to make decisions about gastrostomy by any healthcare professional within a regional MND Care Model. Similar to previous publications, some cases had developed care pathways aim to coordinate the placement of gastrostomy (Oliver *et al.,* 2007; Marsden *et al.,* 2016). A coordinated MDT approach to decision support requires services to collaborate and come to a consensus about when discussions are initiated, the information shared with people with MND, roles and responsibilities and MDT communication. Coordinated MDT decision support has the potential to not only improve the quality of the decision-making process, but also if it leads to more timely decisions, to better health and quality of life outcomes.

Strengths and Limitations

Strengths

A strength of this research is the excellent access gained on each case. The selection of cases with principal investigators who had good contact with the regional MDT enabled a range of settings to be accessed and contact with participants meeting the inclusion criteria. This enabled the purposive sampling strategy to be effectively employed, supporting the collection of data to answer the research question from a breadth of perspectives. The range of qualitative methods allowed the phenomenon to be described from a range of emic and etic positions, adding to the rich and thick description of the findings. The sequential and iterative approach to data collection allowed for the emerging concepts from observations to be explored further in the interviews and then finally the focus groups.

Limitations

There are limitations to be considered when interpreting the findings of this study. First, it was not possible to gain access to every setting or organisation in which discussions about gastrostomy took place. For example, recruitment could not take place in hospices or community NHS organisations where the researcher did not have research governance approvals. This is a learning point for future case study or ethnographic research that seeks to understand MND teamwork across organisational boundaries to ensure access is gained to all possible settings within the case boundaries. Second, ethical permission was not given for observing diagnostic consultations, despite this being an interaction where gastrostomy may be first introduced to people with MND. This meant the researcher was unable to observe how gastrostomy was addressed during this challenging scenario. Third, this study focused on the coordination of the HCPs delivering decision support rather than the quality of the decision-making process. Further research is required to understand how the MDT can support people with MND to make quality decisions about gastrostomy placement.

Implications for practice

The findings of this study have implications for how MND services coordinate the decision-support about gastrostomy is delivered across the different settings in which discussions take place. First, healthcare professionals who support people with MND with their decision-making should collaborate to understand and identify the roles and responsibilities of the different healthcare professionals involved. Responsibilities may include identifying the need to discuss the option, initiating the discussions, informing about interventions and guiding the timing of gastrostomy placement. Second, improved systems of communication are required that are used and accessible to all healthcare professionals involved in supporting people with MND to make decisions about gastrostomy. Additionally, developing quality standards regarding what information should be shared between the MDT could also contribute to coordination of care. Third, consistency of decision support could be improved through professional consensus about how people with MND are supported to make decisions and the development of decision aids that integrate the inter-professional nature of decision-making in MND care (Legare *et al*., 2011; Lognon *et al*., 2022). Ultimately, the aim of a strategic approach should be for the MDT to function as a coordinated unit of decision support for people with MND making this complex decision (Hogden and Crook, 2017). While this case study is focused on the models of MND care delivery in the UK, it is likely the concepts presented are transferable to other countries with international MND guidance also recommending MND MDT care as the gold standard (Miller *et al*., 2009a; Shoesmith *et al*., 2020).

Reflexivity

The lead researcher (SW) had previous experience of supporting people with MND to make decisions about gastrostomy in his role as an NHS dietitian and some personal experience of the disease. The influence that this experience had on the lead researchers' decisions and thinking were discussed in frequent meetings with the wider supervisory team (VH, AO, CM), to ensure the findings and interpretations were grounded in the qualitative data being collected.

Conclusion

Delivering coordinated decision support to people with MND about gastrostomy across time, space and people is complicated and requires a strategic approach. There are significant communication challenges that impact on healthcare professionals ability to remain aware of the current need to discuss or place a gastrostomy, and about people with MND progress with decision-making. Inter-professional communication solutions are key to supporting a coordinated approach across the MDT, in any setting in which discussions about gastrostomy take place. Future research, service improvement and intervention development should focus on how to support HCPs to be better connected and informed.

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7.6 Summary of chapter

The chapter above has presented the findings from the multiple case study that have contributed to answering the research question:

> ‘How do healthcare professionals, operating within MDTs, support people with MND to make decisions about gastrostomy placement?’.

Two different MND care models were employed on the cases. In summary, within Cases 1 and 2 most of the healthcare professionals were restricted to either the clinic or community setting with responsibility for decision support being widely distributed across the MDT. In contrast, within Case 3, most of the healthcare professionals covered both the clinic and community setting, with a single healthcare professional taking on the majority of responsibility for supporting decision-making about gastrostomy. This difference in care model allowed for conceptual comparisons to be made between cases in relation to how MDT decision support was coordinated across the MDT. The findings are summarised below:

- The home of people with MND was identified as the ideal setting in which to have the complex discussions about gastrostomy.
- There was a lack of strategy across all cases in relation to how the MDT would plan and deliver coordinated decision support.
- The detail and accessibility of professional communications influenced the MDTs’ ability to coordinate decision support.
- When more healthcare professionals are involved who are operating in different settings or organisations, increased lines of communication are required which contributed to a lack of coordination of decision-support across the MDT.
- Healthcare professionals lacking awareness of the current indication for gastrostomy, or the previous progress people with MND had made with decision-

making, resulted in repetition and in some cases contradiction of information shared.

- The quality of coordinated MDT decision support is reliant on having healthcare professionals with the right knowledge and skills in the settings in which these discussions take place.

8 Discussion

8.1 Chapter outline

The following chapter identifies the key findings from the three components of the study and how they have contributed to answering the research questions. Following this, the study findings are integrated and compared to the published literature and theory. The strengths and limitations of the research is discussed, followed by suggested implications for practice and future research. Finally, the conclusions are presented.

8.2 Summary of research completed

8.2.1 Research questions and aims

The overarching doctoral research question this mixed-methods study has answered was:

> 'How do MND multidisciplinary teams support people with MND to make decisions about gastrostomy tube placement?'

The research questions for each component of the study (which contributed to answering the overarching doctoral research question) were:

- Qualitative evidence synthesis:
  > 'What are the organisational, team and individual level factors that influence decision making by people with MND when making the decision to start an invasive healthcare intervention?'
- HCP survey:
  > 'What are healthcare professionals practice and beliefs in relation to supporting people with MND making decisions about gastrostomy placement?'
- Multiple case study:
  > 'How do healthcare professionals, operating within MDTs, support people with MND to make decisions about gastrostomy placement?'

The key findings from the study are described below including how each component of the study contributed to the finding and the answering of the research questions.

8.2.2 An emotional response influences engagement of people with MND in discussions and the timing of decisions made

The qualitative evidence synthesis identified how people with MND decisions about interventions extend beyond the clinical indications. Discussion about interventions trigger an emotional response in relation what the options signify in terms of disease progression, declining independence and the impact on their sense of normality. In addition, people with MND undertake an existential debate about the value and consequences of commencing a life-prolonging intervention. The survey identified how a third of healthcare professionals were unable to initiate discussions with people with MND as early as they would prefer. Also, half of healthcare professionals believed gastrostomy tubes were placed too late though there was a lack of consensus about the optimal timing of gastrostomy. The multiple case study was an opportunity to explore why discussions and decisions about gastrostomy were happening later than healthcare professionals believed was ideal. The case studies confirmed that people with MND often experience an emotional response to being presented information about the disease and the gastrostomy option which drove avoidant strategies designed to enable people with MND to cope with living with MND. Healthcare professionals may also make subjective judgements about whether people with MND are ‘ready’ to discuss the gastrostomy option. Strategies that identified and addressed the emotions, goals and values of people with MND and promote control may be key to timely engagement and decisions about gastrostomy.

8.2.3 A misalignment of the preferences, perceptions and priorities of people with MND and healthcare professionals

The qualitative evidence synthesis identified that healthcare professionals play an influential role in what and when decisions are made, through the provision of information, support and guidance. The survey was an opportunity to understand what informed healthcare professionals reasoning in relation to how they support people with MND to make decisions about gastrostomy. While a broad range of intervention information is shared with people with MND, there was a lack of consensus in relation to the timing of discussions and decisions, and in relation to how healthcare professionals share their preferences for certain options. The multiple case study found that healthcare professionals and people with MND enter the collaborative process of decision-making with different preferences and priorities in relation to considering the gastrostomy option. Healthcare professionals are in possession of a wealth of tacit knowledge about MND, through their experience of caring for people with MND and knowledge about the published research and guidance. This informs their preference to engage in early discussions about gastrostomy and to encourage people with MND to make decisions prior to experiencing significant disease progression. Healthcare professionals' motivation to reduce disease and procedure related risks or crisis takes priority over discussion about the risks and benefits of enteral nutrition. People with MND, part driven by an emotional response, can prefer to delay discussions and decisions, through wanting to extend a sense of normality and avoid acceptance of disease progression. Healthcare professionals have a complicated ethical task of supporting people with MND to make informed decisions about whether or not to engage in discussions, and about the timing of gastrostomy.

8.2.4 Concrete healthcare professional guidance assists people with MND to implement their decisions

The qualitative evidence synthesis identified how not just symptom severity but also healthcare professional guidance drives the decisions people with MND make about interventions. People with MND experienced uncertainty about their options with some expecting healthcare professionals to use their MND expertise to guide their decision-making. However, Healthcare professionals delivering guidance was complicated by simultaneously seeking to respect people with MND right to autonomy and their preference to remain in control including avoiding placing people with MND under pressure to make decisions. The survey found that healthcare professionals were divided in relation to whether they had a responsibility to give recommendations about whether to accept, delay or decline gastrostomy; with these beliefs being mirrored in reported equally mixed practice. Interestingly, doctors and nurses were more likely to give people with MND recommendations than dietitians or speech and language therapists. The lack of consensus about what level of disease progression indicated a need to discuss or make decisions about gastrostomy had potential to impact on how healthcare professionals might frame these choices to people with MND in practice. These findings were reflected in the multiple case study with some people with MND feeling uninformed about the optimal timing of gastrostomy placement, with HCPs communicating a generic preference for 'earlier rather than later'. The use of abstract descriptions given by healthcare professionals did not always give people with MND the concrete information they required or wanted to be able to make decisions about the timing of gastrostomy. There was evidence that a more direct approach, including explicit sharing of healthcare professional preferences, was sometimes needed to convince people with MND to implement decisions.

8.2.5 Challenges associated with coordinating decision support that is distributed over time, space and people

The qualitative evidence synthesis including reports of miscommunication and contradiction between the information shared by different healthcare professionals involved in supporting the decision-making of people with MND. Despite healthcare professionals being a valued source of information, a lack of consistency had the potential to cause decisional conflict. The survey identified a lack of consensus about how healthcare professionals support the decision-making of people with MND about gastrostomy including differences between the beliefs and practice of different professional disciplines. The survey further identified differences between the beliefs and practice of the various disciplines involved in supporting decision-making. Healthcare professionals responding to the survey lacked some satisfaction with the consistency and communication of information within their MDT. These findings highlighted that there were challenges associated with delivering consistent and coordinated decision support across the MDT which could be explored in more depth in the multiple case study. Selecting cases with differing MND care models allowed for some interesting analytical comparisons to be made about how decision support is coordinated across the MDT. As confirmed in the earlier research, many different healthcare professionals are involved, operating across both the clinic and community settings. All cases were lacking a clear and agreed strategy about how and when discussions are initiated and in relation to guiding the decisions of people with MND about if and when to have a gastrostomy. The quality and accessibility of inter-professional communication was key to healthcare professionals remaining aware of progress made with decision-making to allow them to continue to inform the decisions of people with MND rather than disrupt them.

8.3 Comparing the findings of the research to current literature

8.3.1 Identifying and addressing the emotional response: key to engagement in timely decisions

The emotional response of people with MND to information about the diagnosis, disease progression and gastrostomy option

The qualitative evidence synthesis identified that the decisions people with MND make about interventions were driven by emotional responses to the option being presented that extended beyond concern for any functional changes or future health threats. This was a finding that was explored in more depth in the multiple case study and provided some possible explanations for healthcare professionals reports in the survey of delayed discussions and decisions about gastrostomy. Despite professional guidance recommending people with MND are engaged in discussions about gastrostomy early in the disease course (NICE, 2019), it is important to question whether such discussions are actually 'better' for people with MND. This should include understanding how people with MND perceive and experience being asked to consider a time in the future where they may need to consider gastrostomy placement and what this may represent to them. When seeking to address this question, a further consideration is how is 'better' defined? When only defined clinically, there are rationales for earlier discussions including how they may promote earlier intervention and therefore support achieving improved outcomes, as discussed further below in section 8.3.2. There may also be an ethical argument for earlier discussions supporting the autonomy and control of people with MND. The ethical argument though is a little more nuanced and complex when including the varied preferences and responses people with MND have to discussing gastrostomy particularly prior to any functional signs that such an invasive and life-changing intervention may be needed.

The findings from the case study observations and interviews are consistent with those of the qualitative evidence synthesis which describe an emotional response which people with MND experience when informed about the diagnosis, disease progression or the gastrostomy option (Martin *et al.,* 2016; Aoun *et al*., 2018; White *et al*., 2023). Indeed, one complicating factor associated with informing about their options is being unable separate discussion about the decision from the prospect of disease progression. The two topics are intricately linked with the latter having to be understood for people with MND to appreciate why the option is being presented to them. Discussing them near to each other or together adds to the emotional response experienced. The emotional response likely explains the survey finding that healthcare professionals' preference for earlier discussions does not always translate into their practice (White *et al*., 2024). A number of people with MND and caregivers had concerns about discussing gastrostomy too early and the impact this may have on their ability to psychologically cope with life with the disease. It is clear from the current research that the emotional responses of people with MND, including their stage of acceptance and the coping strategies they employ play an important role in when they choose to engage in discussions about gastrostomy (King *et al*., 2009). Acknowledgment of this concept is missing from not only guidance but also from current models of SDM in healthcare and in MND. Therefore, there is an opportunity for the development and validation of theory to guide future research, interventions and care provision that account for how humans engage in challenging discussions when emotionally responding to the information about the diagnosis and disease progression.

Addressing the emotional response as part of a decision support strategy

Current SDM models do not always explicitly account for how patients' emotional response may influence their involvement in decision-making (Elwyn *et al.,* 2017). Others have but not elevated the relevance and importance in the schematics developed to represent the model of SDM (Legare *et al.,* 2011b). A recently developed SDM model for end-of-life decisions in dementia, explicitly includes the need to address emotions (Davies *et al.,* 2021). Any model of decision-making validated for MND should include and expand on how the emotional response could be identified and mediated to enable engagement in the subsequent criteria for successful SDM practice. If accepting the argument that all people with MND should at least be given the opportunity to understand their future options prior there being a significant clinical need, then how can this be promoted? The findings from the qualitative evidence synthesis and the multiple case study highlight addressing the emotional response as a key mechanism through which increased engagement could be enabled. The following section discusses some of the potential strategies open to MND teams.

Advanced communication skills and strategies

Healthcare professionals have the complex dual responsibility to share clinical information while simultaneously managing the emotional responses of people with MND that such discussions can trigger (Edwards *et al.,* 2021b). This involves having not just good clinical knowledge, but also advanced communication skills required to sensitively deliver bad news about the disease. Indeed, healthcare professionals and people with MND within all cases identified how it is not just what is said, but how it is said that is important. Doctors have previously cited a lack of training as a barrier to the effective delivery of bad news to people with MND; and informing someone they may need to consider having a gastrostomy placed is considered 'bad news' for the majority of people with MND (Schellenberg *et al.,* 2014). In line with previous research exploring the preferences of people with MND for professional communication, including a robust rationale for having discussions about gastrostomy promoted engagement (Genuis *et al.,* 2023). As identified by one person with MND in Case 2, having gastrostomy discussed triggered similar feelings to that of being given the diagnosis. Therefore, the principles associated with delivering the diagnosis which the majority of the literature is focused on (Bongioanni *et al.,* 2023), could equally be applied to attempts by healthcare professionals to initiate discussions about gastrostomy (or other interventions commonly discussed in MND care). Individual preferences for information and how it should be delivered, differs between people with MND, with each experiencing MND and interactions with healthcare professionals differently (Genuis *et al.,* 2024). HCPs have been reported to not always take into account what people with MND already understand or what their current information needs are (Bongioanni *et al.,* 2023). This is consistent with the case studies finding that healthcare professionals frequently lacked awareness of previous assessments or discussions.

Therefore, healthcare professionals seeking to open discussions about gastrostomy should use similar approaches and acquire the same communication skills required to deliver the diagnosis. These include taking consideration of the setting, use of empathy and active listening, exploring what is already understood, using simple language and avoiding using abstract descriptions (Miller *et al.,* 2009b; Bongioanni *et al.,* 2023). When compared to these attributes of effective communication skills there was evidence of both effective and possibly less effective qualities during observations of healthcare professionals discussions with people with MND within all cases. There has been interest in learning from and adapting the approaches taken in oncology, including the use of communication frameworks or protocols to guide how healthcare professionals deliver bad news (Edwards *et al.,* 2021b). For example, the SPIKES (setting, perception, invitation, knowledge, emotion and strategy) protocol developed in oncology, details how healthcare professionals can share unwelcome information and engage patients in discussions about their treatment plans (Baile *et al.,* 2000). A third of people with MND when surveyed reported dissatisfaction with how the diagnosis was communicated with them in relation to the steps outlined in SPIKES (Aoun *et al.,* 2016). The SPIKES protocol has been proposed as being applicable to MND care (Miller *et al.,* 2009b) and subsequently adapted further for the MND context (Edwards *et al.,* 2021b). Edwards *et al.,* (2021b) developed the ALS ALLOW protocol (figure 11), which is aimed not just at delivering the diagnosis but also when establishing the goals of people with MND, which could include making decisions about gastrostomy.

Figure 11. Proposed ALS-Specific Guidelines for Breaking Bad News: ALS ALLOW. Edwards *et al.,* 2021b Delivering Bad News in Amyotrophic Lateral Sclerosis: Proposal of Specific Technique ALS ALLOW, Table 2, pp. 524. Table reused with permission from Wolters Kluwer Health, Inc.

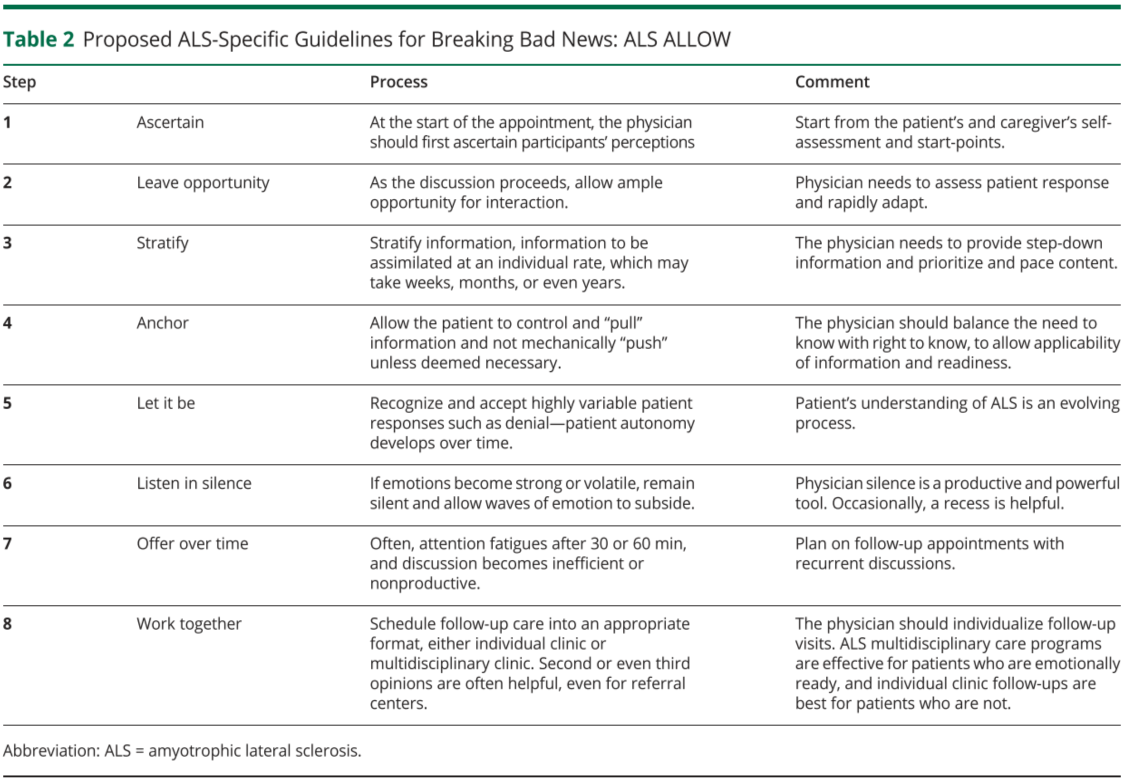

Table 2 Proposed ALS-Specific Guidelines for Breaking Bad News: ALS ALLOW

Step		Process	Comment
1	Ascertain	At the start of the appointment, the physician should first ascertain participants' perceptions	Start from the patient's and caregiver's self-assessment and start-points.
2	Leave opportunity	As the discussion proceeds, allow ample opportunity for interaction.	Physician needs to assess patient response and rapidly adapt.
3	Stratify	Stratify information, information to be assimilated at an individual rate, which may take weeks, months, or even years.	The physician needs to provide step-down information and prioritize and pace content.
4	Anchor	Allow the patient to control and "pull" information and not mechanically "push" unless deemed necessary.	The physician should balance the need to know with right to know, to allow applicability of information and readiness.
5	Let it be	Recognize and accept highly variable patient responses such as denial—patient autonomy develops over time.	Patient's understanding of ALS is an evolving process.
6	Listen in silence	If emotions become strong or volatile, remain silent and allow waves of emotion to subside.	Physician silence is a productive and powerful tool. Occasionally, a recess is helpful.
7	Offer over time	Often, attention fatigues after 30 or 60 min, and discussion becomes inefficient or nonproductive.	Plan on follow-up appointments with recurrent discussions.
8	Work together	Schedule follow-up care into an appropriate format, either individual clinic or multidisciplinary clinic. Second or even third opinions are often helpful, even for referral centers.	The physician should individualize follow-up visits. ALS multidisciplinary care programs are effective for patients who are emotionally ready, and individual clinic follow-ups are best for patients who are not.

Abbreviation: ALS = amyotrophic lateral sclerosis.

The findings from the case studies are consistent with the steps described in ALS ALLOW. For example, the need to understand the perceptions of people with MND before delivering information, the use of advanced communication skills, tailoring the approach to individual needs and addressing the emotional response. Additionally, while at diagnosis there may be the opportunity not to pursue discussions if the person with MND wishes to avoid them i.e. Step 5 of ALS ALLOW: ‘let it be’ (figure 11), the protocol acknowledges this may not be appropriate later in the disease (e.g. when discussing gastrostomy) without risking a negative impact on outcomes. It also promotes the benefits of using the home setting for these discussions later in the disease consistent with the preferences of people with MND, caregivers and healthcare professionals in the case studies.

Most people with MND were very satisfied with the communication of their healthcare professionals, reporting not just good therapeutic relationships but also a personal connection. However, there were examples of practice that were not consistent with the principles of breaking bad news summarised. These included a lack of awareness of the understanding of people with MND and a reliance on abstract analogies to describe the decision problem. Further research is required to understand how healthcare professionals could more effectively open timely discussions, particularly in the absence of symptoms indicating a need for gastrostomy or in the presence of emotionally driven resistance. It is important that healthcare professionals tailor their approach to the individual, including the use of advanced communication skills. Any development of interventions to enable engagement in early discussions should integrate the principles of psychological interventions that support the management of any emotional response.

Targeted mediation of the emotional response

An important principle of the breaking bad news protocols is the psychological support people with MND need to help manage the emotional response the discussions trigger; a consideration that people with MND have found lacking in their interactions with healthcare professionals (Maksymowicz *et al.,* 2022). A finding from the multiple case study was that emotional responses were common, even if not always overtly expressed by people with MND. Identifying and addressing the emotional response to the diagnosis and information about the disease may not only support people with MND adjustment but also facilitate engagement in decision-making discussions. A wide range of psychological and psychosocial interventions have been identified in recent scoping reviews, including education, counselling, psychotherapy, behavioural therapy and social support interventions (Zarotti *et al.,* 2021; Oh *et al.,* 2024). Though the included studies did not use decision quality outcome measures they did infer psychological improvements which could empower people with MND to engage in discussions about gastrostomy. Acceptance and commitment therapy (ACT) is a psychological therapy that promotes people to engage in action aligned with their values, even when experiencing significant emotional distress (Hayes *et al.,* 2006). The recent large multicentre RCT in the UK, COMMEND, found that delivering ACT in MND care significantly improved the quality of life of people with MND (Gould *et al.,* 2024). The same study also found improvements in the psychological flexibility of people with MND after 9 months, which can support them to be aware of and accept their emotions, including any that are negative. Improved psychological flexibility has been found to be associated with improved understanding and acceptance of the gastrostomy option (Pearlman and Thorsteinsson 2019). With discussions about gastrostomy being influenced by the emotional responses of people with MND, psychological interventions could play an important preliminary role in preparing and empowering them to engage in timely discussions through addressing the emotional response.

8.3.2 People with MND and healthcare professionals need to be on the same page: sharing of perceptions, preferences and priorities

Healthcare professionals proactive preferences

A core principle of shared decision-making (SDM) is that healthcare professionals and patients share information, preferences and attain an understanding of each other's reasoning (Makoul and Clayman, 2006; Breckenridge *et al*., 2015). However, the study findings highlighted that the preferences, perceptions and priorities of healthcare professionals and people with MND, for engaging in discussions or making decisions about gastrostomy were not always aligned. Healthcare professionals and people with MND sharing the rationales for their preferences for or against intervention, may support a better understanding of the decision problem and allow the values of people with MND to be integrated into their informed decisions (Breckenridge *et al*., 2015). The initiation of discussions about gastrostomy was identified as a crucial phase of the decision-making process though both healthcare professionals and people with MND found it challenging. Despite the overwhelming support from HCPs healthcare professionals for discussing gastrostomy early in the disease course reported throughout the study, this entry into the deliberative process of understanding options and decision-making was often delayed by either healthcare professionals or people with MND.

Healthcare professionals’ reasoning and preferences for earlier discussions was informed by their tacit knowledge and the published research and guidance. Healthcare professionals frequently referred to the recommendations in MND guidance (NICE, 2019) and findings from studies associating weight loss and delayed gastrostomy placement with poorer outcomes, particularly the large prospective observational study, ProGAS (ProGAS study, 2015). In the UK, MND care is guided by three sets of clinical guidelines, each of which address the decision to place a gastrostomy (Burgos *et al.,* 2018; NICE, 2019; Van Damme *et al.,* 2024). Each set of guidance uses very similar short, generic, statements about when discussions about gastrostomy should be initiated with people with MND which do not extend beyond that the option should be discussed at an early stage. Other than linking to the subsequent earlier consideration of gastrostomy placement, these recommendations fail to justify why discussions should be commenced, do not provide a definition of ‘early’ or acknowledge the contextual complexities associated with opening conversations about these difficult topics with people with MND. A review of guidelines from other countries similarly does not include any recognition of the contextual complexity associated with engaging people with MND in early meaningful discussions about gastrostomy, including any guidance or justifications about timing of discussions or about how to address emotional responses the current study found to be commonplace (Miller *et al.,* 2009a; Shoesmith *et al.,* 2020; Petri *et al.,* 2023).

The published guidance portrays the gastrostomy choice mostly in clinical terms; decisions based on addressing clinical risks presented by deteriorating markers of clinical function. The prioritisation of evidence from quality clinical research e.g. RCT’s, in MND guidelines, does not reflect the reality of how humans respond or engage in such discussions (Lewin and Glenton, 2018; Flemming *et al.,* 2019). Without acknowledging the contextual, real-world experience of having such discussions healthcare professionals will continue to meet barriers to adhere to the recommendation which may have subsequent consequences for the timing of enteral nutrition and patient outcomes. There is a strong argument for clinical guidelines to integrate qualitative evidence (including that performed in real-world practice) to inform or at least provide contextual nuance to recommendations, (Tan *et al.,* 2009; Lewin and Glenton 2018; Flemming *et al.,* 2019). Practice informed by more contextually informed professional recommendations may be the key to healthcare professionals meeting other clinical goals e.g. the earlier placement of a gastrostomy. The research presented in this thesis has generated new contextually rich knowledge about engaging people with MND in discussions about their options and could be used to inform future updates of professional guidelines.

The clinical and ethical rationales for earlier discussions

While MND guidance encourages healthcare professionals to discuss gastrostomy early in the disease course, it is important to be able to provide a robust rationale for having such discussions which people with MND often can instinctively prefer to avoid (White *et al.,* 2023). Consistent with the professional recommendations and previous research, most healthcare professionals (81%) responding to the survey believed that discussions with people with MND should be initiated early i.e. prior to, or following identification of, the presentation of indications for gastrostomy (Van Eenennaam *et al*., 2021). Similarly, there was broad agreement of healthcare professionals within all cases in the case study for seeking to open early conversations about the option. Clinical and ethical arguments for opening early discussions were captured in the qualitative evidence review and case studies.

The ethical argument for earlier discussions broadly relates to supporting the autonomy and control of people with MND. First, relates to opening all possible options and therefore also the level of choice available to people with MND. People with MND lacking a perception of choice about gastrostomy has been a consistent theme reported in a number of qualitative studies (Greenaway *et al.,* 2015; Paynter *et al.,* 2020; Lisiecka *et al.,* 2021a; Van Eenennaam *et al.,* 2024). The concept of lacking choice often reflects a perception that continuing without gastrostomy is not a valid option; associating the option with starvation or death (Greenaway *et al*., 2015). Despite the uncertainty about predicting the rate of disease progression, what is certain is that once dysphagia presents it will continue to progress and present a significant risk of malnutrition or aspiration, particularly for those diagnosed with bulbar onset or fast progressing disease (Mariani *et al.,* 2022). Some people with MND, if given the choice, may wish to be proactive and decide to have a gastrostomy prior to the development of malnutrition, dysphagia or respiratory failure rather respond to the presentation of these indications for the intervention. For people with MND to have the widest scope of choice possible, healthcare professionals would be required to actively introduce and discuss the options prior to the presentation of symptoms as it is unlikely people with MND would have the motivation or foresight to trigger the discussion themselves. This rationale is consistent with the ethical principles of autonomy (Beauchamp and Childress, 2013), Mental Capacity Act (MCA, 2015), professional guidance on informed consent (BMA, 2024) and the Montgomery Supreme Court ruling (Chan *et al.,* 2017) that healthcare professionals have a responsibility to offer all clinically indicated options with patients.

A second ethical argument for why early discussions support people with MND to make autonomous decisions relates to the ability to engage in meaningful discussions or even have the mental capacity to make their own decision. A number of healthcare professionals identified the importance of giving people with MND the opportunity to understand and discuss their options while they can still speak well or are cognitively able to make their own decisions. Problems speaking (i.e. dysarthria) can affect 80-95% of people with MND with some losing the ability to speak altogether (i.e. anarthria) (Beukelman *et al.,* 2011). Consistent with reports in a previous qualitative study, several consultations were observed where people with MND were less able to be involved in discussions as a result of dysarthria and the reliance on communication aids (Munan *et al*., 2021). When people with MND are unable to communicate their wishes by any means, even with using communication aids, they could be assessed as lacking the mental capacity to make their own decisions (MCA 2015). Up to half of people with MND experience cognitive changes, including 15% developing frontotemporal dementia, which has the potential to impact on how they perceive and process decisions and at worse could result in lacking mental capacity (Ringholz *et al.,* 2005; Radakovic *et al.,* 2024). The healthcare professionals within Case 2 described finding scenarios where people with MND were experiencing cognitive changes particularly challenging in relation to making best interest decisions about gastrostomy. Therefore, giving people with MND the opportunity to consider their options while they still have the ability to engage in discussions, to cognitively deliberate about the information shared and to express their preferences, is a strong ethical argument for initiating discussions early.

The clinical arguments for earlier discussions relate to expectations of disease progression and the safe placement of gastrostomy and timely commencement of enteral nutrition. As discussed in section 2.4 (Chapter 2), malnutrition and aspiration are associated with worse outcomes for people with MND (Sorenson *et al.,* 2007; Ning *et al.,* 2019). The lack of empirical evidence for the effectiveness or timing of enteral nutrition should be acknowledged (Sulistyo *et al.,* 2023), with the recommendations for earlier placement being informed by cohort or observational studies (ProGas Study, 2015) and professional consensus (Burgos *et al.,* 2018; NICE 2019; Van Damme *et al.,* 2024). A reasonable case could be made for generalising research findings from other clinical contexts (Norman *et al.,* 2008), that weight loss and dysphagia are associated with worse outcomes for people with MND and concluding that gastrostomy placement is an obvious solution to address these health threats. However, the argument for what represents the effectiveness of enteral nutrition is more complicated than referring only to the nutritional, clinical or survival outcomes. First, there are many different indications (e.g. malnutrition, aspiration risk, respiratory failure and mealtime burden) for considering enteral nutrition in MND, and for this reason multiple outcomes that may be relevant to evaluate the interventions' effectiveness against (e.g. weight changes, chest infection incidence, procedure associated morbidity and mealtime experience). People with MND will each value these outcomes differently highlighting the need for a personalised approach that integrates individuals' values and preferences into the decision-making process. Second, the outcomes associated with enteral nutrition in MND are associated with the speed of disease progression and the timing of intervention (ProGAS study, 2015). There are likely diminishing benefits of enteral nutrition and increasing procedural risks associated with gastrostomy placement, particularly when nearing the final stages of the disease. Some of the poor outcomes associated with malnutrition are less likely to be relevant to people with MND when compared to people living with conditions with a longer disease course (NICE, 2017). This does not mean there are no positive nutritional benefits associated with the decision, but that these disease-related nuances need to be factored into decision-making.

There is an argument that discussions about gastrostomy should be triggered by presentation of the risks of malnutrition including early signals of dysphagia, changes in appetite or rate of disease progression, rather than waiting for clinical indicators of malnutrition or aspiration to present (Johnson *et al.,* 2012; Mariani *et al.,* 2022; Shijo *et al.,* 2023; Gebrehiwet *et al.,* 2023; Mendes *et al.,* 2024). For example, Mariani *et al.,* (2022) retrospective cohort study found that people with MND with fast progressing and bulbar onset disease reached a level of dysphagia defined as indicating need for gastrostomy faster than those with slower progressing or spinal onset disease. Information about rate of disease progression, which is available at diagnosis and throughout the disease course, could also be used to guide the timing of discussions and decisions about gastrostomy. In relation to taking a more strategic approach to MDT decision making, MND teams could dedicate increased decision support resources for those people with MND expected to reach the milestone of being indicated for enteral nutrition sooner than others with slower progressing disease.

There is a subtle but important distinction between there being a decisional and procedural cut-off; a concept highlighted by a Case 1 healthcare professional. This distinction is not always captured in the professional guidance or commentaries on the topic. Healthcare professionals' tacit knowledge about the risks of people with MND reaching crisis and the time it takes to organise gastrostomy placement often motivated a preference to discuss gastrostomy early. Previous research has demonstrated that nutritional status can deteriorate further between gastrostomy being recommended and the placement of gastrostomy (Son *et al.,* 2024). Healthcare professionals report having a responsibility to remain one-step ahead of the disease (McConigley *et al.,* 2014); a remit that some people with MND and caregivers agreed with. Part of this remit involved healthcare professionals using their tacit knowledge to judge when to initiate discussions, working backwards from predictions about when the optimal time to place a gastrostomy would be. These judgements involved taking into consideration the time it can take people with MND to make decisions, the predicted rate of disease progression and the time it takes local services to organise the placement of a gastrostomy.

Therefore, there are several ethical and clinical justifications which inform healthcare professionals preferences for initiating early discussions about gastrostomy. These should be made more explicit in the professional guidance and be included in healthcare professionals' rationales when trying to open these sensitive conversations. While these justifications exist, it remains important to consider how people with MND perceive and respond to having discussions about future disease progression and treatment options, particularly before they are experiencing any indications for intervention.

Informing people with MND *about their choice to engage in discussions about gastrostomy*

A number of healthcare professionals in the multiple case study identified that people with MND may avoid discussions about gastrostomy without being fully informed about the consequences of delaying gaining an understanding of how they may wish to manage current or future risks of dysphagia. Therefore, there is a requirement for healthcare professionals to find ways to share their rationales and reasoning with people with MND so that they can make an informed choice about whether to engage in discussions about gastrostomy or not, while being sensitive to their emotional responses. This could also promote the preferences and priorities of people with MND, being better aligned. A SDM approach (Makoul *et al.,* 2006) could be taken to supporting people with MND to make decisions about their option to engage (or not) in discussions about their options. Any approach would need to focus on promoting cognitive, problem-focused coping strategies and mediate any emotional, avoidant coping strategies. For example, following a more considered approach could support people with MND to make an informed decision to avoid such discussions if they believe this allows them to have some more time to come to terms with diagnosis or to focus on another priority. This would be a cognitively driven decision rather than one driven by any emotional response and would be made with knowledge and an acceptance of any risks associated with delaying an understanding of the options.

Healthcare professionals enter the collaboration of decision-making armed with considerable tacit knowledge having often had many similar discussions before. However, people with MND have recently had all expectations about how their future years may look destroyed by receiving the MND diagnosis (Locock *et al.,* 2009). People with MND can enter decision-making lacking an understanding of the disease and the decision, while seeking a way to cope with the emotional, psychological and physical consequences of the disease

(King *et al.,* 2009). People with MND are also not well-practised in making clinical decisions; a complex task that may be taken for granted by healthcare professionals. Following diagnosis, people with MND are faced with having to engage in multiple complex discussions including information about the disease and treatment options without having been empowered with the skills and knowledge required. Therefore, people with MND require support to prepare them for engaging in meaningful deliberation of their options in collaboration with their healthcare professionals, include addressing the emotional consequences of decision-making.

Such support could come in the form of a decision coaching role within MND teams. Decision coaching is defined as '*non-directive support delivered by a healthcare provider to help patients prepare to actively participate in making a health decision*' (Jull *et al.,* 2021). The decision coaches are an adjunct to the healthcare team, remain neutral in relation to the decisions being made and aim to enable patients to participate in good quality SDM (Stacey *et al.,* 2008). A Cochrane review found low quality evidence for decision-coaching delivered with evidence-based information to improve patient knowledge, though there was uncertainty about whether the intervention improved preparation for decision-making (Jull *et al.,* 2021). As identified previously, MND is a 'worst-case decision-making context' (Hogden 2014), and therefore the impact of such supportive measures may be more significant than in other clinical contexts. A previous case study found that patients making decisions about renal replacement therapy, were clearer about the decision problem following a session with a decision coach (Buur *et al.,* 2023). There is potential for the decision coach role within MND teams to have a wider remit than just the gastrostomy decision. It is expected that people with MND will have to make a range of complex decisions throughout the disease course, often simultaneously, contributing to the complexity of decision-making in MND care (Martin *et al*., 2014). Therefore, there may be substantial benefits of empowering people with MND with the skills, knowledge and coping strategies to engage in multiple future decisions. Further research is needed to develop and evaluate decision coaching and other interventions that may empower and promote people with MND to engage meaningfully in discussions about their treatment options.

8.3.3 Informing decisions about timing of gastrostomy: people with MND need concrete guidance

The guidance and evidence informing the timing of gastrostomy

The findings of all components of this study pointed towards a lack of healthcare professional consensus and uncertainty among people with MND about the timing of gastrostomy decisions. The uncertainty had the potential to contribute to delays in gastrostomy placement with half of healthcare professionals in the survey believing people with MND had gastrostomy placed too late. Healthcare professionals referred consistently to how the published guidance and research informed their clinical reasoning and preference for the timely placement of gastrostomy to promote positive nutritional outcomes and reduce disease and procedure related risks. However, there are some contradictions in the published guidance that may not be consistent with the clinical recommendation to encourage the placement of gastrostomy prior to significant progression of malnutrition, dysphagia or respiratory failure. For example, in the absence of good quality evidence to support the optimal timing of gastrostomy placement the authors of UK's NICE MND guideline (NICE, 2019) conclude that early placement of a gastrostomy should be considered to 'prevent' malnutrition and reduce the procedural risks associated with placing gastrostomy later in the disease course. However, in contrast, the same authors state that *'gastrostomy placement should only be considered with the presence of symptoms including weight loss, dysphagia and compromised respiratory function'*. These two positions outlined above are not consistent or compatible with each other for a number of reasons.

First, if gastrostomy is considered following the presentation of indications, then this does not represent the intervention as a preventative measure. Instead, it is being framed as a reactive measure to correct nutritional losses. Consistent with previous case series more than half of healthcare professionals responding to the survey believed gastrostomy should be placed following the progression of dysphagia or >10% weight loss since symptom onset (Beggs *et* al., 2010; Rio *et al.*, 2010). Second, using the presentation of symptoms to inform the timing of decisions about gastrostomy placement fails to acknowledge the time it takes services to arrange the admission for the procedure (Labra *et al.,* 2020). This was an important consideration in the deliberations of healthcare professionals and people with

MND about the timing of decision-making. The survey finding that 44% of healthcare professionals informed people with MND they would wait >4 weeks to have their gastrostomy placed was consistent with all the cases reporting up to a 3 month wait for gastrostomy placement following a decision being made. Previous research has confirmed that people with MND can experience additional nutritional losses following being identified as indicated for enteral nutrition while waiting for gastrostomy to be placed (Son *et al.,* 2024). The risks associated with procedural and disease-related risks escalating while waiting for the placement of a gastrostomy should feature prominently in discussions between people with MND and healthcare professionals. There is a need for professional guidance, and healthcare professionals, to be more concrete about the timing of decisions about gastrostomy, acknowledging the uncertainty associated with disease progression and the limitations of healthcare services to quickly place gastrostomy tubes. People with MND require support to make decisions in the presence of this uncertainty and on behalf of their future selves, if they wish to have the tube in-situ at the optimal time to optimise any benefits of enteral nutrition and reduce procedural risks. Ideally decision support should include the use of objective information that may help inform people with MND about likely progression of any of the indications for enteral nutrition. For example, the rate of progression could be informed by recent changes of functional scores or measures of nutritional status, dysphagia, respiratory failure or mealtime burden. In the absence of this objective risk information, healthcare professionals have a responsibility to use their experience of the MND disease course and local services to help guide decision-making. Healthcare professionals need to clearly delineate the timing of the decision from the timing of gastrostomy placement. As one Case 1 healthcare professional identified, there is a need to understand that there is both a decisional and procedural cut-off for gastrostomy placement.

Healthcare professional’s role in guiding the decisions of people with MND about timing of gastrostomy placement

Healthcare professionals play an important role in relation to the decisions of people with MND about timing of gastrostomy not just in terms of the information they share but ‘how’ they share it. Unlike people with MND, healthcare professionals are in possession of considerable experience of the likely disease progression including the consequences of worsening swallow and subsequent risk of aspiration and malnutrition. This includes not only the risk of poorer clinical outcomes, but also in relation to how people with MND and their caregivers experience life with progressing dysphagia (ProGAS study *et al*., 2015; Lisiecka *et al*., 2021b; Lisiecka *et al*., 2022). In line with previous research, people with MND in the case studies often required healthcare professionals to validate they had reached the right time to agree to gastrostomy (Genuis *et al.,* 2023). How healthcare professionals framed information about timing, including the use of recommendations, influenced the decisions made by people with MND. The survey revealed a lack of consensus about whether healthcare professional should share their preferences for, or against, gastrostomy placement in the form of giving clinical recommendations. Interestingly, there was also variation in whether healthcare professionals shared their preference for people with MND accepting, delaying or declining gastrostomy and differences in the use of recommendations between different professional disciplines. The shared model of decision-making proposes that not just patients, but also healthcare professionals share their preferences for treatments (Charles *et al.,* 1999; Makoul *et al.,* 2006, Breckenridge *et al.,* 2015). However, healthcare professionals have a responsibility for sharing their preferences for all the options available and not just focus on that of intervention. A subtle but important shift in framing of SDM would remind healthcare professionals of their responsibility to share information and preferences of *all options* available to people with MND including of continuing without or declining gastrostomy (Chan *et al.,* 2017; BMA 2024).

One challenge of employing SDM when making decisions about gastrostomy placement, surrounds supporting people with MND to evaluate how the range of indications and associated outcomes, align with their values. The current study captured how healthcare professionals can focus on the procedural and disease-related risks of delayed placement and less on the outcomes of enteral nutrition. For people with MND to effectively understand the complexities of the decision, the approach to supporting decision making should aim to meet a number of important objectives. First, to support people with MND to understand the health threat, including the consequences of the key indications for enteral nutrition on their outcomes and experience of living with the disease. Second, facilitating people with MND to explore how the different outcomes align with their values and which they may place more weight on in relation to making a decision. Third, identifying and addressing the cognitive and emotional representations people with MND have of the diagnosis and disease progression, and how these influence their behaviours during decision making. Fourth, support people with MND to manage the uncertainty associated with decision making, in particular having to consider making decisions ahead of their being a significant clinical need.

8.3.4 Coordinating decision support across the MDT

Distributed decision-making

The model of MND care on each case influenced healthcare professionals ability to coordinate decision support across the MDT. The current study confirmed the findings of previous research that decision-making about gastrostomy in MND care is distributed over people, space and time (Labra *et al*., 2020; Van Eenennaam *et al.,* 2021; Son *et al*., 2024). Decisions about gastrostomy were made longitudinally following interactions with multiple healthcare professionals working in different settings. A distributed model of decision-making has been proposed as being more appropriate than those that focus on decisions made solely between dyads of people with MND and their healthcare professional (Rapley *et al.,* 2008). For example, a longitudinal account of decision-making was captured in the case study medical note reviews with a range of assessments and interactions between healthcare professionals and people with MND taking place between gastrostomy first being introduced through to decisions being made. The healthcare professionals taking responsibility for discussing gastrostomy with people with MND operated across the clinic and community settings including belonging to different NHS Trusts (OPM, 2016). Decision-making distributed over so many people and settings consistent and coordinated decision support, not just in relation to content but also how and when information is delivered (Chapman *et al.,* 2021). The MDT nature of decision support adds another layer of complexity to a decision that is already a very difficult one for healthcare professionals and people with MND to make.

The risks associated with an inconsistent approach

The distributed nature of decision support has the potential to impact on the ability of people with MND to make informed and timely decisions, if the approach taken is not well coordinated. The survey confirmed the findings of previous studies (Oliver *et al.,* 2011;

Ruffell *et al.,* 2013; Van Eenennaam *et al.,* 2021) of a lack of professional consensus about how clinical outcomes are used to inform the timing of discussions and decisions, the information shared with people with MND and how information is framed. These differences in beliefs and practice of different healthcare professionals, increase the risk of people with MND receiving mixed messages that could impact on uncertainty and lead to decisional conflict (O'Connor 1995; Garvelink *et al.,* 2019). Such decisional conflict would be a significant concern if associated with delays in decision-making that could allow disease and procedure related risks to escalate (ProGAS study, 2015; Son *et al.,* 2024). There is a need to better understand how the decision support delivered across the MDT effects the quality of decision-making, the timing of decisions and the impact on health, quality of life and experiential outcomes.

Coordinated decision support: a possible mechanism for the efficacy of MND MDT care

While MDT care is broadly accepted as the optimal model of MND care delivery supported by evidence that it improves survival and quality of life (Ng *et al.,* 2009; Aridegbe *et al.,* 2013; Rooney *et al.,* 2015; De Almeida *et al.,* 2021) there remains a lack of evidence focusing on the mechanisms behind any beneficial effect (Soukup *et al.,* 2018). Improved clinical decision making between HCPs with expertise in MND, good quality shared decision-making between healthcare professionals and people with MND, and the efficient access to interventions may contribute to the survival benefits associated with support from a specialist MND MDT (Aridegbe *et al.,* 2013; Rooney *et al.,* 2015). A recent review applied a complexity lens to MND MDT care capturing the varied and complex clinical considerations that healthcare services are expected to cover (Berlowitz *et al.,* 2023). However, the review did not extend to how MDTs interact, collaborate and coordinate the care delivered. There has been a lack of research evaluating regional MND healthcare services perform against the principles of good quality teamwork. The coordination and delivery of decision support about gastrostomy which uniquely involves so many professional disciplines across many settings, could be one variable to focus on in such research.

There was evidence of the purposeful use healthcare professional expertise and settings, and the staggering of information delivery. However, there was no strategy documented or implicit, agreed across the healthcare professionals involved about when and how the MDT would initiate and continue discussions about gastrostomy, or criteria by which people with MND would be encouraged to make decisions. Previous qualitative studies have identified how the MDT care model may impact on the experience people have of living with the disease (Hogden *et al*., 2012a). A key to understanding and evaluating how people with MND are supported to make decisions is the development of interprofessional models of SDM (IP-SDM) models or frameworks that have been validated in the context of MND MDT care delivery. IP-SDM models have been developed and validated or evaluated in other settings including primary care (Legare *et al.,* 2011b), home care (Legare *et al.,* 2013), end of life care (Davies *et al.,* 2021) and diabetes (Yu *et al.,* 2015). Decision-making models developed for MND care have focused on the cyclical nature of decision-making of individual people with MND, but lack reference to the involvement of the MDT, which is particularly relevant for decisions about gastrostomy (King *et al.,* 2009; Hogden, 2015). There is a need to develop models or frameworks that capture the importance of MDT coordination or teamwork, and the wider contextual factors that influence the engagement (e.g. emotional response) and deliberations of people with MND.

Combining the principles of inter-professional shared decision making and high performance teamwork: a route to delivering quality MDT decision support

MND guidance consistently recommends that people with MND are supported by a range of health and social care disciplines following diagnosis to assess and manage the range of consequences of the disease (Miller *et al.,* 2009b; NICE, 2019; Shoesmith *et al.,* 2020). The World Health Organisation (WHO) have defined good quality neurology care as an integration of tertiary, secondary and primary care that demonstrates coordination, accessibility, continuity, comprehensiveness and equity (WHO, 2006). The delivery of SDM across the MDT is intricately linked to the quality of interprofessional practice in general (Legare *et al.,* 2011b). Inter-professional communication and taking an MDT strategic approach, were identified in the case studies as possible mechanisms of a coordinated approach to decision support across the MDT. The finding from the survey that healthcare professionals were not fully satisfied with the quality of communication or consistency across the MDT was explored in more depth in the case studies.

The importance of understanding what constitutes good quality teamwork in healthcare and the impact on patient care has received a lot of attention in the literature particularly in acute settings (Rosen *et al.,* 2018; Soukup *et al.,* 2018). Components of quality teamwork include organisation, leadership, communication, responsibilities, inclusiveness, cohesiveness and coordination (Rydenfalt *et al.,* 2017; Rosen *et al.,* 2018; Soukup *et al.,* 2018). However, there is a paucity of research that has sought to evaluate teamwork in healthcare teams outside of the acute setting (Rosen *et al.,* 2018). The competencies of an effective MND team are likely to differ from those required of teams in different clinical context such as emergency departments or those supporting life-long chronic diseases (Rosen *et al.,* 2018). The findings from the current study suggest that good quality coordinated teamwork is synergistic with good quality coordinated decision-support. For example, previous research found that joint visits with MDT colleagues were associated with indicators of effective teamwork including shared team goals, efficient organisation, and increased expertise (Nygren *et al.,* 2021). Within the case studies healthcare professionals valued joint working as an effective mode of communication and cohesive teamwork. Significantly more research has focused on MDT performance in oncology (Soukup *et al.,* 2018). While there are significant differences between decisions made in oncology and MND care in terms of the disease course, available treatments and the organisation of care, the approach to studying teamwork could be transferable to the MND context. Developing, applying and evaluating teamwork frameworks to the MND care context could generate a more nuanced understanding of how MND teams' impact on the outcomes of people with MND. Even within the three MND teams included in the case studies there were differences in how MND services were organised, which influenced their ability to deliver coordinated decision support. Supporting MND services to become high-functioning teams could be the key improving the delivery of SDM across the MDT.

Good quality interprofessional communication is key

A lack of accessible and detailed communications within the MDT limited HCPs awareness of the current indication for gastrostomy of people with MND and any progress made with decision-making. Guidance includes a recommendation that all HCPs are informed about any decisions that people with MND make about their care (NICE, 2019). To improve coordination of decision support, this recommendation could be expanded to include sharing with the MDT progress made with initiating and continuing discussions about care options, rather than only the making of a decision. A previous study identified how healthcare professionals lacking awareness of the current understanding of people with MND influenced the quality of clinical interactions, particularly when discussing sensitive topics (Bongioanni *et al.,* 2023). Such breakdowns in MDT communication increase the risk of healthcare professionals missing opportunities to open or progress discussions about gastrostomy. Strengthened lines of communication between healthcare professionals could facilitate people with MND involvement in discussions and support a more coordinated approach. The introduction of an interdisciplinary communication tool into four different clinical settings supported the flow of information that was relevant to patients' decisions about life prolonging treatments across the MDT (Sohi *et al.,* 2015). An organisational approach to improving teamwork performance can include facilitating communication that is open (i.e. HCPs feel able to share views), of good quality (i.e. shared frame of reference) and available (i.e. opportunities to communicate) (Rydenfalt *et al.,* 2017). A systematic review of interventions designed to improve teamwork included the use of communication tools that facilitate MDT interaction, trigger action and improve knowledge about patients (Buljac-Samardzic *et al.,* 2020). Coordination of care is dependent on good quality team communication (Rydenfalt *et al.,* 2017). Healthcare professionals within the cases studies had implemented similar strategies to communicate with the MDT such as databases, clinic proformas and an IT dashboard synthesising the results of assessments of people with MND. However, none were shared across all settings and accessible to every healthcare professional involved in discussions about gastrostomy. Further barriers to communication were the geographically and organisationally distributed nature of the MDT, which prevented frequent quality interactions between healthcare professionals.

Taking a strategic approach to coordinating decision support across the MDT

A complex intervention development approach including the collective implementation and evaluation of communication tools, organisational measures and healthcare professional training (on teamwork and SDM) may create an optimal framework within which effective MDT SDM can thrive (Rydenfalt *et al.,* 2017; Rosen *et al.,* 2018; Buljac-Samardzic *et al.,* 2020; Skivington *et al.,* 2021). Simultaneously enhancing the competencies of healthcare professionals and MND teams in teamwork and SDM could address the barriers to consistency, communication and coordination identified in the current study. A key difference between the cases included in the case study was the continuity of care across the clinic and community setting. Within Case 3 most HCPs reviewed people with MND in clinic and community settings, whereas within Cases 1 and 2, the teams were mostly distinct (and even belonged to different organisations). The continuity of care systems (i.e. same team involved in acute and community setting) in mental health care was found to be associated with better outcomes and preferred by patients and healthcare professionals (Omer *et al.,* 2015). Consistent with the findings of the current study, barriers to delivering SDM across MDT's included poor communication between healthcare professionals who are disconnected from each other (Brogan *et al.,* 2019). There are good reasons why the MND model implemented in Case 3, and therefore their approach to coordinating MDT decision support may be challenging to implement across all MND Centres. Case 3 was able to implement this model partly because of the smaller MND population they were responsible for. Repeating this for all regions may require a lot more healthcare professional resource or a larger number of MND centres which may not be feasible from a financial and human resource perspective. However, this does not mean that other MND Care Centres and the wider community MDT cannot seek to strengthen the communication between healthcare professionals and collaborate to develop strategies that support coordination of decision-support.

8.4 Strengths, limitations and reflexivity

8.4.1 Strengths of the research

The series of research presented in this thesis is the first to aim to understand how MND teams support people with MND to make decisions about gastrostomy placement including through studying the phenomenon in real-world settings. A major strength of the research presented is the combination of breadth and depth focused on the topic within the same study. The quality of the research is supported by the rigour with which the study was conceptualised and designed. The conceptualisation of the project has been years in the making including meeting with healthcare professionals working in MND care, collaborating with a variety of PPI groups and people with MND, and exploring the research design options with multiple academics. The rigorous design process confirmed that the research question was relevant to those people who are involved in making decisions about gastrostomy in MND care and that there was a clear gap in both academic understanding that also represented a real-world problem in MND practice. The qualitative evidence synthesis provided a rigorous review of the literature. The rationale for including in the review, studies about ventilation and gastrostomy decision-making, was that both interventions have far reaching implications for people with MND, caregivers and healthcare professionals. Therefore, important conceptual findings were strengthened through synthesising qualitative transferable across these different decisions. These important conceptual findings transferable across decision-making contexts, were then able to be explored in more depth in the survey and multiple case study.

Each component of research guided, designed and implemented by published methodological texts and reporting guidelines, and through my own learning from attendance at courses on the each of the methods used. This knowledge, in addition to collaboration and feedback from supervisory, academic and PPI groups, informed the development of detailed protocols for each component of research. A protocol is an essential part of performing case study research (Yin, 2018; pg 93-105) as it not only supports the credibility of the study, but it also provides a guide with which recruitment and data collection will take place within each case. Documenting the methods used in study protocols strengthens the dependability of the qualitative components and reliability of the survey.

Credibility of the findings are further supported by the sampling strategies used. The systematic approach, including a second reviewer, to identify all the qualitative papers that met the inclusion criteria in the review, ensured that the conceptual themes developed represented the current understanding in the literature including any alternative explanations. While snowball sampling will always be associated with selection bias, significant efforts were made to ensure that the survey was distributed to the target population with the results showing representation from the key professional disciplines involved in supporting people with MND to make decisions about gastrostomy. Finally, the purposive sampling of cases, participants, observations and settings in the case studies broadened the data made available for inclusion in the analysis. The successful implementation of the sampling strategies, approach to recruitment and the rigorous analysis, supports the credibility and transferability of the case study findings.

A further strength of the case study is the rigorous approach taken to the framework analysis and the thick description of the case study findings. The data sought to represent a credible account of the phenomenon and context as it occurs in real-world practice through accessing multiple perspectives and using methods that viewed the phenomenon from both emic and etic positions. This supports the theoretical transferability of the findings that could be applied to settings outside of those included in the study. The studies have likely identified concepts that are also transferable to other decisions in MND care and to MND MDT care in general. For example, the emotional responses people with MND have to the diagnosis and discussions about future disease progression could also be relevant to how MND teams engage people with MND in discussions about NIV, home adaptations or end of life care.

8.4.2 Limitations

There are limitations associated with the study. First, due to limitations of time and resource, it is inevitable that important views, experiences, perceptions have not been captured. Confidence in the dependability of the review findings could have been improved through including a second reviewer to screen all abstracts and full text papers for inclusion, or through having a second coder during the analysis (Green and Thorogood, 2014, p. 229). The survey was likely biased towards the beliefs and practice of dietitians due to my increased accessibility to this profession (see reflexivity section 9.5) and lack of funding to pay other groups to disseminate the survey. Recruiting increased numbers of doctors, nurses and SLT would have strengthened the validity of the comparisons made between professions responses. Finally, only a limited amount of time was spent in the field in each case. While I was successful in recruiting a wide range of professional disciplines within each case, there were many interactions that I would have not been present at which would have provided a more in-depth understanding of the phenomenon. I was able to observe healthcare professionals practice a lot more frequently than the behaviours of people with MND. Future research should work with ethical panels, PPI and cases to optimise the opportunities to observe patients, including considering longitudinal research designs.

There are limitations to how generalisable or transferable the findings are. For example, defining a sample frame for the survey would have strengthened the generalisability of the findings (Blair *et al.,* 2014, p. 111-127). This could have been achieved through only performing the survey on the cases included in the cases studies, though this would have limited the breadth of the survey and excluded evaluating the practice and beliefs from across UK MND care. Inclusion of only three cases in the multiple case study will limit the transferability to other MND teams in the UK. The case studies alone have highlighted how the models of MND care can differ between care centres. It is likely there are other models of MND care employed in the UK which may exhibit other characteristics that could influence how people with MND make decisions about gastrostomy. Therefore, though many of the concepts presented are theoretically transferable to other settings they may not apply completely to other centres. Equally, MND care around the world also varies in terms of how it is funded and organised, in addition to the cultural and legal differences that may impact on how decisions are made (WHO, 2006; Kierkegaard *et* al., 2021; Bublitz *et* al., 2024). It is not possible to comment on how relevant these findings are for MND teams and people with MND outside of the UK setting.

8.4.3 Reflexivity

Reflexivity involves 'an examination of the foundations of frameworks of thought' (May and Perry, 2017, p. 3). It involves thinking about how my previous experiences, values, and beliefs have shaped how I think about the phenomenon under investigation, the research I am conducting and about the social world in general, and how my positionality influenced my analytical decisions and interpretations (Cresswell 2013, p. 216). My subtle realist position involved taking the position that there is an external social reality available to be studied, and that credible knowledge could be attained through the use of mixed-methods methods. Acknowledging that I cannot completely separate myself from the construction of the accounts I present, increased the importance of reflexivity throughout the conceptualisation, design, conduct, analysis and reporting of the research. Below I move beyond the personal history provided in the introduction (see section 1.5) and give an account of my reflexivity while conducting the research.

The personal connection I had with MND, through my grandad passing away with the disease when I was 15 years old, played an important role in my initial motivation to focus my clinical and academic careers in the MND field. Even though this happened when I was at a young age, it is something I continue to reflect on and motivates me to want to support people with MND to live better lives with the disease. This experience drives me to want to make a difference either in the present through directly supporting people with MND to live with the disease (i.e. through my clinical role), or in the future through generating new knowledge in my research about how healthcare services can better support people living with this dreadful disease. During, and since, completing this research I have reflected on whether my personal connection with MND influenced my thinking or decision-making during the study. I do not think it directly influenced my perceptions or interpretations of the qualitative data being collected; certainly, less so than my professional experience of MND. However, it did partly influence my decision to explore the topic using qualitative methods, motivated by a belief that the disease is experienced within an extremely complex clinical, social and psychological context and not in terms of objective measures of clinical decline we refer to in practice and as primary outcomes in studies. I have a strong belief that understanding how people with MND experience the disease is key to improving the care healthcare services deliver. It was important to be aware of these potential influences on my thinking to ensure any decisions made about research methods remained academically sound, and that any analytical interpretations were informed by the qualitative data collected.

My position as a dietitian of >20 years including having a wide network and presence on national specialist dietetic groups, meant that dietitians and dietetic groups were more accessible to me when recruiting to the survey. This was evident from the recruitment of significantly more dietitians than doctors, nurses or SLT, which was not representative of MND teams. I met a lot more resistance from non-dietetic groups to disseminate the survey or the requirement for payment which I did not have the budget for. However, with dietetic groups, as a result of my professional connections with them, the gatekeepers were more accommodating and provided significant assistance in facilitating the snowball sampling strategy. With hindsight this should have been expected and highlights the need to be reflexive when planning and costing similar surveys. I could have spent more time in the planning phase to ensure that I spent additional time and resources collaborating with other professions to allow wider distribution of the survey.

A further challenge associated with being a dietitian researcher relates to how my professional experience supporting people with MND to make decisions about gastrostomy may have influenced the analysis of the qualitative data. Such beliefs may have influenced the findings that I chose to present or how they were framed. I developed my analytical ideas through discussion with my supervisors and continued to return to the data summaries and original transcripts to ‘check in’ that they were grounded in the data. Thick descriptions are reported that position the findings within the cases in which they were captured including the careful use of quotes and the active search for alterative explanations.

While I purposely did not inform participants of my clinical experience, inevitably this became known to many, particularly the healthcare professional participants. I had previously presented widely at conferences on the topic of gastrostomy decision making and developed online resources to support the decisions of people with MND about gastrostomy; resources used by healthcare professionals and people with MND within each case. There were some incidents where participants awareness of my previous clinical experience impacted on data collection. While gaining consent a caregiver made it known that he had researched me online and my professional experience. This took me by surprise, and I was unsure of how to respond. My response involved acknowledging that he was correct and clarifying I was present as a researcher and not an healthcare professional. Subsequently, while observing the same participant discussing gastrostomy in clinic, the caregiver broke from their discussion with the neurologist to identify myself as an expert in this area. Again, I sought to minimise any engagement in conversation, to allow the clinical consultation I was observing to continue. At a later date, when interviewing the same person with MND and caregiver, they questioned whether the neurologist had only discussed gastrostomy because I was present. I acknowledged that I could not say for sure what the healthcare professionals’ motivations were or control the conversations that they chose to have while being observed. I reassured the participants that I encouraged all HCPs observed to continue with their normal practice, including to only discuss gastrostomy if this was what they would normally do. I also advised that I had observed lots of consultations where gastrostomy had not been discussed. Another scenario involved a neurologist showing a person with MND an online video featuring me during a clinic consultation. This revealed me as not just a researcher but also a healthcare professional interested in and with knowledge of the clinical area. In both situations the observation and interview were able to continue but there is the risk that the data collected was influenced by my presence as a healthcare professional researcher.

The concerns highlighted above capture the challenge associated with performing observations as a healthcare professional researcher with a reputation and experience in the topic area. Therefore, maintaining a reflexive approach was even more important. With regards the sampling of participants and settings, I was careful to ensure this was informed by my understanding of the literature, my increasing familiarity with each case and the evolving analysis, rather than any preconceived ideas I may have constructed as part of my own practice. Despite all efforts, I acknowledge that that the practice and disclosures of particularly healthcare professional participants may have been affected by having an awareness of my professional background. I have attempted to counter any threats to credibility through the rigour with which I have collected, analysed and interpreted the data. This has included ensuring fieldnotes and transcriptions accurately reflect the data being collected, checking the decisions I made in relation to the coding or development of themes was grounded in the data, and discussing the analytical phase of the framework approach with supervisors.

However, I believe there were examples of where my previous professional experience supported the conduct of the study. For example, having an underlying knowledge of how MND care is delivered supported the initial conceptualisation of the problem and enabled me to identify the gaps in understanding in both the current literature and practice. I was also able to use my position to easily reach out and organise meetings with other MND healthcare professionals to discuss my evolving ideas, confirm the research was relevant and inform the development of the research plan. This experience also supported my collaborations with the range of PPI groups and individuals with MND I met, allowing me to build rapport and have some insight into the topics discussed. I also benefited from being comfortable in the clinical settings and with communicating with people with MND with speech difficulties or who can become emotional during interviews. My familiarity with these scenarios allowed me to be sensitive to the experience of people with MND participating in the research while still remaining focused on the aims of the research.

8.5 Implications for future research

A number of research priorities have been identified through completing the series of research presented in this thesis. First, research is required that seeks to evaluate approaches to engage people with MND in timely discussions about gastrostomy. The current study highlighted the opportunity and potential benefits of better preparing people with MND for engagement in discussions. This could include evaluating the effect of psychological interventions or decision support interventions on the preparedness or participation of people with MND in decision-making discussions.

Second, is the need for better quality data that supports people with MND and healthcare professionals' decision making about the timing gastrostomy placement. The current study revealed that the timing decision was associated with significant uncertainty. Some people with MND wanted more concrete information to guide their decisions. While a randomised controlled trial of enteral nutrition in MND care is unfeasible for ethical reasons, observational studies could provide better quality risk data through employing a stratified sampling approach e.g. recruiting people with MND with different MND phenotypes, rate of progression or severity of gastrostomy indication. Such data could be integrated into risk communications in decision aids.

Third, there is a need to perform a decisional needs assessment in relation to the decision people with MND make about the timing of gastrostomy (Jacobsen *et* al., 1999). The decision aids currently available either lack applicability to the UK setting (Hogden *et al.,* 2016) or were developed to inform decisions about 'if' rather than 'when' to have a gastrostomy (Maunsell *et al.,* 2019). In addition to understanding the risk communications required, such a decisional needs assessment could also seek to account for there being a decision and procedure cut-off and the impact of the waiting time for gastrostomy placement.

Finally, there is a need to generate a better understanding of how MND MDTs perform as teams, including in relation to how healthcare professionals coordinate decision support. Research opportunities include developing, adapting and/or validating interdisciplinary shared decision-making models (Legare *et al.,* 2011a) for the MND care context, including identifying the mechanisms that are associated with optimal decision process outcomes

(Waldron *et al.,* 2020). Other opportunities include informing the design and conduct of interprofessional SDM using teamwork frameworks.

8.6 Implications for practice

The considerable time spent in the field in different centres, discussions with supervisors and colleagues and my own reflections as a practicing HCP have informed the identification of a number of implications for practice of the current study. First, relates to healthcare professionals aiming to adhere to the professional recommendation to discuss gastrostomy with people with MND early in the disease course. Healthcare professionals intending to introduce gastrostomy should prepare people with MND for these discussions. For example, healthcare professionals should ensure they are aware of the outcome of previous assessments or discussions relevant to the decision. This could facilitate healthcare professionals to progress the understanding of people with MND rather than repeating or contradicting what has already been covered. Understanding what people with MND already know and establishing their perceptions and responses to previous discussions, will enable healthcare professionals to identify any emotional response or coping strategies being employed. Healthcare professionals should communicate a clear rationale for why they are discussing the option now. Finally, it is important that people with MND remain in control. Healthcare professionals providing a clear rationale for discussions (particularly in the absence of any indications) and seeking permission for discussing disease progression and intervention options, gives people with MND the opportunity to learn about their options while still respecting any informed preference to delay the discussion.

Second, people with MND want and need clear and concrete guidance about the timing of gastrostomy placement to address the uncertainty associated with the decision. People with MND value healthcare professionals sharing their preferences for intervention in the form of recommendations and some prefer a direct approach. However, it was evident from the survey that not all healthcare professionals believe they have a responsibility to give people with MND recommendations and also use them selectively. With consent, healthcare professionals should share their preferences for or against gastrostomy, ideally accompanied with a rationale and having established knowledge about the values and goals of people with MND. The risk of further disease progression while waiting for gastrostomy placement after making a decision, is an example of where healthcare professionals could better support people with MND with concrete guidance, to make informed decisions about the timing of gastrostomy placement.

Third, gaining an understanding of the preferences of people with MND for how they wish have information shared with them will support healthcare professionals to tailor their communication to the individual needs of the people with MND. Use of effective communication skills are crucial to promote engagement in discussions and support people with MND to feel listened to. These are skills that not all healthcare professionals possess and therefore it should be mandatory healthcare professionals involved in these discussions undertake training in advanced communication skills.

Finally, individual healthcare professionals should acknowledge the longitudinal nature of decision-making including interactions with the MDT. This highlights the importance of understanding what has been discussed before and feeding forward what is discussed during interactions. This can be achieved firstly through improved interprofessional communication, including enough detail for the next healthcare professional to understand what progress has been made. Regional MND teams, including healthcare professionals in the clinic and community setting should seek to collaborate to come to a consensus about how they plan the initiation of discussions, the localised information shared, realistic expectations about how long it takes to organise placement, and assignment and clarification of professional responsibilities.

8.7 Unique contribution to knowledge

This is the first study to apply a mix-methods approach to develop an in-depth understanding about how the MND MDT supports people with MND to make decisions about gastrostomy placement. The study has identified that people with MND experience an emotional response to the presentation of information about disease progression that influences their engagement in discussions and the timing of decisions. Proactively addressing the emotional response prior to sharing information about the intervention may be key to opening timely and constructive discussions. The decision about the timing of gastrostomy is particularly associated with uncertainty with people with MND often needing concrete healthcare professional guidance and validation to overcome instinctive avoidant coping strategies.

The study confirmed that decisions about gastrostomy are made over time and are the outcome of interactions distributed across many healthcare professionals and settings. The regional MND care model employed influences healthcare professionals’ ability to deliver coordinated decision support. A coordinated approach is influenced by the quality and accessibility of communication between healthcare professionals that supports an awareness of current indication and progression with decision making. A lack of strategic approach contributes to the risk of inconsistencies, contradictions and missed opportunities to open discussions or build on the informed decision-making of people with MND. The findings of this study have confirmed that this is a complex decision heavily influenced by the context in which it is occurring.

8.8 Conclusion

The findings of this research have confirmed that decision-making about gastrostomy in MND care are complex decisions heavily influenced by the context in which it is occurring. The emotional responses of people with MND and uncertainty about disease progression play important roles in when discussions are initiated and the timing of decisions. Healthcare professionals have a complicated responsibility to inform people with MND about their options while simultaneously managing the emotional responses that are experienced in relation to their perceptions about the diagnosis, disease progression and the prospect of needing a gastrostomy. Decisions are distributed over time, space and people, including people with MND interacting with several healthcare professionals operating in different settings or organisations. Variations in healthcare professionals’ beliefs and practice, an absence of MDT decision-support strategy and a lack of quality and accessible interprofessional communication can contribute to a less coordinated approach to delivering decision-support across the MDT. MND teams should develop a strategic approach, grounded on strong lines of interprofessional communication to coordinate the decision support they deliver to people with MND.

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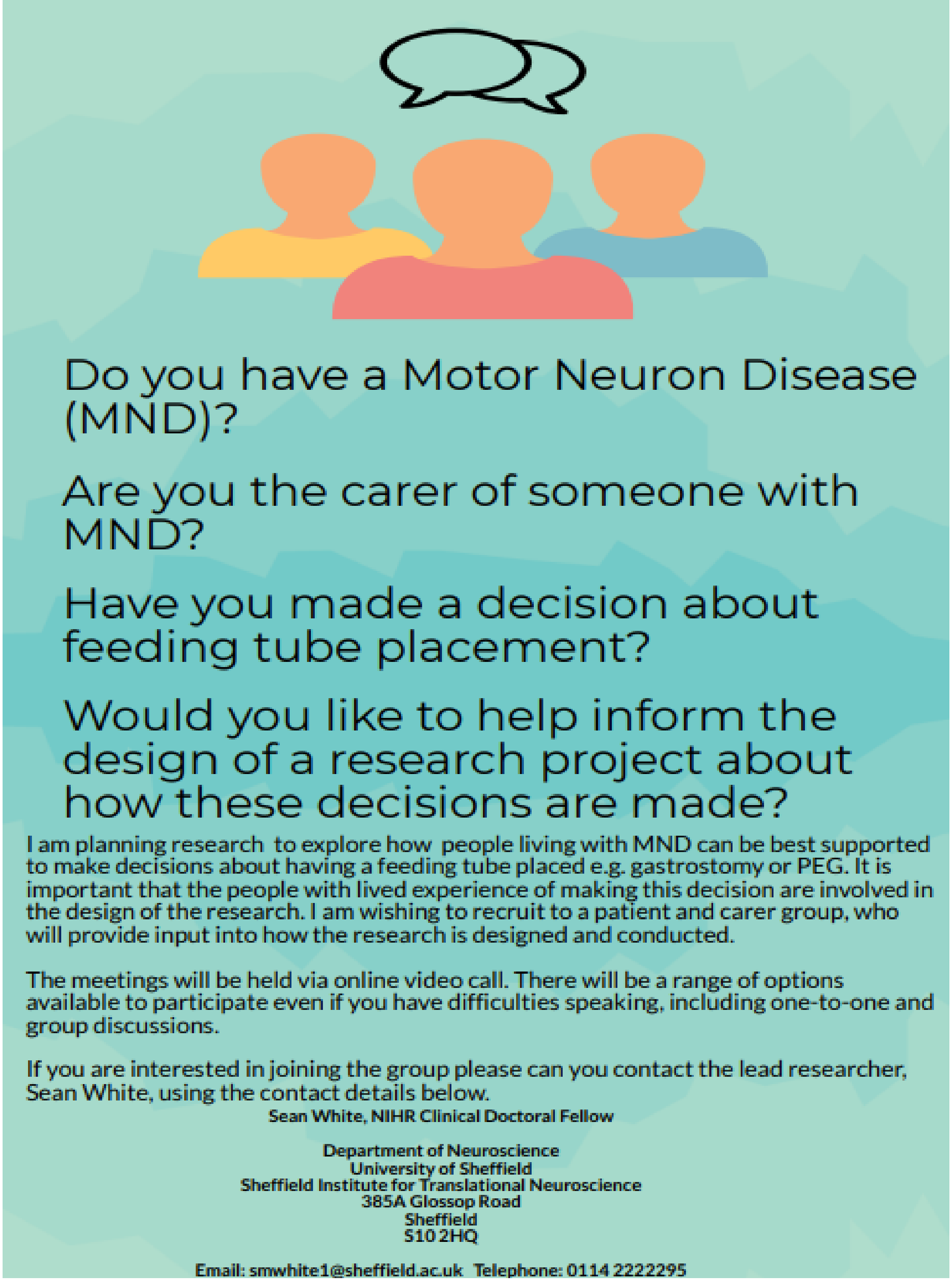
Appendices

Appendix 1 Advert for PPI panel.



Do you have a Motor Neuron Disease (MND)?

Are you the carer of someone with MND?

Have you made a decision about feeding tube placement?

Would you like to help inform the design of a research project about how these decisions are made?

I am planning research to explore how people living with MND can be best supported to make decisions about having a feeding tube placed e.g. gastrostomy or PEG. It is important that the people with lived experience of making this decision are involved in the design of the research. I am wishing to recruit to a patient and carer group, who will provide input into how the research is designed and conducted.

The meetings will be held via online video call. There will be a range of options available to participate even if you have difficulties speaking, including one-to-one and group discussions.

If you are interested in joining the group please can you contact the lead researcher, Sean White, using the contact details below.

Sean White, NIHR Clinical Doctoral Fellow

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Appendix 2 Example of feedback sent to PPI panel.

Sean White's NIHR Clinical Research Doctoral Fellowship

UPDATE

Issue of
June 24, 2022

The literature review is nearly complete. After 9 months of bringing together all of the qualitative literature (studies that collect data using interviews, focus groups and observations), the findings are now being written up for publication. A summary of the findings a described below.

The survey is out there. The review findings informed the development of an online survey to understand healthcare professionals practice and attitudes towards supporting people with MND to make decisions about gastrostomy placement. The survey was launched on Monday 13th June 2022

Planning for the multiple case study is underway. The major part of the research plan is due to take place next year, observing how decisions are made in real-world practice. I will value your input into the development of this stage of the research. More information below.

Decisions in MND care: 'it's complicated'

Following an extensive search of the published qualitative literature that focused on the experience of making decisions about interventions (ventilation and gastrostomy) of people living with MND, their caregivers and supporting healthcare professionals, 28 papers were found. The findings from these papers were extracted and re-organized to identify what factors influence how decisions are made. The review findings are presented under four major themes which are summarized below.

Values based judgements

People with MND create a vision of how an intervention will affect their **quality of life**. These include the impact the intervention will have on their independence, body image and on their caregivers. Quality of life is often a higher priority than any potential to extend life.

Sharing the burden

The decision-making process and decisions made are influenced by others. **Healthcare professionals** are a valued source of credible information and support. However, the manner in which information is shared affects how it is received by people with MND. There are challenges associated with the many different professional roles involved in supporting decisions, with evidence that this can lead to mixed messages and confusion. **Caregivers** play a supportive role, seeking information and providing someone to discuss the options with. Decision-making can be complicated when the person with MND and caregivers' preferences for an intervention are not aligned.

Autonomy and control

People with MND, caregivers and healthcare professionals believe that the person with MND should have **control over final decisions**. However, there are instances where this **control may be threatened** such as when placed under pressure to make decisions by others. Healthcare professionals describe the challenge they face providing neutral and balanced information, when they believe interventions will have benefit for the person with MND.

Tipping the balance

Increased threat posed by symptoms of MND, can be the tipping point at which decisions are made. Prior to this, people with MND can prefer to focus their energy on coping with the **'here and the now'** which can present a challenge to beginning discussions about future interventions. The message of starting interventions **'earlier rather than later'** consistently encouraged by healthcare professionals can be viewed as unhelpful, with more concrete guidance preferred.

What next?

The review is currently being written up ready to submit for publication. A copy of the draft will be circulated for your information, and I would welcome any feedback you may have.

Survey of healthcare professionals

Using the findings from the review, discussions with yourselves (my PPI panel), topic academics and my supervisors, I developed a survey to investigate the practice and attitudes of healthcare professionals towards supporting people with MND making decisions about gastrostomy placement. The survey is split into two main sections: 1. How do healthcare professionals introduce and discuss the option to have a gastrostomy; 2. How do healthcare professionals coordinate and communicate while supporting decision making. The online survey was launched on 13th June and will remain open for 2 months. I will be interested in discussing the results with you.

Multiple case study plan

The final stage of the research involves myself going onto 3 MND 'sites' to observe how healthcare professionals support people with MND making decisions about gastrostomy. I will be collecting data using interviews, focus groups, observations and asking people with MND to record a diary about their experience of decision making. I need to develop lots of documentation to undertake this research including participant information sheet, consent forms, interview topic guides and the diaries. I will value your feedback and would like to spend the next time we meet discussing these.

Next meeting...

I would like to try to meet with you again within the next couple of months. I will send out some possible dates. Please tell me if you no longer wish to be involved and I can remove you from the emails. I cannot thank you enough for your support and it really does help guide how the research is designed and carried out. Looking forward to seeing you all again.

Are you interested in helping us understand how to best support people with motor neurone disease (MND) making decisions about gastrostomy feeding tube placement?

Appendix 3 ENTREC statement

APPENDIX A from Paper 1

See table A1

Table A1. A summary of how the review met the recommendations of the Enhancing transparency in reporting the synthesis of qualitative research (ENTREC) statement[24]

Item	Guide and description
Aim	Aim of the review is clearly described.
Synthesis methodology	Rationale given for thematic synthesis
Approach to searching	A comprehensive search strategy was planned to seek all available studies that relate to the research question.
Inclusion criteria	Inclusion criteria stated including population, language, year limits, type of publication, and study type.
Data sources	All database and supplementary sources included with dates of searches.
Electronic Search strategy	Search strategy described including the databases searched and the scope of the supplementary searches. Example of the full Medline search terms used included in appendix.
Study screening methods	Comprehensive description of study screening method, including role of second reviewer who screened 10% of the title/abstracts and full texts.
Study characteristics	Description and table of study characteristics including year of publication, country, population, number of participants, data collection, methodology, aim.
Study selection results	Study selection results provided in text and PRISMA flow diagram including data sources, duplicates and reasons for exclusions.
Rationale for appraisal	The results of the quality appraisal using the CASP qualitative appraisal tool was presented to inform the readers critical appraisal of the review findings.

Appraisal items	CASP qualitative appraisal tool used to assess the quality of the included studies
Appraisal process	Appraisal process conducted by single reviewer
Appraisal results	Appraisal results present in tabular form and included as an appendix.
Data extraction	All the PDFs of the selected studies were imported into NVivo for data extraction. Only text in the findings/results sections of publications were analysed.
Software	EndNote used to store the study publications. Excel used to manage the study selection process. NVivo used to perform the line by line coding and to manage the data while developing the descriptive and analytical themes.
Number of reviewers	The primary investigator (SW) screened all title/abstracts, full texts, completed quality assessment and analysis and synthesis. Second reviewer (LC) screened 10% of title/abstracts and full texts.
Coding	Thematic synthesis including line by line coding described
Study comparison	Where present differing perspectives on similar concepts were highlighted and discussed. The reviewer actively looked for alternative explanations for concepts throughout the analysis phase.
Derivation of themes	An inductive approach to the synthesis was taken
Quotations	Mostly participant quotes were included, including indicating whether the quotes were from a pwMND (P), caregiver (C), or HCP (H). A few author extracts (R) were also included within the findings.
Synthesis output	A pathway of care model was developed to capture

Appendix 4. Selecting the qualitative evidence synthesis using the RETREAT framework (Review question–Epistemology–Time/Timescale–Resources–Expertise–Audience and purpose–Type) (Booth 2018)

RETREAT component	Reasoning
Review question	The review will address an emergent question, that will take an interpretative approach to explore the lived experience of decision-making about invasive interventions in MND care.
Epistemology	Method choice not dependent on epistemological underpinning.
Time frame	6-9 months available to complete review.
Resources	The review will be lead by the researcher, with a colleague assisting with the process of study abstract and full text screening and study selection. Funding available for reference manager (Endnote), qualitative analysis software (NVivo) and access to a broad range of bibliographic resources.
Expertise	Single PhD student with some experience in primary qualitative research design and theoretical knowledge of qualitative evidence synthesis supported by attendance at qualitative evidence synthesis short course.
Audience and purpose	The outputs will inform healthcare professionals and academics interested in MND care and decision making.
Types of Data	The scoping searches have suggested the qualitative data will be containing conceptual detail and thick, in terms of the contextual descriptions provided.

Reporting standards	ENTREC

Appendix 5. Search strategy described in terms of the components of the STARLITE framework.

STARLITE component	Search strategy description
Sampling strategy	Aimed to identify all relevant studies on the topic
Type of studies	Search using a comprehensive list of search terms related to qualitative research methodology
Approaches other than bibliographic searches (i.e. grey literature	*Thesis reports* ProQuest dissertation and theses database (includes index of theses) was searched for theses that met the inclusion criteria. *Citation searching* Web of Science citation search feature used to identify publications that have since referenced papers included in the review. *Reference list searching* Searched reference lists of all studies included in the review and of selected other reviews (n=23), comment pieces or other relevant documents that did not meet the inclusion criteria. *Hand searching journals* The last 3 years contents lists of the ALS specific journal: ‘Amyotrophic Lateral sclerosis & Frontotemporal Degeneration’ were searched

	Hand searching conference proceedings Search the last 3 years (2018, 2019 and 2020) plenary, poster and work in progress abstracts for the annual International MND Symposium. *Individuals/Organisations* Key professional networks, specialist groups, HCPs and the authors of included studies were contact to request any studies or grey literature not captured in the searches.
Range of dates (start date - end date)	With limited studies identified on the topic and no further rationale identified to limit the dates of the search, no date limits were set on the search.
Limits	Limited to studies reported in English due to lack of resources to translate papers
Inclusions and exclusions	*Inclusion criteria:* 1. pwMND variant, their caregivers (defined as family members or non-paid carers) or HCP's 2. Participants in studies over 18 years old 3. Studies focus on pwMND decision making about commencing invasive interventions 4. Employs any qualitative methodology 5. Peer reviewed publication 6. Published in English language
Terms used	The following concepts and their synonyms 'MND', 'decision making or named intervention/care decision' and 'qualitative

	study' were combined using the Boolean term 'AND'. Full Medline See appendix ## for full Medline search.
Electronic sources	Searches were adapted for Ovid MEDLINE, Embase via OVID, CINAHL via EBSCO, PsycINFO via OVID, Cochrane (Wiley) Library database of systematic reviews.

Appendix 6. Conceptualisation of the survey research question.

Concept: Decision making process

Dimensions	Sub-dimensions	Further subdimensions
Demographic information	Professional role	
	MND experience	
	Role in the MND MDT	
	Age/gender	
Initiating discussions about gastrostomy	Timing of introducing gastrostomy	
	Triggers for introducing gastrostomy	
Decision making process	Choice presentation	
	Communicating about the current health threat	Progression of disease/symptoms

		Nutritional
		Respiratory
		QoL
		Prognosis
	Information exchange	Procedure
		Home enteral feeding
		Care requirements
		Impact on care needs
		Impact on caregivers
	Benefits and risks of gastrostomy and enteral feeding	
	Outcomes with/without gastrostomy	Prognosis
		Function
		Quality of life
	Goals of gastrostomy placement	

	Timing	Respiratory failure
		Frailty
		Before/after symptom onset
	Withdrawal of enteral feeding	
	Sources of information	
	HCP recommendations	
	Time spent discussing gastrostomy	
	Autonomy	HCP/caregiver pressure

Concept: multidisciplinary team organisation of decision support

Dimensions	Sub-dimensions	Further subdimensions
MDT decision support	Professional roles involved	
	Role in decision making	
	Guidance	
Coordination	Policies and guidance	
	Care pathways	
	Communication	

Referral	Time to placement	
HCP influence on decision making process	Pace of delivery	Length of interactions
		Number of interactions
	Roles/responsibilities in decision making process	
	Knowledge relevant to decision	Training

Appendix 7. Multiple case study protocol.

Protocol for multiple case study exploring how people with motor neuron disease (MND) are supported by their clinical teams to make decisions about gastrostomy feeding tube

Chief investigator: Sean White

Supervisors: Professor Christopher McDermott (lead supervisor), Professor Alicia O'Cathain and Dr Vanessa Halliday

IRAS project ID: 317939

Sheffield Teaching Hospitals Clinical Research and Innovation Office number: 22400

Table of Contents

Background

1.1 Gastrostomy placement in MND

The clinical decision to offer a gastrostomy tube to a pwMND is complicated by the unpredictable disease progression and the number of clinical indications for initiating the intervention (Van Eenennaam *et al.,* 2021). It remains unclear what impact gastrostomy placement has on survival in pwMND, with studies presenting mixed results (Castanheira *et al.,* 2021). A recent prospective study found no difference in survival when comparing pwMND who accepted or refused gastrostomy placement, where the intervention was deemed clinically indicated (Vergonjeanne *et al.,* 2020). The timing of gastrostomy placement influences post-placement survival, with later placement and increased weight loss being associated with shorter survival (ProGas Group 2015; Castanheira *et al.,* 2021). The lack of evidence about clinical and quality of life outcomes presents a challenge to HCPs communicating the benefits and risks of gastrostomy placement to pwMND who have an unpredictable disease course.

Once a decision is made about gastrostomy tube placement, pwMND are then required to decide when they wish to place the tube. A disconnect has been reported in the qualitative literature between when pwMND perceive there is a need for intervention and when supporting HCPs feel the intervention is indicated (Greenaway *et al.,* 2015). HCP recommendations are made in response to observing the decline of clinical and nutritional outcome measures and respiratory failure (Van Eenennaam *et al.,* 2021). In contrast, pwMND have been reported to be guided by how they are experiencing the disease, sometimes willing to tolerate symptom burden rather than start an intervention (Stavroulakis *et al.,* 2014; Greenaway *et al.,* 2015). There is little evidence or clinical guidance available to guide when the optimal time to have a gastrostomy tube, leading to varied HCP practice about when the option is presented and recommended by HCPs (Van Eenennaam *et al.,* 2021). Clinical markers need to be integrated into HCP recommendations about gastrostomy timing, while considering the psychosocial factors that influence when pwMND choose to have a gastrostomy tube placed.

1.2 General decision-making in MND care

Published reviews have described what the indications for invasive interventions such as gastrostomy and ventilation are, how they are initiated and the evidence with regards outcomes (Barc and Kuźma-Kozakiewicz 2020). While this objective data about the benefits, risks and outcomes of an intervention are important to inform HCPs clinical reasoning and pwMND decisions, it does not address the psychosocial and contextual factors identified to influence decision making in MND care (Hogden 2012a; Hogden 2012b; Hogden 2013; Foley and Hynes 2018). pwMND are constantly re-evaluating their views and preferences about an intervention, in response to and acceptance of the continual changes in their physical condition (King *et al.,* 2009). The readiness of pwMND to engage in discussions about interventions influences the timing of information exchange (Hogden *et al.,* 2012a). Healthcare professionals have reported how pwMND who appear to be in denial about their diagnosis delay discussions about future interventions (Hogden *et al.,* 2012b). While denial has been shown to be an effective coping strategy to reduce psychological distress in pwMND, information seeking behaviour was associated with a better quality of life (Matuz *et al.,* 2010). It is important that HCPs understand how and why pwMND respond to intervention options being introduced, and how to engage them in discussions about interventions, while continuing to respect their preferences for information and coping mechanisms. A flexible and individualised approach to supporting decision-making is required, taking into account both intervention information and the preferences of the pwMND involvement in the decision.

1.3 Making decisions about gastrostomy placement in MND

A recent meta-ethnography identified a wide range of factors that pwMND reported influenced the decisions they made about gastrostomy placement including influence of other people; perceptions of choice and control; the influence of dysphagia on decisions made; the role of personal values; timing of gastrostomy placement; impact of emotions and feelings and the place of gastrostomy within the wider context of MND (Lisiecka *et al.,* 2021). pwMND and their families have little time to psychologically and physically adapt to the rapid decline in function, creating a barrier to engaging in decision-making activities (Martin *et al.,* 2014; Greenaway *et al.,* 2015). Guidance recommends healthcare professionals have early discussions about gastrostomy placement with pwMND (NICE 2019). However, pwMND often report not wishing to consider the future and only making a decision when they perceive there is a physical need (Pols and Limburg 2016). This creates a challenging juxtaposition between HCPs wishing to introduce interventions and pwMND wishing to delay discussions until they are ready (Greenaway *et al.,* 2015). pwMND do not make decisions by themselves, and are heavily influenced by caregivers and the support they receive from HCPs.

Caregivers have a strong influence over the decision-making process; the decisions made; and when decisions are made. Caregivers play a supportive role during decision-making, including screening and sourcing information; providing emotional support; and enabling them to access expert advice (Hogden *et al.,* 2013). However, challenges have been highlighted when conflict exists between promoting the pwMND voice and the caregivers' own views about what is in the best interests of the pwMND (Foley and Hynes 2018). pwMND make decisions based on not just the impact on themselves but also the perceived burden the interventions will have on caregivers (Foley and Hynes 2018). With pwMND and their caregivers often making the decision to have a gastrostomy together, any differences in opinion may present a threat to the pwMND autonomy if the views of pwMND and their caregivers are not aligned.

HCPs play an integral role during the decision-making process in terms of being gatekeepers to interventions and the support they provide to pwMND. pwMND and their families identify HCPs as a credible source of expert information and support in relation to decision making (Greenaway *et al.,* 2015). While pwMND prioritise the information they receive from HCPs, they are also influenced by the approach taken by HCPs towards having these difficult discussions (Paynter *et al.,* 2020). HCPs use a range of health outcomes to guide their clinical reasoning but are not always consistent in what cut-offs they may use or when they introduce interventions to pwMND (Stavroulakis *et al.,* 2013; Van Eenennaam *et al.,* 2021). The lack of evidence-based guidance on these important issues when communicating information about interventions, opens the door to varied ‘best practice’. Varied HCP knowledge and attitudes towards gastrostomy placement in MND care becomes more of an issue when multiple HCPs are having discussions with a single pwMND about gastrostomy placement. There is a need to understand how the multiple HCPs having these discussions with a pwMND deliver a coherent and consistent message to pwMND about the choice to have a gastrostomy placed.

The HCPs knowledge, previous clinical experience, and values are amongst the factors that influence their clinical reasoning when offering interventions and framing discussions about options available (see figure 1) (Bekker 2021). Shared decisions require HCPs and patients to share and discuss their understanding of the disease, the proposed interventions, and of the preferences for the options available. Both HCPs and patients undergo a process of reasoning about intervention options that are influenced by the context in which decisions are being made. To be able to effectively support patient decision making, HCPs require an understanding of both their own and the patient's understanding and reasoning.

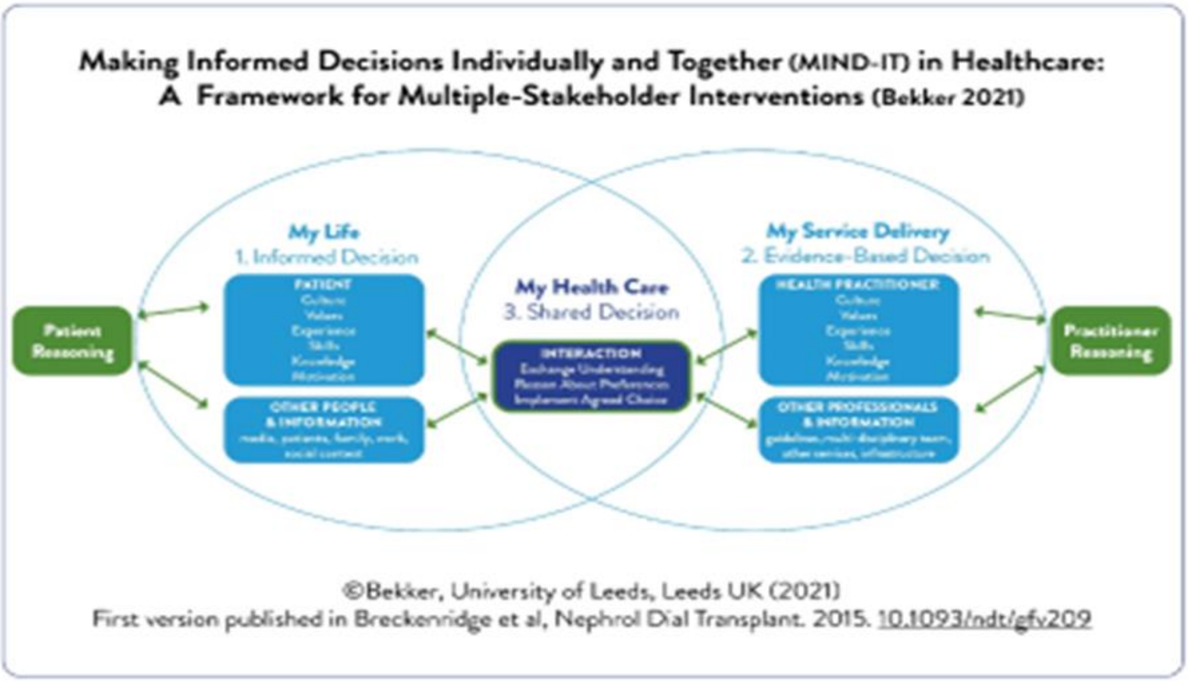


Figure 1. **Making Informed Decisions Individually and Together (MIND-IT) framework**

Within MND care the involvement of multiple HCP roles, the organisation and resources of healthcare services and local and national guidance, are likely to influence when interventions are offered and how the options of commencing, deferring or declining interventions are framed. Further research is required to understand how HCPs clinical reasoning, individually and as teams, informs how and when options are presented and to what extent patient understanding, values and reasoning are incorporated into decisions made.

1.4 MND MDT: a unit of decision support

Following diagnosis, pwMND are supported by a range of HCP roles acting within the MND MDT, each with different expertise and responsibilities. Evidence of a survival benefit in those pwMND supported by a MDT with MND experience, has informed how MND services are now organised and delivered (Aridegbe *et al.,* 2013). The mechanisms by which MND MDT's infer a survival benefit remain unclear. One hypothesis could be the expert decision support and the timely initiation of supportive interventions such as gastrostomy placement. Some complex decisions, such as about gastrostomy placement, require input and support from multiple HCP roles within the MDT (Van Eenennaam *et al.,* 2021).

Research has highlighted some of the challenges of delivering coordinated decision support in MND MDT care. HCPs are often not all situated under one roof, making it more complicated to coordinate and communicate across all the stakeholders involved within and outside the formal neurology clinic MDT (Hogden *et al.,* 2017). HCPs taking part in focus groups reported power imbalances between professions that may inhibit communication and MDT communication was identified as key to coordinating support given to patients (Sohi *et al.,* 2015). A rapid ethnography identified how different HCP roles working within primary care teams still work in silos, with physicians having little communication in busy clinics with the other staff (Chesluk *et al.,* 2010). With decision support being delivered by multiple professionals and teams across different healthcare settings, the team dynamic and lines of effective communication are likely to impact on the experience the pwMND have of decision making. Issues such as care coordination are not comprehensively addressed in international MND management guidelines (Janssens *et al.,* 2016). HCPs and pwMND have reported mixed messages being delivered about the need for interventions potentially contributing to pwMND decisional conflict (Greenaway *et al.,* 2015; Chapman *et al.,* 2021). Differences in opinion were found between medical and allied HCPs when asked about the timing of gastrostomy discussions, policy compliance, and how informed pwMND and caregivers are about gastrostomy placement (Ruffell *et al.,* 2013). The is a paucity of research that has focused how the MND MDT functions as a unit of decision support, including what the roles/responsibilities of each profession involved are, coordinating the sharing of the information pertinent to the decision and how interactions with the MDT influences how and when decisions are made. Improving the way the MDT supports the decision making requires an understanding of these contextual complexities from the perspectives of all the main stakeholders, including the pwMND, caregivers and HCPs. An action research project found that professions had different perceptions about their role in sharing information about treatment options and guiding decision making (Sohi *et al.,* 2015). While communication was reported as key for involving patients in decision making, hierarchical challenges included power imbalances between different professions (Sohi *et al.,* 2015). The MND MDT, as a unit of decision support, has the complex task of coordinating communication about disease progression and intervention, while accounting for the facilitators and barriers that have been reported to affect decision-making. Further research is required to understand how HCPs coordinate with each other to have a consistent approach to navigating the many decision-making challenges and enable pwMND to make an informed and timely decision about gastrostomy placement.

1.5 Theoretical framework guiding the study

The CI has completed two previous pieces of research on this topic area. The findings of this research have informed the development of the research question, objectives, data collection and analysis.

Firstly, the CI used a qualitative evidence synthesis (QES) to systematically collate and synthesise the findings from the qualitative literature that focused on the perspectives of pwMND, caregivers and HCPs on the process of making decisions about invasive interventions, namely ventilation and gastrostomy. The outcome of the review were four analytical themes:

Making values-based judgements: pwMND create positive or negative perceptions about how the intervention may impact on their daily routines or quality of life and have differences in how they value the prospect of commencing a life prolonging treatment.

Sharing the burden: External stakeholders influence the decision-making process and what decisions are made. HCPs and caregivers can both support and present barriers to decision making, through the sourcing/providing of information, and how their priorities and values are projected onto the decision-making process. Where present, the perceived impact on caregivers, is an important consideration whether or not to accept an intervention.

Control/autonomy: All stakeholders support and seek to protect the pwMND preference for autonomous decision making. However, there are scenarios where autonomy can potentially be threatened by disease progression, and perceived pressure to make a specific decision by others e.g. caregivers and HCPs. pwMND exert agency over a preference to focus on the present which can be a barrier to planning or discussing future interventions.

Tipping the balance: Symptom progression and HCP recommendations are key drivers for when interventions are accepted. All stakeholders find it a challenge to judge the optimal time to commence interventions, with pwMND wanting concrete guidance from their HCPs

The review findings describe how HCP and pwMND knowledge, perceptions, experiences and preferences, related to the disease and interventions, interact throughout the decision-making process, from the point of an intervention being indicated through to the acting on decisions made. HCPs clinical judgement and reasoning is informed by objective markers of functional decline, the information that pwMND share with them about their experience of the disease and views about interventions, and their prior experience of other pwMND outcomes. The outcome of the clinical reasoning influences when interventions are first discussed with pwMND, and how information about the interventions are framed. The pwMND reasoning is informed/influenced by the cognitive and emotional representations of the disease and proposed interventions, with comparisons about perceptions about quality of life with interventions being compared to the current self-schemata, guiding coping strategies and action planning. The reasoning and interactions of HCPs and pwMND is repeatedly updated in response to further functional decline. The next phase of research aims to understand how HCPs can best support pwMND at the key decision points: introducing the option; informing about options; timing of gastrostomy placement. Though maintaining a focus on the role that HCPs play in the decision making process, through understanding how pwMND respond to, process and act on the support they receive from HCPs.

The findings from the QES and the concepts investigated in a recently completed survey of HCPs currently being analysed have informed the design of this study and the initial framework which will be iteratively developed in response to the emerging data from the case studies.

1.6 Research question

- How do HCPs, operating within MDTs, support pwMND to make decisions about gastrostomy placement?

1.7 Aim

To understand how HCPs, as individuals and as part of the wider MDT, support pwMND to make decisions about gastrostomy placement

1.8 Objectives

- To understand the roles of different HCP professions and teams involved in supporting pwMND decision making
- Understand how HCPs working within an MDT structure communicate with each other about pwMND considering gastrostomy placement
- Understand what informs HCPs reasoning to introduce and offer gastrostomy placement to pwMND
- Understand what informs pwMND reasoning to accept, defer, or decline gastrostomy placement
- Identify what information HCPs share with pwMND about the option to continue without and to choose gastrostomy
- Identify what information pwMND share with HCPs about their perceptions, understanding, and goals in relation to the decision to have a gastrostomy placed or not
- Understand how HCPs, individually or as part of the MDT, influence pwMND response to being presented with the option and information about gastrostomy placement
- Understand what enables or hinders pwMND to act on their decision to accept gastrostomy (e.g. for those pwMND who have accepted they will have a gastrostomy but choose to defer the timing of placement)
- To develop evidence based recommendations to guide HCPs, individually and as MDTs, in how they can inform pwMND about their option to have (or not) a gastrostomy placed

Methods

2.1 Methodology

Case study design provides a method that can meet the objectives of this research providing an approach that enables the phenomenon and context to be the focus of qualitative enquiry. Yin (2014) defined case studies as a 'method that investigates a contemporary phenomenon (the "case") in depth and within its real-world context, when the boundaries between phenomenon and context may not be clearly evident' (Yin 2014; pg 15). For the proposed study, the decision support that HCPs deliver to pwMND considering gastrostomy placement, as individuals and an MDT, represents the phenomenon of interest. Case study design involves observing the phenomenon from multiple perspectives, using different research methods to generate a rich and thick description of what is actually happening in 'real-world' practice (Crispin *et al.,* 2017). Including multiple cases enables a broader qualitative enquiry into the phenomenon, in similar contexts but different settings, allowing for the search for atypical cases or explanations. Triangulation of a range of data sources allows a detailed understanding of a phenomenon and particularly when multiple cases are studied, theoretical generalisation (Anthony and Jack 2009, Crowe *et al.,* 2011; Yin 2014; Stake 2006; Walshe 2011).

2.2 Case definitions

- Case definition: 'the MND multidisciplinary team operating across the clinic and community setting that supported pwMND to make decisions about gastrostomy'
- pwMND embedded cases: The longitudinal process of decision making by an individual pwMND, including their interactions with members of the MND MDT
- Boundaries: Includes pwMND, caregivers, HCPs and the healthcare organisations involved in making decisions about gastrostomy placement.

2.3 Case study site selection

2.3.1 Rationale for study site selection

One priority when selecting case study sites is that the researcher has access to observe and record data about the phenomenon of interest on each site (Stake 2006 pg 24-27). Though there are differing models of MND care provision, most pwMND will receive care from multiple professional roles, including discussions about gastrostomy where indicated. The care of pwMND usually includes the medical care delivered by a medical consultant, who could be a neurology or palliative care consultant in addition to a range of nursing and allied health professionals (AHPs) within and outside (i.e. community or out-reach) the MND clinic setting. Though not universal, many pwMND are supported by a MND nurse or coordinator. However, there are differing models of MND MDT care structure and organisation in the UK. A number of different models of MND care organisation exist in the UK (MND Association Information Sheet 3A):

1. Tertiary MND Care Centres. These are specialist MND clinics that include different members of the MDT that pwMND attend for routine follow up after being diagnosed with MND. The care centres are often centred around a central MND clinic that is hosted in either an NHS hospital or charity funded hospice, including availability of a range of clinical specialities. These centres provide tertiary care, supporting pwMND over a wide geographical area, outside of the boundaries of care provided locally. However, pwMND attending tertiary MND care centres will also receive support from HCPs in their local area that are employed by other organisations.
2. Non-tertiary MND clinic. These are specialist MND Care clinic, that include a range of different clinical specialties relevant to the care of people with MND. They differ from tertiary care centres in that their catchment area for pwMND remains local to the clinic.
3. MND Networks. There are 5 MND Networks in the UK part funded by the MND Association, that provide multiple MND clinics over a region, particularly where the MND population is sparsely populated (e.g. South Wales, South West Peninsula and Northern Ireland).

2.3.2 Approaching potential case study site

Firstly, a scoping exercise identified the MND services expressing an interest in being a case study site. A number of approaches have been taken to approach HCPs within MND services about the opportunity for their local MND clinic to be the centre for one of the case studies:

1. The CI has completed a survey of UK HCPs practice and attitudes towards supporting pwMND making decisions about gastrostomy placement, including asking HCPs who may be interested in participating in the case study to contact the CI. The HCPs

replying to this request will be contacted to explore whether their local MND service would be a feasible case study site.

2. The contact details available on the MND Association website were used to contact the clinical leads and/or MND coordinators/nurses of MND centres and networks.
3. The CI sent invitational emails to his established clinical network

The scoping email gave a brief description of the study and what being a case study site would involve for the PI and wider MDT. Those receiving the scoping emails were asked to respond if they wished to express an interest in becoming a case study site and would be willing to answer some screening questions to inform study site selection.

2.3.3 Selecting case study sites

The HCPs responding to the invitational emails were sent a subsequent screening email which aimed to gain more information about the model of MND care delivered locally and the feasibility of them becoming a case study site. The CI also offered to have further discussions via video or telephone call to answer any clarification questions the HCP replying to the invitations may have. The screening process included asking for the following information:

- Whether the local MND service is neurology or palliative care lead
- Whether the service is a tertiary or non-tertiary service
- Whether the service is part of a MND care centre, network or other model
- The name of the organisation that hosts the MND clinic
- The HCP roles employed by the organisation where the MND clinic takes place
- The HCPs roles that support pwMND outside the organisation where the MND clinic takes place and where they are located (geographically and in terms of the organisation that employs them).
- The geographical spread of where pwMND who attend the clinic are located.
- Identification of a gatekeeper HCP who would be willing and able to fulfil the role of PI

The case studies would be purposively sampled based on the following criteria:

- The model of MND care provided i.e. network, care centre or local service; tertiary or non-tertiary service; neurology or palliative care lead service, aiming for a variation in models.
- A gatekeeper HCP who could fulfil the role of PI on the case study site
- Engagement of the wider MDT to participate in the case study
- Commitment to recruit 3-5 pwMND to the study

2.4 Preparation for case study data collection

The success of the study on each site depends on recruiting 3-5 pwMND and a broad range of HCPs involved in discussions about gastrostomy placement. Prior to engaging in the fieldwork, the CI will require a good understanding about the local MND MDT, including who the relevant HCPs are, where they are located, how, where and when pwMND engage with the MND MDT. The CI will also need to be familiar with how local MND clinics operate, to understand how to ensure everyone is aware of the research taking place, to identify and optimise opportunities for non-participant observations, and to minimise any disruption that may be caused by the CI’s presence.

The CI will firstly arrange to meet with the site PI, to generate a good understanding of the issues above. Following this, the CI will offer to meet virtually with any HCPs who are interested in learning more about the study and considering taking part, including the delivery of an online presentation about the study, recruitment and data collection. This session will introduce the aims of the study, what participating will involve, process of pwMND recruitment and consent, and be an opportunity to better understand the organisation and membership of services, and aid planning for data collection. The session will be recorded and with permission from those attending, shared with other members of the local MDT not able to attend. The CI, in collaboration with the site PI, will develop a summary of the local MND service and map out how pwMND interacts with the MDT for each site, with a specific focus on discussions about gastrostomy placement.

If a Welsh MND service is selected to be a case study, a local translation service will be used to translate the participant facing documentation into Welsh including the invitational emails, study leaflets, posters, participant information sheets, and consent forms. Verbal translators would be asked to join people with MND or caregiver interviews where the individual does not speak English.

2.5 Inclusion and exclusion criteria

2.5.1 pwMND participants

Inclusion criteria

- Confirmed diagnosis with MND
- The pwMND has had discussion(s) with HCPs about the option to have a gastrostomy placed
- The pwMND is considering having a gastrostomy or has made a decision about gastrostomy placement (they may not have had their gastrostomy placed yet)
- Attends the MND clinic within the organisation of the case study site
- Are able to communicate (i.e. in the interviews) verbally or by using a communication aid
- Can speak and read in English or Welsh

Exclusion criteria

- If the person with MND has had a gastrostomy placed, it was placed less than 1 month ago

2.5.2 Caregivers participants

Inclusion criteria

- Non-paid informal caregiver of a pwMND recruited to the study
- pwMND has consented to the caregiver being approached to be a participant
- Can speak and read in English or Welsh

Exclusion criteria

- The caregiver is paid to be carer e.g. the caregivers is employed to provide care but has no other personal relationship with the pwMND

2.5.3 HCP participants

Inclusion criteria

- Provides any decision support about gastrostomy placement to pwMND including introducing the option and sharing information.
- Is a qualified healthcare professional
- Supports pwMND that attend the case study site MND clinic

2.6 Sample size

2.6.1 pwMND and caregivers

3-5 pwMND attending MND clinics on each case study site and their caregivers (where present) will be recruited on each case study site (across all three case study sites 9-15 pwMND and up to 15 caregivers (where present) will be recruited).

Further pwMND and caregivers will be observed during their interactions with HCPs by the CI.

3-5 pwMND and their caregivers will be invited to participate in workshops to inform the development of HCP guidance.

2.6.2 HCP participants

Up to six HCPs who discuss gastrostomy placement with pwMND on each case study site will be recruited to participate in the semi-structured interviews (across all three case study sites up to 18 HCPs will be recruited).

Up to eight HCPs with representation from different HCP professional roles, will participate in a single multidisciplinary focus group on each case study site (across all three case study sites up to 24 HCPs will be recruited).

Further HCPs may be observed during their interactions with other HCPs, pwMND or caregivers.

Up to 8 HCPs will be invited to participate in workshops to inform the development of HCP guidance.

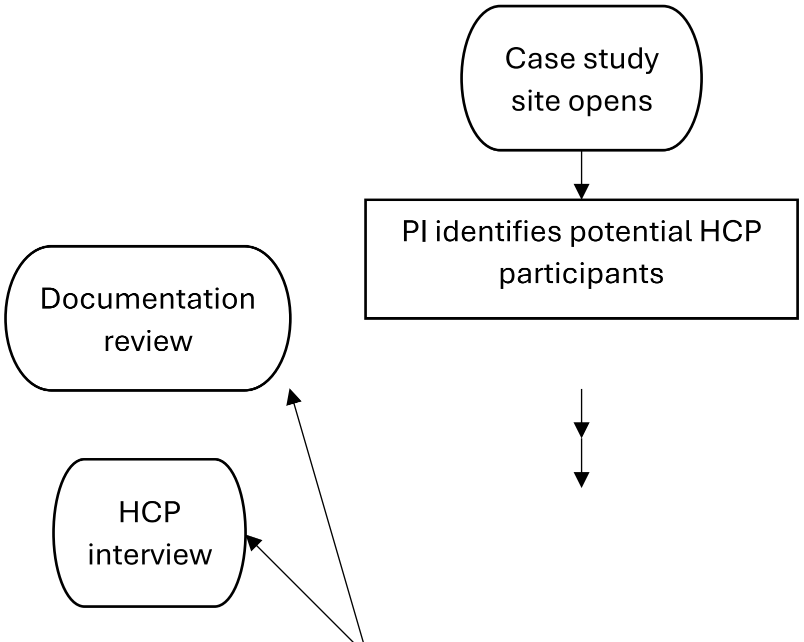
2.7 Sampling

A convenience sample of 3-5 pwMND (and their caregivers where present) will be recruited on each case study site to take part in a single semi-structured interview. pwMND will also consent to a review of their medical record.

HCPs will be purposively sampled to participate in the focus groups, non-participant observations and semi-structured interviews, to ensure representation from the range of professions involved in supporting pwMND with the decision to have a gastrostomy placed within the local MDT including HCPs that work in the acute, clinic and community settings. The sampling frame for each site will be informed by discussions with the PI, information obtained during the online case study preparation sessions with the MDT and what is learnt about the local site during non-participant observations. On most sites the sampling frame will include clinicians (most often neurologist and/or palliative care clinician), speech and language therapist, dietitian and MND specialist nurse or coordinator. However, when on site other members of the MDT who are identified to have discussions about gastrostomy placement will also be approached, including gastroenterologists, nutrition teams, nutrition nurses, respiratory teams and psychologists. The sampling strategy will include recruiting HCPs from the different organisations, teams and healthcare settings.

2.8 Approach, Recruitment and consent

A summary of the approach to recruitment is described in the recruitment flow diagram below (figure 2):



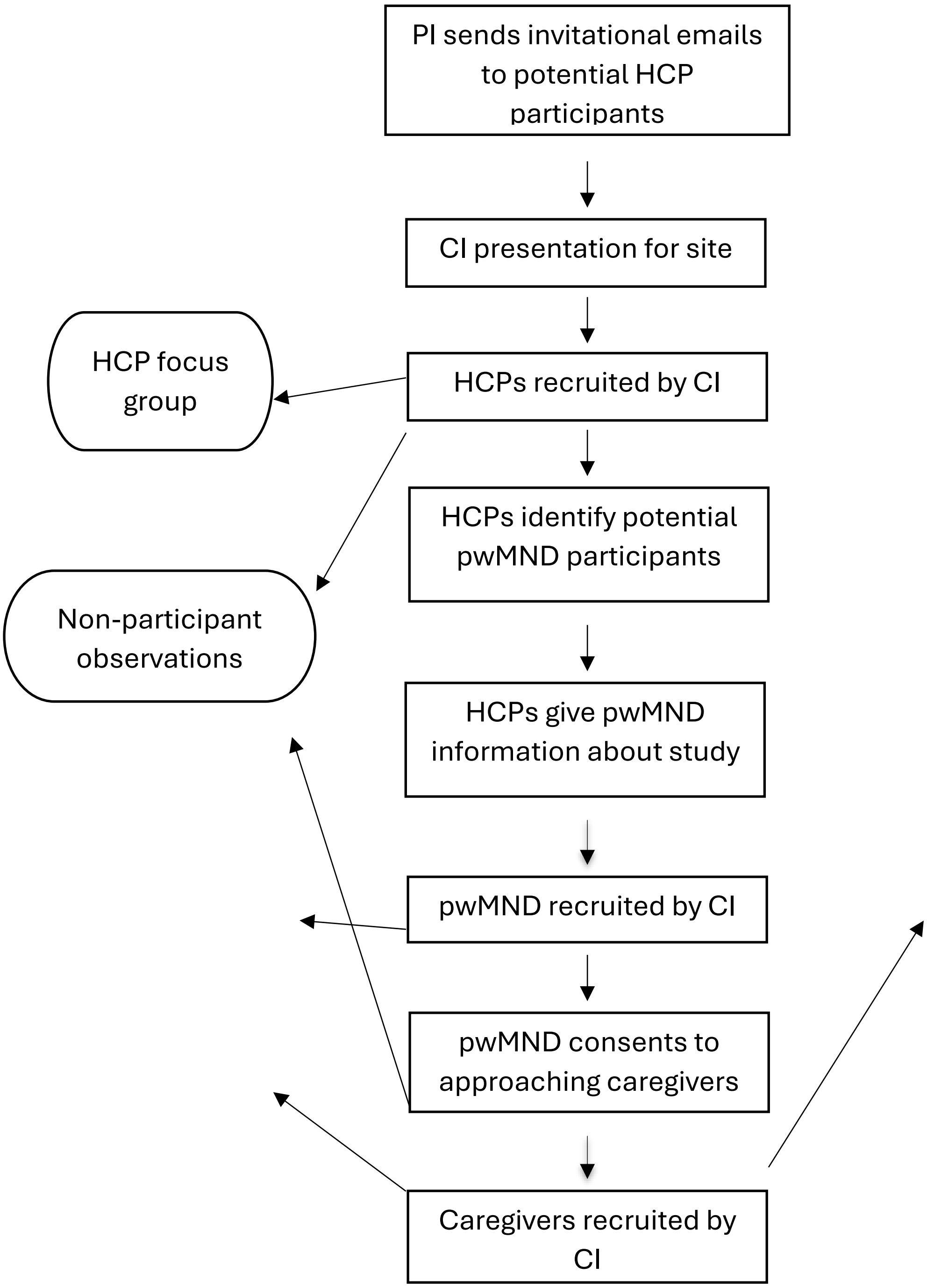


Figure 2. Recruitment flow diagram (does not represent order of data collection)

2.8.1 Recruitment of a Principal Investigator (PI)

Recruitment will begin with that of a gatekeeper (or key informant) HCP from within each MND clinic who will also be designated as the primary investigator (PI) for the case study site (Pope and Allen 2020). The PI will be identified through the scoping and screening exercise to identify the case study sites. This individual is most likely to be a MND specialist nurse or coordinator, who has a good knowledge of the case sites MND caseload and contacts with the local MND team and service (Millen 2000). The PI will have up-to-date Good Clinical Practice (GCP) training and have a good awareness of the study protocol including the inclusion/exclusion criteria and approaches to recruitment.

2.8.2 Recruitment of HCPs

Interviews, focus group and workshop

HCPs will have the opportunity to participate in one, some or all of the research activities including non-participant observations, interview, and focus group. The PI will have responsibility for forwarding invitations, HCP participant information sheets (PIS) and consent forms, by post or email, to all HCPs who are known to have discussions with pwMND about gastrostomy placement that attend the MND centre/clinic, including those that are located within and outside the local site organisation. The CI PPI panel and the survey completed by the CI identified that many different HCPs discuss gastrostomy with pwMND including those that attend the MND clinic and those that provide support in the community. The PI will follow up by contacting the HCPs sent the invite by phone or email one week later to discuss the study and answer any questions they may have. HCPs interested in taking part can either give verbal consent for the PI to pass on their contact details to the CI or by contacting the CI directly themselves. The CI will also arrange to put up posters advertising the opportunity for HCPs to participate in the study in areas where local HCPs are situated such as clinics, staff rooms or offices. When contacted, the CI will arrange a time to discuss the study with the potential participant and seek to obtain informed consent. HCPs belonging to organisations where the CI has local research department approval for the study, will be asked to forward invitations to other MDT colleagues who they are aware are involved in discussions with pwMND about gastrostomy placement that attend the case study site MND clinic. All HCPs forwarding on the information to other HCPs will be on the delegation log. All potential participants will be allowed at least 24 hours to read the PIS and given the opportunity to ask questions. All HCPs who participate in the study will also be offered the opportunity to consent to being approached to take part in the post case study workshops.

When consenting to take part in the research HCPs will be asked for permission to approach them to take part in the other research activities taking place i.e. interviews, focus groups or workshop.

Non-participant observations

In relation to non-participant observations, the CI will forward the PIS to all HCPs that are known to be present in the setting in which observations are planned. The PIS and consent forms will be forwarded by the PI or other HCP from the case study site. For observations planned in the clinic setting, all HCPs due to attend the clinic will be sent the PIS and the CI will seek to obtain consent in advance. The CI will place posters up in the clinic with a picture of the CI and inform those in the clinic that observations are taking place for research purposes. The CI will have information to give to people and consent forms to complete. For observations in meetings or HCP offices, all HCPs known to be present will be sent the PIS and asked to provide consent. At the start of meetings the PI or HCP chairing the meeting, will advise those present that observations are taking place. All HCPs will need to have provided consent prior to a meeting being observed by the CI. Where consent is declined or not obtained the meeting will not be observed.

2.8.3 Recruitment of pwMND

Interviews, medical note review and workshop

The PI and local HCPs will be asked to identify pwMND who meet the inclusion criteria. pwMND who meet these criteria will be provided with some brief information about the study and the PIS. Potential participants could be approached to take part in the study in face-to-face clinic, via video or phone calls or via email. pwMND who consent to being observed, will also have the option to give consent to the CI approaching them to take part in the interviews and review of their medical notes. Also, during the periods of non-participant observation e.g. during MND clinics, the CI will be available to discuss the study with any pwMND who have had the study introduced to them by a HCP and if interested, arrange a time to gain informed consent. The CI/PI will place posters in the clinic waiting room about the opportunity for pwMND and caregivers to participate in the study, including the CI contact details. The local MND Association support groups will be asked to advertise the opportunity to participate in the study using their communication channels or support group meetings. With consent from the local branch of the MND Association, the CI will offer to attend support groups online or face-to-face to discuss the study.

Any pwMND or caregiver interested in taking part can either give permission for their contact details to be forwarded to the CI or they can use the contact details included in other recruitment adverts to make contact independently. pwMND participants are able to take part in the interview, medical note review and workshop if their caregiver were to decline consent. The caregiver may remain present in the interview to assist the pwMND with communication if required, but only data provided by the pwMND will be recorded. The CI will then arrange a time to screen them for eligibility, discuss the study with the potential participant(s) and arrange a time to obtain informed consent. In most scenarios potential participants will be allowed at least 24 hours to read the PIS and given the opportunity to ask questions. Consent will be sought to approach pwMND and caregivers to take part in the post case study workshops. However, participation in the workshop is optional and declining involvement does not prevent participants from being interviewed or having their medical notes reviewed.

With consent from the participant, the researcher will write to the GP of all pwMND recruited to participate in the study using a standard letter template.

Non-participant observations

The CI will be observing interactions between HCPs and pwMND. These may occur in any setting including clinic, hospital wards, the patient’s own home, or via telephone/video call. The approach to recruitment will involve the HCP identifying pwMND whom they are aware will be attending planned interactions with a HCP from their records e.g. clinic lists. The HCP will send potential participant(s) (i.e. pwMND and caregivers if present) information about the study, a participant information sheet and consent form in relation to the CI to observing their interaction with their HCP at least 24 hours in advance. Potential participants will have time to read the PIS and ask questions of the PI or CI. The potential participant(s) will be informed that the CI will be present and observing the interaction for research purposes.

The HCP and/or the pwMND may not be aware in advance of a planned interaction that a discussion about gastrostomy placement will take place during the planned interaction. This will include the scenario when gastrostomy placement is being discussed with the pwMND for the first time. Discussions about gastrostomy can be spontaneous rather than pre-planned as a result of the HCPs assessment or the pwMND or caregiver raising the issue. For this reason, the CI will need to observe multiple interactions between HCPs and pwMND including where gastrostomy is not discussed. It could potentially cause the pwMND anxiety and distress if they were to be informed in advance (i.e. by sending them information about the study) that the HCP may be discussing gastrostomy placement with them. A previous review described scenarios where not gaining full informed consent may be legitimised including where the consent process itself could lead to increased stress or anxiety (Rebers *et al.,* 2016). Even if gastrostomy has been discussed before it may still cause the pwMND anxiety if they are made aware in advance that the planned interaction will include discussion about gastrostomy placement. However, observing the discussions between HCPs and pwMND and caregivers remains important to fully understanding how HCPs support people making these decisions. The issues explained above make seeking full informed consent to observe interactions between HCPs and pwMND/caregivers prior to the interaction occurring, a challenge.

This issue was discussed with the CI's PPI panel on 30/8/22. The panel agreed it would not be appropriate to send pwMND information about the intention to observe their interaction with a HCP, that included reference to studying how decisions are made about gastrostomy placement. If the pwMND had not discussed gastrostomy before or the issue was contentious for them, receiving information that suggests their HCP will be discussing gastrostomy with them the panel agreed this could raise their anxiety prior to the interaction and may interfere with clinical practice. Though the panel recognised that not disclosing all information about the study was not ideal, they accepted that the approach described below would ensure pwMND and caregivers had the opportunity to provide full explicit consent for the observational data to be used in the study and the lack of full disclosure was appropriate and proportionate to avoid unnecessary distress to the participants.

To avoid causing the pwMND any distress the information sent to them will not include that the study is focused on the decision to have a gastrostomy tube placed. Instead, the information will explain that the research is aiming to understand how HCPs share information with pwMND without specifically mentioning the focus on gastrostomy decision making (see the justification for this in the consent section 2.8.5). The pwMND will have the opportunity to discuss the study further with the CI prior to the interaction taking place. Where possible the researcher will obtain informed consent prior to when the pwMND attends the interaction to avoid interrupting normal practice in the clinical setting. If this is not possible the pwMND can bring a signed copy of the consent form to the interaction or the CI will have copies available to complete on the day of the interaction.

2.8.4 Recruitment of caregivers

All pwMND recruited to participate in the interviews will be also asked for permission to approach their caregiver, where present, to participate in interviews. Where this consent is provided the CI will provide the caregiver with the PIS, give time for them to read and understand the information, and provide opportunity to ask any questions. Caregivers will ideally be interviewed alone but where requested interviews can be in the presence of the pwMND.

The approach to recruiting caregivers to be observed will be the same as it is for pwMND participants. Where the pwMND does not give consent for their caregivers to be approached to be observed during interactions with their HCP, or where the caregiver does not give consent to be observed, then the interaction will not be observed by the CI. This will mean that even if the pwMND consents themselves to participate the observation would still not occur. Consent is required from all participants present during an interaction (including the pwMND, caregivers and HCP) for the observation to take place. Where the pwMND does consent to approaching the caregiver, the caregiver will be provided with the pre-observation PIS which does not explicitly refer to the focus of the study being on decision making about gastrostomy placement. Where discussions about gastrostomy placement do take place, the CI will arrange a time after the interaction to seek full informed consent including informing about the focus on gastrostomy decision making.

2.8.5 Procedures for obtaining informed consent

Documenting consent

Where it is feasible face-to-face written consent will be obtained from participants. However where this is not possible other methods of obtaining informed consent will be used. For those participants (pwMND/caregivers/HCPs) where it is not possible to obtain face-to-face consent, consent will be obtained remotely using one of the following methods in this order of preference (this is a method of consent used in the OptiCALS randomised controlled trial on which the CI is co-applicant):

1. Consent is obtained electronically via email or post

Where discussions are unable to take place face-to-face, consent may need to be obtained remotely. An electronic PDF or paper copy of the consent form will be emailed or posted to the participant in advance of arranging a phone/video call between the CI and participant. During the phone call the CI will answer any questions and read out each statement from the consent form. For electronic consent forms the participant will then sign with a type-written signature and email it back to the CI. For paper consent forms the participant will sign the form and post it back to the CI

2. Consent form signed by an independent witness (only applicable for pwMND participants)

Where the pwMND is unable to sign a consent form electronically or on paper (e.g. due to poor hand-grip strength) or the participant is unable to speak due to communication difficulties, an independent witness can sign the form on their behalf. The CI will arrange a phone call or video call (using University of Sheffield's Google Meet account). The pwMND can indicate their consent through typing their agreement with the statements read out by the CI on their communication aid, hand-writing their response or through typing within the chat function of Google Meet. The independent witness who will join the video/phone call, will sign the form to indicate the participant has given informed consent and return the form back to the CI via email or post. With permission from the participant, the call will be recorded on an encrypted and password protected dictaphone. Once the consent form has been signed, a copy will be sent to the participant for their records.

3. Audio recorded verbal consent (only applicable for pwMND participants)

A copy of the consent form is emailed or posted to the participant. The CI will arrange a phone or video call (using secure Google Meet software) with the participant and read out each statement with the participant stating that they understand and agree with them. This may include a pwMND participant using a communication aid with a speech output. The CI will initial each box to confirm the participant has given verbal consent to each statement on the consent form. With permission from the participant, the call will be recorded on an encrypted and password protected dictaphone. The file will be downloaded and saved on a secure University of Sheffield server. Once the consent form has been signed, a copy will be sent to the participant for their records.

Different approaches need to be taken for the different participant types and data collection methods. These are described in detail below.

HCP participant consent

Consent for focus group/interview

Each HCP who agrees to participate in the focus group/interview will be sent the PIS, given at least 24 hours to consider the information and have an opportunity to arrange a call with the CI prior to the focus group/interview to discuss any aspect of the research or data collection. HCPs will be asked to bring a signed copy of the consent form on the day of the focus group/interview. The CI will have copies of the consent form if the HCP does not bring it with them. At the beginning of the focus group/interview the CI will describe the aims of the study, what will be involved, remind participants that they can withdraw without needing to give a reason and discuss how the data will be managed.

Consent for observation

HCPs may be observed in interactions with each other or with pwMND. HCPs will be provided with the PIS prior to the planned observation and allowed time to read it and ask questions. HCPs will be asked to provide written consent prior to any planned observations of interactions they have with other HCPs or with pwMND/caregivers. For the scenario of a meeting being observed, those attending will be reminded that the CI is present for the purpose of the observations.

For the scenario of observing an MND clinic, the CI will put up posters at the entry to the clinic and where HCPs are located e.g. staff room, with the CI photo, information about the study and that the CI will be observing for research purposes. Copies of the PIS and consent forms for HCP's will be available within the clinic and from the CI. The CI will seek to gain consent from every HCP captured in the observations in the clinic setting.

For observing HCP interactions with pwMND outside of the clinic setting e.g. telephone/video calls or home visits, the HCP will be asked to provide informed consent prior to the interaction taking place.

pwMND and caregivers

The CI will seek full consent from all pwMND and caregivers prior to each research activity. PIS and consent forms have been developed for each research activity.

Consent to interviews and review of medical notes

The CI will arrange a time that is convenient for the potential participants, to discuss the study and given at least 24 hours to consider whether they wish to consent to take part. The CI will arrange a time to gain informed consent from the participants. All pwMND participants consented to take part in the interviews will also be able to consent to be invited to take part in the post case study workshop. However, participation in the workshop is optional and not consenting to take part does not preclude them from taking part in the interviews or review of their medical notes.

The pwMND recruited to participate in interviews will be asked to provide consent to view and collect data from their medical notes. The pwMND will be made aware that all data collected from their notes will be anonymised including removing any information that may identify the pwMND, their HCPs or the location of the MND clinic. No direct quotes from the medical notes will be included in any reporting, presentations or publications.

With the consent of the participant, the participant's GP will be sent a letter informing them that the participant is taking part in the study.

Consent for observations

Potential pwMND participants to be observed will be sent information about the study by the local HCPs at least 24 hours before the planned observation of their interaction with a HCP including a participant information sheet and a consent form. The information will describe how the research is focused on how HCPs share information with pwMND, how the observation will take place and the CI contact details if they wish to discuss further. The information will not describe the focus on gastrostomy decision making to avoid causing any distress to the pwMND or any caregivers. During the interactions that the CI is observing, if a pwMND attends that has not received the study information beforehand or has not been able to signa consent form prior to the interaction, the CI will not observe the interaction Where the pwMND or caregiver declines to be observed the CI will immediately leave the interaction. The pwMND, caregiver or HCP can ask the CI to stop observing at any point without needing to give a reason. At the end of the interaction where discussions about gastrostomy have taken place the HCP will seek permission for the CI to contact the pwMND and caregiver at a later date to provide the full PIS describing that the focus of the study is on the decision making about gastrostomy placement, discuss the study and seek explicit consent to add the observation to the data set. When interactions are observed that do not include discussion about gastrostomy placement, no further discussion will take place between the CI and pwMND or caregiver. Only pwMND and caregivers who have provided consent to participate in the study will have their data collected during the observations included in the analysis.

HRA Confidentiality Advisory Group (CAG) approval will be sought to gain ethical approval to observe HCPs having discussions about pwMND e.g. ad hoc discussions or in MDT meetings without having obtained consent from the pwMND whose cases are discussed. The CAG approval is being sought because the CI will be observing discussions about pwMND cases not relevant to the study. Where gastrostomy placement is an issue discussed between HCPs it will not be possible to approach pwMND prior to these discussions taking place. The aim of the observation of HCP-HCP discussions is to understand how HCPs discuss these cases with other HCPs, what informs their clinical reasoning and what actions are planned for after the meeting. These discussions will not be audio recorded to limit the exposure of the CI to patient identifiable information. The CAG approval also relates to the CI having to observe interactions between HCPs and pwMND/caregivers where gastrostomy placement is not discussed.

Consent for the workshop

All pwMND and caregivers who consent to take part in interviews and observations will be asked within the consent form for permission to approach them to take part in the workshop. Following completion of the data collection those pwMND and caregivers who expressed an interest in participating in the workshops will be sent the PIS and consent forms relating to the workshop. Participants will have the opportunity to discuss what is involved prior to attending the workshop. At the start of the workshop the CI will remind the pwMND about the purpose of the workshop, their involvement and how the information will be used. Participants will be reminded that they only need to contribute as much as they feel comfortable and can leave at any point without needing to give a reason.

2.9 Data collection

2.9.1 Information about the case study

The case study site PI will be asked to provide some data about the local MND clinic and patient population for the purposes of describing the demographic of the site in reports and publication. Any data that may help directly or in-directly, identify the case study site will not be included in any reporting. The information that will be collected from each site includes:

- The geographical region in the UK e.g. North East, South East
- An estimate of the total population of the region the MND clinic accepts referrals from
- The point prevalence of pwMND currently on the case sites caseload
- The number of new MND diagnosis made in the clinic each year
- The number of pwMND who are referred for gastrostomy placement by the clinic each year
- The HCP roles that attend the MND clinic

- A description of the different HCPs, teams and services involved in the care of pwMND who attend the MND clinic

The CI will have a 60 minute discussion with each PI at the beginning of each case study to discuss the structure and location of local services, provide a detailed explanation of the case study protocol and recruitment procedures, and discuss the role of the PI during the study.

2.9.2 Information about the participants recruited

To maintain anonymity of HCP recruited to the study limited data will be reported about the demographic characteristics of each participant. The HCP roles recruited participating in each data collection method in total will be recorded. The HCP roles of individuals recruited to the study will not be reported on or linked to any of the data e.g. participant direct quotes. To maintain the anonymity of the pwMND and caregivers recruited to the study no demographic characteristics will be reported of each participant.

Further person identifiable information will be collected and securely saved on Sheffield Teaching Hospitals NHS Foundation Trust (STH NHS FT) servers to enable participants to be contacted with regards data collection, inform the purposive sampling of participants, and to inform participants about the publication of the study or guidelines. Data will include name, address (where required), email address, telephone number, professional role (HCP participants only) and participant type (pwMND, caregiver or HCP). This data will be stored separately to the pseudo anonymised research data which will be stored on encrypted restricted access folders on the University of Sheffield server.

The case studies will include a range of data collection methods, including the different stakeholders involved in the decision-making process, namely, pwMND, their caregiver and supporting HCPs. These data collection methods are described below.

2.9.3 HCP data collection

Non-participant observations

Aim

To understand how HCPs, as individuals and members of the MDT, organise, deliver and coordinate decision support to pwMND about gastrostomy placement

Data collection

The CI will spend up to 7 days, not necessarily consecutively, at each case study site with the aim to observe HCPs interacting with each other and where possible pwMND/caregivers with regards the decision to have a gastrostomy placed. The plan for non-participant observations will be optimised for each specific site during the early discussions with the PI and local HCPs, but could include:

- HCP interactions with pwMND at out-patient clinics, via telephone/video call or domiciliary visits
- Attendance at face-to-face and virtual HCP meetings where pwMND cases or services are discussed
- Observation of the informal or ad-hoc discussions that HCPs may have with other HCPs in their place of work.

At the start of any discussions between HCPs, a HCP already recruited to the study will remind those present that the CI will be observing for research purposes, giving HCPs the opportunity to decline (they should all have provided consent for the observation to take place). During face-to-face meetings the CI will agree with the HCPs attending the meeting where to situate himself to minimise disruption to the meeting. During online meetings, the CI will turn off his camera and microphone and have no interactions with the HCPs during the meeting. The observations will include:

- Which HCPs attend the MDT.
- Discussions about when MDTs decide to introduce gastrostomy placement to pwMND.
- Any challenges related to discussions with pwMND that are discussed.
- Which professional roles are involved in discussions with pwMND about gastrostomy and what their responsibilities are in the decision making process.
- How decisions are made about referring pwMND for gastrostomy.

- What actions or referrals are planned for post meeting.

During observations the CI will take field notes including documenting what was observed; relevance towards the case study objectives and any analytical insights, immediately after consultations using an observation plan checklist. Flow diagrams and maps will be constructed to describe how services are organised and linked to one another, including the lines of communication used. Field notes and data recorded on the observation plan checklist will be transcribed by the CI at the earliest opportunity in preparation for analysis.

The CI will be present in HCP interactions with pwMND where gastrostomy placement is not discussed. When this is the case any field notes recorded will be crossed out in ink, initialled by the CI, and the data will not be included in the data set.

Focus group with MDT members

Aim

To understand the MND MDTs experience of supporting pwMND to make decisions about gastrostomy placement

Data collection

A single focus group of up to eight HCPs, will be conducted on each case study site, following the non-participant observations. To compensate for some of the invited HCPs not attending on the day, the focus groups will be over-recruited (i.e. 10 HCPs will be invited) (Green and Thorogood 2014; pg 143). The CI will offer dates and times for the focus group and choose a date that optimises participation and ensures broad representation of clinical roles. Focus group participants may or may not be the same as those who participate in the non-participant observations or interviews. Using the topic guide, the CI will facilitate group discussion about participants' experience of supporting pwMND making decisions about gastrostomy, and the enablers and challenges of delivering MDT decision support. The focus group will be audio recorded, take place on the case study site premises, and will last up to 90 minutes. Though face-to-face focus groups are preferred where it is not possible the local team will be offered the opportunity to perform it online, using Google Meet.

Interviews with HCPs

Aim

To elicit the views and perspectives of HCPs about how pwMND are supported to make decisions about gastrostomy placement in MND MDT care.

Data collection

HCPs will be recruited to participate in a single interview during the period of case study data collection. Up to 6 HCPs will be interviewed on each case study site. The CI will conduct the interview at a time that is convenient to the participants. Where possible the interviews will take place face-to-face on the case study site premises and last up to one hour. However, the option of a video or telephone call interview will be available if this suits the HCP. The CI will use a topic guide to guide the interview through the key issues being researched. The semi-structured interview will explore the participants' experiences, role and responsibilities in the decision-making process and their perceptions about how the MND MDT functions as a unit of decision support. The interview will be used to further explore and expand on the practice observed in the non-participant observations. The interviews will be audio recorded to allow for transcription of the interviews in preparation for analysis.

2.9.4 pwMND and caregivers’ data collection

Interviews with pwMND and caregivers

Aim

To understand the pwMND perceptions of having the option of gastrostomy placement introduced to them by healthcare professionals

To understand the pwMND perceptions of making the decision to have a gastrostomy and the influence interactions with HCPs have on their decision-making process.

Data collection

Each pwMND and any caregivers recruited will take part in a single semi-structured interview. Ideally pwMND and caregivers will be interviewed individually to allow the individual's perspectives, views and attitudes to not be influenced by that of the ‘other’. However, in view of the communication and emotional challenges pwMND face, a joint interview will always be available. Interviews will take place at a venue of the participants choice, which may include their own home, a private room on an NHS site or if preferred or via video call using Google Meet or telephone call. The interviews will last approximately one hour, with the opportunity for breaks if required. The CI will use the topic guide to guide discussions, while also allowing the participant to discuss the issues they feel are of importance. The interviews will explore the pwMND and caregivers experience of making the decision to have a gastrostomy placed, with a focus on how their interactions with HCPs influenced their decision-making process and the decisions made. The interviews will be audio recorded to allow for transcription of the interviews in preparation for analysis.

pwMND can experience difficulties speaking. For some this can be significant, meaning they are unable to speak at all, and need to use communication aids. Where appropriate, the CI will forward the topic guide to the pwMND one week before the planned date of the interview. The pwMND will be able to type their answers in advance of the interview. These answers can then be further probed by the CI during the interview. Additional time will be allowed to enable the pwMND to express their views if needed. Where it is the participants preference, they will be able to return further answers to the questions and probes after the interview has been completed.

Non-participant observations

Aim

To understand how HCPs, pwMND and caregivers interact throughout the process of decision making

Data collection

There are a number of contexts and settings in which discussions about gastrostomy may take place between pwMND/caregivers and HCPs, or between HCPs, including:

- HCP and pwMND/caregiver interactions. These could occur face-to-face, via telephone or video call, or in writing by letter or email.
- Interactions could take place in hospital wards, out-patient clinics, via telephone/video call, or as domiciliary visits

- HCPs will have discussions with their immediate team members and the wider MDT about patient cases, in formal pre-arranged MDT meetings and ad-hoc communication verbally via telephone or video call, in person or in writing.

The observations will involve the CI sitting in on the interaction between the HCP, pwMND and caregiver. For telephone calls this will involve setting up a conference call or listening in to the conversation taking place on loudspeaker. For video calls the CI will be invited into the call but will turn his camera and microphone off during the interaction. For face-to-face interactions the CI will agree with the HCP, pwMND and caregiver, where the best place to sit is, with an aim to disrupt the interaction as little as possible. The CI will not interact with the HCP, pwMND or caregiver throughout the interaction unless asked a direct question or asked to stop the observation. The HCP, pwMND or caregiver can ask the CI to leave the interaction at any point without giving a reason. The CI will complete short-hand notes in a field diary being mindful to not disrupt the interaction. Following each observed interaction, the CI will document detailed notes about the interaction on the observation plan checklist. Other than discussing the study and gaining consent there will be no direct contact between the CI and the pwMND or caregiver during or after the observation.

The CI will also be observing HCPs discussing pwMND during MDT meetings. During these observations the researcher will be recording data relating to the case discussion with regards gastrostomy placement and the planned actions of the HCPs after the discussion. No patient identifiable information will be recorded. Only field notes will be recorded, when observing these interactions.

Documentation review

Aims

To understand the policies, care pathways and guidance that underpins the process by which HCPs support pwMND making decisions about gastrostomy placement.

To understand what information is documented and shared by healthcare professionals with regards when supporting a pwMND making the decision to have a gastrostomy placed.

Data collection

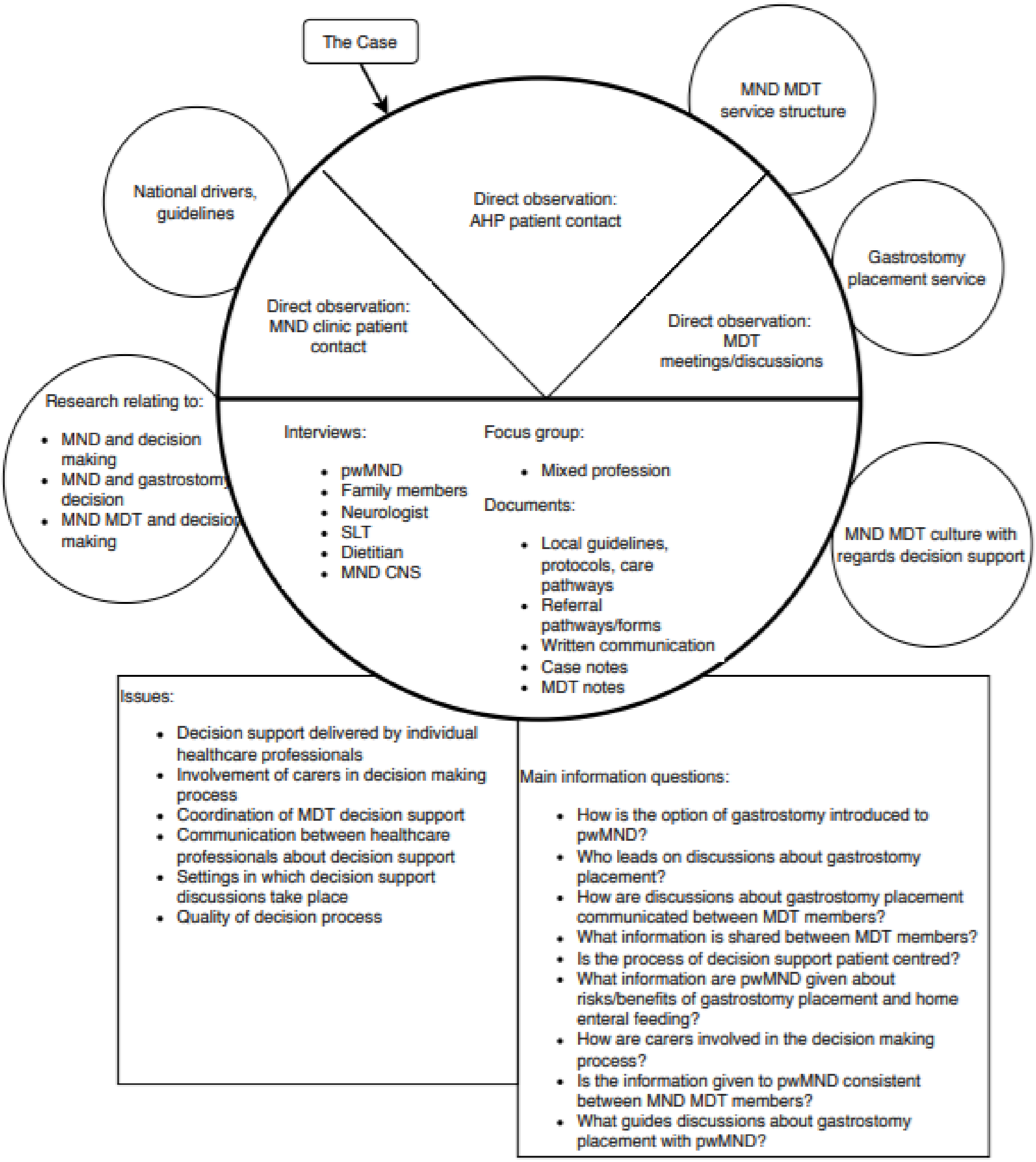
Policies, guidance, care pathways and standard operating procedures

HCPs will be asked to share paper or electronic versions of any documents that the HCPs, teams or services refer that may be relevant to guiding or informing the decision-making process. These may include but are not limited to guidance, policies, care pathways, and standard operating procedures. These may have been developed locally, nationally or internationally, and also maybe single profession or MDT in focus. A documentation review pro-forma will be completed for each piece of documentation shared with the CI, recording information about when the document was written and reviewed, who the authors are, the intended audience and any initial analytical notes. All pieces of documentation will be recorded in an excel database, including a brief summary of what is included in the document and a hyperlink to the whole document which will be stored on University of Sheffield computers in line with the data management plan. The documents and the data entered into the pro-forma will also be available to be uploaded into NVivo for the analysis phase.

Medical notes review

The medical notes related to the pwMND recruited to take part in the interviews on each site will be reviewed by the CI for any text related to the indication for gastrostomy; discussions about gastrostomy; or the decision to proceed or decline gastrostomy. The CI will have obtained a letter of access and research passport for the organisation on which the research is taking place. The CI will also gain access to the systems or paper notes on which medical notes are recorded. Consent will be sought from the pwMND to gain access to and extract verbatim data from their medical notes. Only documentation that is part of the pwMND medical record will be accessed for data collection. Data will be copied verbatim onto a paper pro-forma which will be anonymised, transcribed and saved on secure University of Sheffield servers at the earliest opportunity after periods of time in the field. Once transcribed, the CI will destroy the paper pro-formas. Any data that could be used to identify either HCPs or patients, or the case study site will be anonymised. No data from the medical notes will be used for direct quotes in the reporting.

Figure 3. Summary of multiple case study data collection



2.9.5 Order of case study and analysis

The order and timing of data collection on each case study site is guided by the aims of each method and how each will help develop and adapt data collection to future data collection and strengthen the analysis plan. As explained below the data collected will be analysed concurrently and iteratively using the Framework approach.

Order of activities on each case study site including data collection methods:

1. Online meeting with the PI

2. Online presentation to and meeting with members of the MDT on the case study site
3. Non-participant observation of practice on case study site
4. Collection and review of any guidance, policies, care pathways or standard operating procedures that relate to how pwMND are supported to make decisions about gastrostomy
5. Review of pwMND medical records
6. pwMND and caregivers' interviews
7. MDT focus group and HCP interviews
8. Workshop to feedback findings and develop guidance

2.10 Analysis

2.10.1 Rationale for data analysis approach

The Framework approach developed by Ritchie and Spencer (1994) is a method of data management and thematic qualitative analysis developed originally for use in social policy research but now used extensively in healthcare research (Gale *et al.,* 2013). Framework approach has been used to analyse data collected during case study research conducted in a variety of healthcare settings, including emergency care (Edwards *et al.,* 2020); palliative care (Dodd *et al.,* 2020); evidence-informed decision making (Lukeman *et al.,* 2019); care delivery in Parkinson's disease (Tod *et al.,* 2016) and decision making in dementia care (Smebye *et al.,* 2102). Framework is ideally suited to analyse case study data for a number of reasons, including allowing a deductive and inductive approach; managing a large volume of different data types; facilitating the comparison of findings within and between cases; and triangulating different sources of data. For each case study Framework approach will be used to analyse the data from the embedded pwMND cases and the case study as a whole. The framework approach includes the use of a central matrix that displays summarised data relating to both cases (rows) and themes (columns) (Gale *et al.,* 2013). The organisation of the analysis in this way, allows for the analytical interrogation of the findings to identify relationships, that can provide theoretical explanations about the phenomenon (Goldsmith 2021). It is these higher level and more abstract interpretations that will support the transferability of the findings and aid the development of professional guidance that can be relevant to all MND HCP's and MDT's. Finally, the analysis will be an iterative process that will be undertaken concurrently with data collection. This will allow emerging themes to inform changes to the topic or observation guides, allow exploration of pertinent issues in more depth and facilitate alternative lines of enquiry including actively searching for outliers.

2.10.2 The analysis process

The anonymised transcribed data from the non-participant observations, interviews and focus groups, the examples of local guidelines, and the data taken from pwMND medical notes will be uploaded onto NVivo computer assisted qualitative data analysis software (CAQDAS). NVivo will be used to manage the large and varied data set, while the analytical process will involve the CI, in collaboration with his supervisors, organising the data into a format that allows for answers to the research question to be developed. The framework approach will ensure that the final analysis would have at its disposal, the breadth and depth of data that will allow for the research questions to be answered in full. The framework approach to qualitative analysis will include the 5 steps described by Ritchie and Spencer (1994): 1. Familiarisation; 2. Identifying a thematic framework; 3. Indexing; 4. Charting; 5. Interpretation of patterns (Ritchie and Spencer 1994).

2.10.3 Triangulation of findings

The case study will implement two different types of triangulation as defined by Denzin (1978) and summarised in figure 4. Multiple methods are being used to collect data on each case study site including interviews, focus groups, documentation review and non-participant observations (methodological triangulation). These different methods provide varying degrees of naturalistic data from the observations and review of medical notes capturing what participants 'do' with regards the phenomenon in real-world practice, through to focus groups and interviews which capture data about how the participants 'perceive' the phenomenon. Within each case, data is being collected from different participants including pwMND, caregivers, and healthcare professionals and in different settings including clinics, HCP offices, patients homes and online (data triangulation). Data triangulation is also achieved through the collection of the same types of data set from three different case study sites.

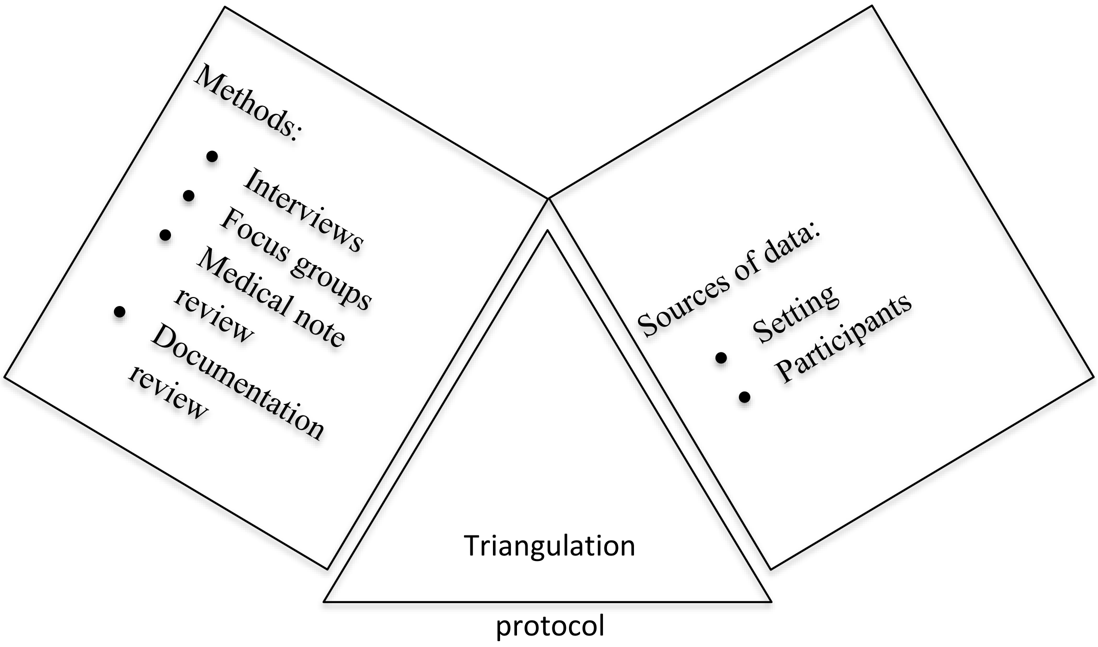


Figure 4. Summary of types of triangulation used.

The data management aspect of the Framework approach, which organises the data and analysis into a matrix, provides an ideal format for triangulation to take place, with the ability to compare across themes and cases (or units of analysis). The triangulation protocol will cover both the triangulation of data from within and between cases. Following the procedural stages described by Farmer *et al.,* (2006), the findings from each individual case study will be triangulated as follows:

1. Sorting

The themes from the different data sources are combined. This is repeated for each case study site.

2. Convergence coding

The themes from each data set will be inserted into the first column of an integration grid with the different data collection methods along the top row (O'Cathain *et al.,* 2020). The grid will be populated with summaries about whether the theme is covered by each data set including reference to the meaning, prominence, coverage and examples (Farmer *et al.,* 2006).

For the triangulation of the findings from the three case study sites, a similar process will be used. Two interrogation grids will be developed. The first to compare the same findings from the embedded pwMND cases. The second to compare the overall findings across each of the three wider case studies.

3. Convergence assessment

The researcher, in collaboration with his supervisors, will perform an assessment of the convergence of the findings based on the convergence coding. This will include describing the degree of convergence and interpreting whether this is due to the method of collecting the data or a chance finding. The convergence assessment will be documented in the final column (labelled 'Triangulation conclusion') of the integration grid where the researcher will provide a summary about whether there is agreement, partial agreement, silence or dissonance in relation to the theme across the different data sets on each case study site. The researcher will interrogate the findings from the embedded cases and the overall case study findings of each of the three case study sites for evidence of agreement, partial agreement, silence or dissonance.

4. Completeness comparison

The data informing the themes for each data set will be compared in terms of the range of evidence it provides. The comparison will allow for a rich description of the overall findings, and how each data set contributes to these and why key differences may exist.

5. Researcher comparison

While only the lead researcher will be conducting the analysis of the primary data collected and implementing the triangulation protocol, continual discussions will be taking place with supervisors about conclusions made about the analysis and triangulation. However, it is not possible to perform a comparison of different researchers' interpretations.

6. Feedback

The interpretations being made will be discussed throughout with supervisors to agree and debate what any differences in convergence mean in relation to answering the main research question. The results of the triangulation process will also be presented to the researchers' expert panel, PPI panel and back to the participants of the study in the workshop. While the feedback received will not alter the findings presented, discussion will take place in the report about how and why they may have differed and what this may mean for future research.

Reporting the results of the triangulation process

The triangulation protocol will be implemented with reflexivity, critiquing the strengths and weaknesses of the process and acknowledging how the researcher made analytical decisions. The large number of different data sets being collected on each site, each method with its own focus and scope will introduce challenges with regards to how convergence is assessed. When triangulating the data, and in particular looking for convergence or dissonance in the findings from different data collection methods or sources, it is important to recognise that the reason for presence or absence of dissonance may be related to how the data was collected (Farmer *et al.,* 2006). For example, the interview data collected will have been influenced by the topic guides which addressed many of the objectives of the study. However, the same objectives may not have been covered within less guided data collection such as non-participant observations or review of medical records. These will be discussed at depth, in the reporting of the study, to ensure the reader is aware of them and to inform their own critique of the conclusions reached.

Workshops and HCP guidance development

The findings of the multiple case study will inform the first draft of a set of recommendations to guide how HCPs can organise and deliver decision support within the context of MND MDT care. These recommendations will underpin the development of professional guidance.

The guidance will be developed in collaboration with the case study sites, supervisors, expert and PPI panels. pwMND, caregivers and HCPs who have participated in the study will have indicated in the consent process whether they wish to be approached to take part in a post case study workshop. In total across the three case study sites, up to 15 people with

MND, 15 caregivers and 24 HCPs will attend the workshops. The workshop will take place on the local organisations premises that has access and facilities for pwMND who may have mobility problems. pwMND and caregiver participants will be offered a £25 voucher for their time, reimbursed for any travel costs. Refreshments will be made available. During the workshop the findings from the case studies (and previous completed research i.e. qualitative evidence synthesis and survey) will be presented followed by the draft recommendations for practice. The workshops will take 2 hours and be audio recorded. The CI will facilitate the group discussion with a focus remaining on how the study findings can be interpreted into practical, pragmatic guidance for HCPs who support pwMND making decisions about gastrostomy placement. In line with previous methods used to develop the gastrostomy decision making information resource, the mytube website, the workshop will be split into 3 parts (White *et al.,* 2017). The first part will involve a discussion with the CI and the pwMND/caregivers only, to hear the voice of the service users first. There will then be a cross-over where the HCPs join the workshop to hear the views of the pwMND/caregivers and to allow discussion. Finally, to manage the energy levels of pwMND, the final part of the workshop will involve only the HCPs, with a focus on the appropriateness, applicability and feasibility of the recommendations and to develop the practical guidance to help HCPs meet the recommendations in practice. Refreshments will be available before, during and after the workshop. With the pwMND and caregivers leaving the workshop before the end, the CI will email each pwMND caregiver after the workshop to check they are ok and that the workshop did not cause any upset or raise any questions. Where indicated the CI will direct pwMND/caregivers to their local HCP for further support. The three workshops on each case study site will happen sequentially, with time between workshops used to draft a new version of the guidance. Finally, all stakeholders will be sent a copy of the final guidance to provide feedback on, before it is completed.

The format of the guidance will include a summary of the research completed and implications for practice. Each recommendation will be followed by practical guidance with regards to how HCPs as individuals and as part of the wider MND MDT introduce gastrostomy placement to pwMND and guide decision making. A focus will remain on how the HCPs coordinate the decision support they deliver with the wider MDT. The guidance will include reference to the contextual enablers and barriers to decision making and practical steps about how to address these.

The CI plans to collaborate with and seek endorsement from the MND Association for the guidance, which will support its dissemination. Further dissemination of the guidance will occur through publishing the guidance in professional publications, sharing on HCP forums and professional groups and presenting at national and international conferences.

Reflexivity

In line with the principles of all research employing qualitative methods of data collection and analysis, it is important for the researcher to undertake every stage of the research from conceptualisation through to writing up the final report, in a reflexive manner (Mason 2002; pg 66). The CI recognises that his clinical role as a dietitian supporting pwMND to make decisions about gastrostomy, and his knowledge of the evidence base on this topic have heavily influenced the research questions asked and the design of the study. While a partly deductive approach to the analysis is being taken, the intention is still for an understanding of the phenomenon to be derived inductively from the data collected during the case studies. To support, reflect and evidence a reflexive approach the following strategies will be employed:

- The CI will maintain a reflexive research diary throughout the data collection, analysis and report writing stages of the research. In addition to recording observations, thoughts and analytical ideas, the diary will be used to capture and reflect on how the CI’s experience may be influencing the direction of the research
- The CI will maintain regular dialogue with supervisors, with discussions about reflexivity being a fixed agenda item in meetings.
- The analytical themes and development of typologies, will be discussed with PPI and expert panels, with the view to challenging them and exploring alternative explanations
- A rigorous and transparent audit trail of the analysis process made possible by the systematic nature of the framework approach, will be available for external scrutiny and provide evidence of the trustworthiness of the findings

Data management

Data will be collected and retained in accordance with The General Data Protection Regulation (2016/679) (GDPR). An Excel database will be kept with the participants names, contact details and pseudo anonymised study identifiers for the CI's reference only. This database will be saved on restricted access folder on the secure STH NHS FT servers where no other study materials such as any reports, publications or research data will be stored. Only the CI and a researcher coordinator of the STH NHS FT Clinical Research and Innovation Office (CRIO) will have access to this information. All research data (e.g. audio recordings and transcriptions) and documents will be securely stored on password protected University of Sheffield server. Audio recordings of interviews and focus groups will be recorded on encrypted and password protected dictaphone. The audio files will be immediately deleted from the dictaphone once they have been downloaded onto University of Sheffield servers.

The audio files will be shared with a professional transcription service in line with the confidentiality policies of the service used. Two transcription services have been identified: Sheffield School of Health and Related Service (ScHARR), University of Sheffield Transcribers group (https://www.sheffield.ac.uk/scharr/research/scharr-information-governance-policy/information-governance/transcription) and an external transcription service White Transcriptions (https://white-transcriptions.co.uk/). Copies of both services' confidentiality agreement or personal data processing agreements have been shared by the services. A confidentiality agreement will be signed by the transcription service with regards to maintaining the confidentiality of the audio and textual data, how the data is securely transferred, how data is processed and how it will be deleted when transcription is completed. Once the completed and checked transcriptions are saved on secure University of Sheffield servers, the CI will delete the audio files and the transcription service will delete the audio and transcription files from their records.

The text from pwMND medical notes recorded on paper pro-formas will be transcribed by the CI, anonymised and saved securely on University of Sheffield servers. Once transcribed the paper proformas will be destroyed.

All of the CI’s field notes and observation plan checklists will be transcribed by the CI and saved on secure University of Sheffield servers. The field notes will be stored in a locked cupboard on University of Sheffield premises when the CI is not using them for fieldwork.

All paper consent forms will be stored in the study file in secure University of Sheffield premises. Electronic copies of completed consent forms will be saved in secure University of Sheffield servers, in the same folders as the database that includes participants names and study ID.

At the end of the study, the study documents, research data and analysis will be securely transferred to password protected folders on the STH NHS FT servers that are separate to the folders where person identifiable data is stored. The person identifiable information will be stored for 3 years after the study has ended to allow for the researcher to inform participants about when the study and guidance has been published. STH NHS FT will archive the study data, analysis and documents anonymously for 5 years.

Other researchers may find the data collected to be useful in answering future research questions. The anonymised data and the analysis will be uploaded and stored on ORDA (the secure University of Sheffield's data repository hub for managing and sharing research data) for 10 years after which it will be deleted. The anonymised data will be made available to other researchers on request.

A data management plan has been developed to provide detailed information about how data will be collected, processed, transferred and stored during the study.

Ethical considerations

6.1 Approvals

Prior to conducting the multiple case study NHS research ethics committee (REC) and HRA approval will be obtained. With it not being possible to obtain the consent of pwMND discussed by HCPs (e.g. in MDT meetings or ad-hoc discussions), a Confidential Advisory Group (CAG) application will be made in parallel with the IRAS application. The NHS REC and HRA application will be submitted via the IRAS online system. Following this, research governance approval will be obtained from each case study site research department, prior to getting green light from the sponsor to commence the study.

The local site approval is complicated by the aim to understand and collect data from HCPs that often span a number of different organisations, both locally and further afield when a tertiary service is provided. Research governance approval will be sought from the organisation that hosts the MND clinic to approach and recruit pwMND and HCPs to participate in the study. The PI at each site will have responsibility for recruiting HCPs who may be employed by other organisations. HCPs recruited through the PI employed by other organisations will only be able to participate in observations of MDT meetings, interviews, focus groups and but not the observations of interactions with pwMND. Where it is deemed feasible and will aid recruitment, organisations external to the case study host organisation will be approached to become a Patient Identification Centre (PIC). Observations of pwMND interactions with HCPs will only occur at organisations where Patient Identification Centre (PIC) approval has been obtained from the HCPs organisation.

Each case study site will have a site file which will include copies of the NHS REC and HRA approval letters, the protocol, the PIS and consent forms.

6.2 Informed consent

All participants taking part in interviews, focus groups, observations and review of medical records will be required to complete a consent form that indicates they understand what is involved, how their data will be used, their rights including being able to withdraw at any point without needing to give a reason. The only scenario where consent is not obtained is when the CI observes HCPs discussing pwMND cases. See section for further information about how consent will be obtained from the different participant types and data collection methods.

6.3 Anonymity

Case studies have been associated with a risk to revealing the identity of participants, and therefore caution should be taken about how participants or case sites are described (Crowe *et al.,* 2011). MND MDTs usually include a small number of HCPs, with some professions only being represented by a single individual. There is a risk that HCP participants could be identified in the reporting of the study. The CI will keep a database of all the participants who have consented to taking part in any of the research activities, including their name, contact details and professional role (HCP participants only). Each participant will be assigned a pseudo anonymised study ID, which will be used to identify participants in the files and documents relating to data collection. Electronic consent forms will also include the name of participants. All electronic documents that include participant identifiable information will be saved in a separate password protected folder on STH NHS FT computer servers, that can only be accessed by the CI and CRIO research coordinator. The research data including transcripts and audio files will be stored in separate password protected folders, and only include the participants study ID as an identifier. Any information that may aid identification of the participant will be deleted or anonymised. This will include names, professional roles, geographical location, and names of clinics. In any reporting or publications participants will only be identified using a pseudo-anonymised study ID, with no reference to the location of the clinic or their professional role. The different sites will be referred to in general terms, for example by region or type of MND care provision.

6.4 Confidentiality

Data will be recorded, stored and processed as per the data management plan. The CI will store field notes in a locked cupboard within University of Sheffield premises, as soon as he returns from site visits. Care will be taken to ensure that field notes are kept secure when on-site visits including keeping them in locked cupboards or safes. Dictaphones used to record interviews and focus groups will be encrypted, password protected and securely stored when not in the possession of the CI. All recordings will be deleted from the dictaphone once they have been uploaded onto secure university computer servers. The recordings will be securely transferred to and from a professional transcription service. Once the audio recordings have been transcribed, they will be deleted from the university computers.

6.5 Potential for participant harm

pwMND making decisions about gastrostomy placement are generally at a later stage of their disease. They may also have had challenging discussions with HCPs and their caregivers about their future care or disease course. Therefore, the observation of their decision making journey and the interviews may be distressing for some. The CI has collaborated with his PPI group to design the research plan in a way that is sensitive to these issues and also use his clinical experience supporting pwMND to minimise the risk of distress. The CI is a HCP who has significant experience in caring for pwMND and will use this experience to observe for and be sensitive to, signs of distress or upset during any of the research activities. The CI will be able to signpost the participants to sources of advice and support, including the MND Association and their local healthcare teams. The identification, management and follow up to pwMND or their caregivers experiencing

If the pwMND or caregivers disclose any information that indicates risk of harm or abuse, this will be discussed immediately with supervisors, to agree an appropriate response. The response may require further discussion with appropriate authorities if the information deemed necessary.

HCPs will be asked to reflect and share their experiences of supporting pwMND to make decisions about gastrostomy and on the practice of the local MDT. Where HCPs share challenges about their own practice or that of others this may have the potential to create issues with colleagues. This scenario may be more relevant during the mixed profession focus groups. At the beginning of the focus groups the CI will remind the participants that everything discussed should remain confidential and not shared outside the group attending.

A distress policy has been developed detailing how the CI will observe for and respond to any signs of participant distress.

6.6 Observing dangerous healthcare practice

During the course of observations, there is potential for the CI to observe HCP practice that is concerning or dangerous and could lead to patient harm. Though it is unlikely that this would occur, in line with the CI’s professional responsibility as a HCPC registered HCP, the CI would report any practice that could be potentially harmful to patients to a member of the clinical leadership within the organisation. The CI would also have discussions with his supervisor (who is also a senior clinician) to agree the most appropriate course of action.

6.7 Response to a patient becoming unwell

The CI will be observing or interviewing pwMND and caregivers in both the clinical setting and potentially alone in their own homes. pwMND making decisions about gastrostomy are likely to be in the later stages of the disease and therefore be living with increasing morbidity, including swallowing, breathing and mobility difficulties. There is a risk that the pwMND could become acutely unwell or experience a fall in the pwMND presence. The CI is a registered HCP, has completed a health and safety course, and through their clinical role undertakes annual resuscitation training courses. The CI would respond to any incident that occurred to minimise the risk to the pwMND while immediately seeking urgent medical assistance.

6.8 Harm to the CI

During observations the CI will ensure all HCPs, pwMND and caregivers are aware that he is in an academic capacity only and will not be assisting with patient care or giving any professional advice.

The CI may be visiting pwMND in their own homes to perform interviews. The CI has been a community HCP visiting patients in their own homes for 17 years, including pwMND, and through this is aware of the risks of performing lone home visits. The CI will ensure that a member of the supervisory team and a family member are aware of any visits to participants home, what the address is and when the visit is expected to finish. Escalation procedures will be in place to manage the situation where the CI does not inform the supervisor or family member that the visit has been safely completed.

The CI has performed a risk assessment and developed a risk management policy that has been reviewed and agreed by the Head of the Neuroscience Department at University of Sheffield (Professor Majid).

6.9 Potential benefits for participants

There will not be any immediate benefits to people with MND and caregiver participants. All participants will have the knowledge that their involvement in the study will help generate a better understanding about how MND healthcare professionals and MDTs support people with MND and their caregivers to make decisions about gastrostomy placement. This will include the development of some professional guidance that aims to improve practice. The completed guidance, including a lay summary for people with MND and caregivers, will be shared with all participants including being made available to the wider MND healthcare professional and patient population.

The MND MDTs will receive a summary of the findings relating to their individual case study site MND MDT practice. This information could be used to evaluate the service they currently provide and to inform service improvement. The publication of the research findings and guidance will have benefits for healthcare professionals in terms of providing an evidence base and practical guidance about how to organise and deliver decision support to people with MND about gastrostomy placement.

Patient and Public Involvement (PPI)

The CI wider doctoral research plan included being informed by collaboration with a range of established PPI groups and individuals with MND. These discussions helped guide decisions when applying for the NIHR doctoral fellowship. Since commencing the fellowship the CI has successfully recruited to a panel of pwMND and caregivers who have experience of making the decision to have a gastrostomy placed. Since commencing the doctoral fellowship, the PPI panel have received training and we have met twice to discuss the research plan. The collaboration with the PPI panel resulted in the following changes to the original protocol:

- Previously there had been a plan to ask pwMND to complete diaries about their experience of making the decision to have a gastrostomy placed. While this would have provided a contemporaneous recording of how the pwMND felt about gastrostomy following discussions with their HCPs, the panel had concerns about this data collection method. The collaboration with the PPI panel included the following discussion and decisions:
    - the ability to recruit people early enough in their process of decision making
    - the challenge in identifying pwMND who may go on to have multiple discussions about gastrostomy placement with their HCPs. Some may have a single discussion as part of the HCP preparing pwMND for future decisions but then not discuss again for many months.
    - The burden associated with recording the diaries and returning them to the CI. This may influence the quality of the data provided.

- The ruling out of any purposive sampling due to the requirement to recruit pwMND early in the data collection period on each case study site.
- The panel suggested that aiming to recruit pwMND at different stages of the decision-making process would provide a broader view of what occurs including purposively recruiting pwMND who have made a decision to have a gastrostomy, decline gastrostomy, are still considering gastrostomy and have had the option discussed early to single interviews rather than completing guidelines.
- The panel made a strong argument for this change which the CI and his supervisors agreed with.
- The issue of not being able to disclose the full details of the study to pwMND who are planned to be observed by the CI generated some interesting discussion.
- The panel advised that pwMND views on not having full knowledge about the focus of the study prior to consenting to be observed may be an issue for some individuals.
- However, it was also agreed that being informed that a discussion about gastrostomy may occur in a future planned consultation with their HCP would potentially cause distress and anxiety.
- The panel agreed it was proportionate and appropriate to not inform pwMND (and their caregivers) that the study focused on gastrostomy decision making and that they would have the opportunity for the observation to not be included after it has taken place.
- The panel felt it was important that HCPs outside of those that attend the MND clinic are recruited to the interviews and focus groups. They advised that often it was these other HCPs that had more detailed discussions with them about gastrostomy placement.
- The panel felt the approach to taking consent from pwMND who were physically unable to sign a consent form was appropriate.

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Appendix 8. pwMND interview topic guide.

1. Introduction

- Introduce self, and research role
- Give summary of the research topic
- Summarise aims and objectives of the research
- Repeat information in PIS about anonymity and confidentiality
- Remind participants that they can have a break or stop interview at any point without giving a reason. They do not have to answer every question.
- Advise that interview will take up to one hour
- Check if participant has any questions
- Seek consent to start audio recorder and to start the interview

2. Background

Aims: to understand the context in which the participants made the decision to have a gastrostomy placed

- When were you diagnosed with MND?
- Who did they discuss important decisions with?
- Where do discussions take place with your healthcare professionals (face-to-face, online, telephone)?
- Have you made a decision about gastrostomy placement yet?

3. Finding out about the option to have a gastrostomy for the first time

Aims: to understand how healthcare professionals introduce the option to have a gastrostomy to people with MND

- Describe the first time you found out about the option to have a gastrostomy placed
    - Who introduced it to them?
    - How was it introduced?
    - Why was it introduced?
    - How did it make them feel?
    - What was their initial response?
    - How was the initial discussion managed by the HCP?
    - What happened after this first discussion?

4. **Process of decision making**

Aims: to understand the content and outcome of the discussions that they have with HCPs and caregivers about the decision to have a gastrostomy placed

- What information do/did you need to make a decision?
- What impact does the symptoms of having MND (e.g. swallow; mobility; breathing) have on your decision making?
- What resources do you refer to during decision making?
    - internal/external information; websites; decision aids
- What was the role of your caregivers during decision making?
    - How have you supported decision making
    - What challenges have you faced
    - How have caregivers interacted with healthcare professionals
- What has been the role of your healthcare professionals in decision making
    - What professional roles discussed gastrostomy with you?
    - What did the different professions focus on?
    - Consistency of support/information (any differences between professions?)

    - Where do discussions take place?
    - Communication skills (language/information shared/frequency/framing)
    - What information did your HCP share with you during decision making?
        - understanding about how gastrostomy tubes are placed?
        - understanding whether enteral feeding is the right decision for participant?
        - Discussing issues important to participant
- Please describe how you made decisions about the timing of gastrostomy placement
    - What information did you need?
    - What factors influenced the decision about timing?
    - How sure were you about timing?
    - How did your understanding/views change with time?
    - How did your understanding/views change with further discussion?
    - How did your healthcare professionals support your decision about timing?

5. Making a decision

Aim: Understand how participants experience of making a decision about gastrostomy placement

- What lead to you making a decision?
- How sure were you about the decision you made?
    - Have you experienced any regret about the decision you have made?
    - Was the decision supported by the caregiver?
- How do you feel about the timing of the decision you have made?

6. **Finish**

- Do you have any suggestions for how healthcare professionals or teams could improve the support they give to people with MND and their caregivers?
- Is there anything else you would like to share?
- Check that the interview has not caused any upset.
- Advise participant about how their data will be used (added to the wider data set, analysed, use of quotes, inform guidance, publications)
- If consent was given advise that you will contact them about the opportunity to attend a workshop to develop healthcare professional guidance
- Ask participant if they wish to receive a printed copy of their diary
- Thank participant for taking part in the study

Appendix 9. Documentation review proforma.

Site ID		Date document obtained	
Title of document			
Authors			
Version number		Date first published	
Date of most recent review		Date of next review	

Aim of document
Description of content
Analytical thoughts

Appendix 10. Full Medline on Ovid search strategy

APPENDIX B (from Paper 1). Full Medline on Ovid search strategy.

See below for the full Medline on Ovid search strategy run on 27/9/2021.

Ovid MEDLINE(R) and Epub Ahead of Print, In-Process, In-Data-Review & Other

Non-Indexed Citations, Daily and Versions(R) <1946 to September 27, 2021>

1 motor neuron disease*.ti,ab.

2 motor neurone disease*.ti,ab.

3 MND.ti,ab.

4 exp Motor Neuron Disease/

5 Amyotrophic lateral sclerosis.ti,ab.

6 ALS.ti,ab.

7 exp Amyotrophic Lateral Sclerosis/

8 Lou Gehrig* disease*.ti,ab.

9 Progressive bulbar pals*.ti,ab.

10 exp bulbar palsy, progressive/

11 Progressive muscular atroph*.ti,ab.

12 Primary lateral scleros*.ti,ab.

13 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12

14 decision.ti,ab.

15 exp Decision-making/

16 (mak* adj2 decision*).ti,ab.

17 decision-making.ti,ab.

18 (shared adj2 decision).mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]

19 patient engage*.ti,ab.

20 exp Patient Participation/

21 patient participat*.ti,ab.

22 exp Decision-making, Shared/

23 exp Patient Preference/

24 choice*.ti,ab.

25 option*.ti,ab.

26 preference*.ti,ab.

27 advance* care plan*.ti,ab.

28 advance* decision*.ti,ab.

29 exp advance care planning/

30 14 or 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22 or 23 or 24 or 25 or 26 or 27 or 28 or 29

31 exp Qualitative Research/

32 qualitativ*.ti,ab.

33 interview*.ti,ab.

34 exp interview/

35 focus group*.ti,ab.

36 exp focus groups/

37 thematic analys*.ti,ab.

38 conversational analys*.ti,ab.

39 content analys*.ti,ab.

40 phenomenol*.ti,ab.

41 ethnograph*.ti,ab.

42 exp Anthropology, Cultural/

43 attitude*.ti,ab.

44 experience*.ti,ab.

45 perception*.ti,ab.

46 exp Attitude to Health/ or exp Perception/

47 theme*.ti,ab.

48 (grounded adj (theor* or stud* or research or analys?s)).mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating subheading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]

49 (biographical adj1 method).ti,ab.

50 theoretical sampl*.ti,ab.

51 (purpos* adj4 sampl*).ti,ab.

52 (observational adj1 method*).ti,ab.

53 (constant adj (comparative or comparison)).ti,ab.

54 ((discourse* or discurs*) adj3 analys?s).ti,ab.

55 narrative analys*.ti,ab.

56 (social construct* or post-modern* or postmodern* or post-structural* or poststructural* or post structural* or post modern* or feminis*).mp. or interpret*.ti,ab. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]

57 31 or 32 or 33 or 34 or 35 or 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49 or 50 or 51 or 52 or 53 or 54 or 55 or 56

58 exp Gastrostomy/

59 gastrostom*.ti,ab.

60 PEG.ti,ab.

61 RIG.ti,ab.

62 percutaneous endoscopic gastrostom*.ti,ab.

63 Percutaneous image guided gastrostom*.ti,ab.

64 per-oral image guided gastrostom*.ti,ab.

65 radiologically inserted gastrostom*.ti,ab.

66 percutaneous radiological gastrostom*.ti,ab.

67 g-tube.ti,ab.

68 feed* tube.ti,ab.

69 tube feed*.ti,ab.

70 artificial feed*.ti,ab.

71 artificial nutrition.ti,ab.

72 enteral feed*.ti,ab.

73 enteral nutrition.ti,ab.

74 exp enteral nutrition/

75 non-oral feed*.ti,ab.

76 exp Ventilation/ or exp Noninvasive Ventilation/

77 non-invasive ventilation.ti,ab.

78 noninvasive ventilation.ti,ab.

79 non invasive ventilation.ti,ab.

80 NIV.ti,ab.

81 exp Respiration, Artificial/

82 mechanical ventilation.ti,ab.

83 tracheostomy.ti,ab.

84 life prolonging treatment.ti,ab.

85 life prolonging intervention.ti,ab.

86 58 or 59 or 60 or 61 or 62 or 63 or 64 or 65 or 66 or 67 or 68 or 69 or 70 or 71 or 72 or 73 or 74 or 75 or 76 or 77 or 78 or 79 or 80 or 81 or 82 or 83 or 84 or 85

87 30 or 86

88 13 and 57 and 87

Appendix 11. Summary of supplementary search strategy.

APPENDIX C from paper 1.

See table C1

Table C1. Summary of the supplementary search strategy.

Supplementary search strategy	Searches performed
Thesis search	Search of the ProQuest dissertation and theses database
Reference list	Reference lists of all publications included in the review and 23 relevant reviews
Forward citation search	Web of Science forward citation search of all publications included in the review on 25/10/21
Journal contents	Scanned the previous 3 years online journal contents of 'Amyotrophic Lateral sclerosis & Frontotemporal Degeneration' on 14/10/21
Conference proceedings	Search the previous 3 years (2018, 2019 and 2020) abstracts for the annual International Symposium on ALS/MND checked on 8/10/21.
Author and subject matter experts	The authors from the publications included in the review and selected academic and healthcare professional subject matter experts were emailed to identify any references that the searches may have

	missed (9 replied). The MND Association and BDA neurology specialist group were also contacted

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Author / Editor

King Edward's Hospital Fund for London.

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White, Sean; O'Cathain, Alicia; Halliday, Vanessa; Croot, Liz; McDermott, Christopher J.

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Author of Portion(s)

White, Sean; O'Cathain, Alicia; Halliday, Vanessa; Croot, Liz; McDermott, Christopher J.

Publication Date of Portion

Appendix 13. The full survey data set.

Supplementary information 5 from Paper 2.

Table 3. The full data set.

Question	n (%)
Do you initiate discussions about gastrostomy placement with people with MND? (N=139)	
Yes	115 (83)
No	24 (17)
Which of the following factors prompt you to initiate discussions about gastrostomy placement with people with MND? (N=115)	
Swallowing difficulties	114 (99)
Weight loss	105 (91)
Person with MND requesting information on gastrostomy placement	103 (90)
Difficulties taking medications	94 (82)
Caregiver requesting information on gastrostomy placement	93 (81)
Dehydration	89 (77)
Poor appetite	80 (70)
Prolonged mealtimes	81 (70)
Low body weight	76 (66)
Respiratory failure	72 (63)
Poor lip seal	51 (44)
Time and effort to prepare meals	49 (43)

Caregivers needing to assist people with MND to eat and drink	45 (39)
Upper limb weakness	37 (32)
Lower limb weakness	10 (9)
Other	20 (18)
In most cases, WHEN DO YOU initiate discussions about gastrostomy placement with people with MND? (N=115)	
Prior to the onset of any indications for gastrostomy	42 (37)
Following the first presentation of any indications for gastrostomy	51 (44)
When there is evidence that the indications for gastrostomy are progressing	15 (13)
Other	7 (6)
In most cases, when do you think gastrostomy placement SHOULD be first discussed with people with MND? (N=137)	
Prior to the onset of any indications for gastrostomy	91 (66)
Following the first presentation of any indications for gastrostomy	33 (24)
When there is evidence of the indications for gastrostomy are progressing	5 (4)
Other	8 (6)
Which aspects of the procedure to place a gastrostomy do you routinely discuss with people with MND? (N=139)	
Detail about the procedure to place the gastrostomy	108 (77)
Length of hospital stay	103 (74)
The impact of respiratory failure on the risks associated with the procedure to place a gastrostomy	97 (70)
Risk of infection post gastrostomy placement	89 (64)

Pain post gastrostomy placement	88 (63)
Risk of aspiration	83 (60)
The sedation required during the procedure	79 (57)
Risk of not surviving gastrostomy placement	54 (39)
I do not discuss the procedure to place a gastrostomy with people with MND	10 (7)
Other	31 (22)
Which of the following issues relating to living with a gastrostomy tube do you routinely discuss with people with MND? (N=139)	
Enteral feed methods available e.g. bolus and pump feeding	119 (86)
The healthcare professional support they will receive once home with gastrostomy tube	118 (85)
Identifying who will use and care for the tube post placement	112 (81)
Care of the gastrostomy	105 (76)
The impact gastrostomy feeding will have on caregivers	91 (66)
Care of the gastrostomy stoma site	90 (65)
Later presenting tube complications and how they are managed e.g. blocked, damaged or displaced tubes	80 (58)
The option to withdraw enteral feeding in the future	79 (57)
Later presenting stoma complications and how they are managed e.g. infection, overgranulation or leakage	68 (49)
Impact gastrostomy may have on body image	63 (45)
Gastrointestinal side effects of gastrostomy feeding e.g. diarrhoea, constipation, bloating	60 (43)
I do not discuss living with a gastrostomy tube with people with MND	10 (7)

Other	12 (9)
Which outcomes of COMMENCING gastrostomy feeding do you routinely discuss with people with MND? (N=139)	
Impact on quality of life	115 (83)
Impact on the person's weight	111 (80)
Time taken to finish meals	98 (71)
Risk of aspiration	95 (68)
Impact on caregivers (e.g. family)	87 (63)
Risk of choking	80 (58)
Estimated length of life remaining (prognosis)	30 (22)
I do not discuss outcomes of commencing gastrostomy feeding with people with MND	5 (4)
Other	10 (7)
Which outcomes of DECLINING gastrostomy feeding do you routinely discuss with people with MND? (N=139)	
Risk of aspiration	111 (80)
Impact on the person's weight	109 (78)
Risk of choking	106 (76)
Impact on quality of life	104 (75)
Time taken to finish meals	87 (63)
Impact on caregivers (e.g. family)	70 (50)
Estimated length of life remaining (prognosis)	50 (36)

I do not discuss outcomes of commencing gastrostomy feeding with people with MND	8 (6)
Other	10 (7)
What goals of gastrostomy feeding do you typically discuss with people with MND during decision making? (N=139)	
Weight maintenance/increase	103 (74)
Reduce risk of aspiration	107 (77)
Prevent choking	89 (64)
Provide an alternative route for medication administration	131 (94)
Reduce fatigue	105 (76)
Prolong life	32 (23)
Prevent/slow muscle wasting	42 (30)
Improve quality of life	107 (77)
Reduce caregiver burden	41 (30)
Improve caregivers quality of life	42 (30)
Reduce stress/burden at mealtimes for person with MND	132 (95)
I do not discuss goals of gastrostomy feeding with people with MND	3 (2)
Other	6 (4)
I do not inform people with MND about how long they will have to wait to have the gastrostomy placed	56 (40)
In your opinion, do healthcare professionals have a responsibility to give recommendations to people with MND about whether or not to have a gastrostomy placed? (N=139)	
Yes	72 (52)

No	67 (48)
Do you give recommendations to people with MND about whether or not they should have a gastrostomy placed?(N=139)	
Never	42 (30)
Rarely	29 (21)
Sometimes	38 (27)
Often	23 (17)
Always	7 (5)
When you believe the following decisions are best for a person with MND do you give recommendations that they should...	
...have a gastrostomy placed? (N=136)	
Never	39 (29)
Rarely	24 (18)
Sometimes	42 (31)
Often	21 (15)
Always	10 (7)
...continue without a gastrostomy tube? (N=137)	
Never	51 (37)
Rarely	27 (20)
Sometimes	49 (36)
Often	5 (4)
Always	5 (4)
...not ever have a gastrostomy placed? (N=135)	

Never	71 (53)
Rarely	30 (22)
Sometimes	28 (21)
Often	3 (2)
Always	3 (2)
Do you discuss the future withdrawal of enteral feeding during decision making discussions about gastrostomy placement? (N=139)	
Never	27 (19)
Rarely	26 (19)
Sometimes	43 (31)
Often	27 (19)
Always	16 (12)
Ideally, by what percentage (%) weight loss from symptom onset do you feel people with MND should have had a gastrostomy placed? (N=124)	
	Mean 8.1% SD 3.8 Range: 1-25%
Ideally, in relation to their respiratory function, when do you feel people with MND should have a gastrostomy placed? (N=138)	
Prior to presenting with any respiratory symptoms or objective signs of respiratory failure	63 (46)

When they have symptoms of respiratory failure but objective respiratory measures remain in normal range	39 (28)
When they have symptoms of respiratory failure and objective respiratory measures are outside normal range	6 (4)
When initiated on non-invasive ventilation (NIV)	2 (1)
When they are using non-invasive ventilation (NIV) for more than 8 hours per 24 hour period	1 (1)
When they are using non-invasive ventilation (NIV) for more than 16 hours per 24 hour period	0 (0)
I do not refer to markers of respiratory failure when making decisions regarding gastrostomy insertion	27 (20)
Ideally, in relation to their swallowing function, when do you feel people with MND should have a gastrostomy placed? (N=138)	
Prior to presenting with any swallowing difficulties	15 (11)
When they are noticing swallowing difficulty but not needing to modify the texture of their diet or fluids	50 (36)
When they need to modify the texture of diet or fluids but continue to manage adequate amounts	61 (44)
When they are presenting with adverse consequences of swallowing difficulties e.g. chest infections, choking episodes, reduced intake, prolonged mealtimes	8 (6)
When the consequences of swallowing difficulties present a significant risk to the respiratory health or nutritional intake of the person with MND	2 (1)
I do not refer to markers of swallow function when making decisions	2 (1)
In your opinion do most people with MND agree to gastrostomy placement: (N=137)	

Too early	0 (0)
About the right time	71 (52)
Too late	66 (48)
Do you encounter any challenges communicating with any members of the care team in your local area when supporting people with MND to make decisions about gastrostomy placement? (N=138)	
Yes	45 (33)
No	93 (67)
Do you experience these challenges specifically with any of the following professionals? (N=40)	
Neurologist	20 (50)
Palliative care doctor	4 (10)
Palliative care nurse	3 (8)
Gastroenterologist	12 (30)
Radiologist	0 (0)
Anaesthetist	1 (3)
Respiratory doctor	10 (25)
Respiratory physiotherapist	3 (8)
Respiratory nurse	2 (5)
Physiotherapist (non-respiratory specialist)	1 (3)
MND/neurology Specialist Nurse	8 (20)
MND coordinator	4 (10)
Community neurology matron	0 (0)

Home Enteral Feed Dietitian	2 (5)
Dietitian (non-home enteral feeding)	2 (5)
Speech and language therapist	4 (10)
Nutrition/gastrostomy nurse or advanced clinical practitioner	2 (5)
Occupational therapist	1 (1)
Psychologist	0 (0)
Social worker	0 (0)
General practitioner (GP)	10 (25)
Other	3 (8)
How effective do you believe the communication is between healthcare professionals in your local area when supporting people with MND to make decisions about gastrostomy placement? (N=139)	
Not effective at all	1 (1)
Slightly effective	10 (7)
Moderately effective	40 (29)
Very effective	72 (52)
Extremely effective	16 (12)
In your opinion, how consistent is the information given to people with MND about gastrostomy placement, by different members of your local MND care team? (N=138)	
Not at all consistent	5 (4)
Slightly consistent	16 (12)
Moderately consistent	60 (44)

Very consistent	48 (35)
Extremely consistent	9 (7)

Appendix 14. Survey proforma.

Supplementary information 2

Survey proforma. See below for a printed version of the questionnaire that was uploaded onto the Qualtrics online survey platform.

The survey is divided into 4 sections: The initiation of discussions about gastrostomy placement with people diagnosed with MND The process of making the decision about having a gastrostomy placed How your local care team collaborate to support people with MND considering gastrostomy placement Demographic questions about you and your professional experience (your answers to these questions are really important to allow the survey results to be analysed)

The following questions focus on when healthcare professionals **first introduce** gastrostomy feeding tube placement to people with MND.

Do you initiate discussions about gastrostomy placement with people with MND?

○ Yes (1)

○ No (2)

Which of the following factors prompt you to initiate discussions about gastrostomy placement with people with MND? (tick all that apply)

☐ Weight loss (1)

☐ Low body weight (9)

☐ Swallowing difficulties (2)

☐ Respiratory failure (3)

☐ Poor appetite (4)

☐ Dehydration (7)

☐ Poor lip seal (5)

☐ Upper limb weakness (6)

☐ Lower limb weakness (10)

☐ Prolonged mealtimes (12)

☐ Time and effort to prepare meals (13)

☐ Caregivers needing to assist people with MND to eat and drink (14)

☐ Difficulties taking medications (15)

☐ Person with MND requesting information on gastrostomy placement (16)

☐ Caregiver requesting information on gastrostomy placement (17)

☐ Other (8) ____

In most cases, **<u>WHEN DO YOU</u>** initiate discussions about gastrostomy placement with people with MND? (tick one option only)

○ Prior to the onset of any indications for gastrostomy (1)

○ Following the **first** presentation of **any** indications for gastrostomy (2)

○ When there is evidence that the indications for gastrostomy are progressing (3)

○ Other (please state) (4) ____

In most cases, when do you think gastrostomy placement **<u>SHOULD</u>** be first discussed with people with MND? (tick one option only)

○ Prior to the onset of any indications for gastrostomy (1)

○ Following the **first** presentation of **any** indications for gastrostomy (2)

○ When there is evidence of the indications for gastrostomy are progressing (3)

○ Other (please state) (4) ____

Do you have any local policies, care pathways or guidance that guides healthcare professionals about when to initiate discussions about gastrostomy placement to people with MND?

○ Yes (1)

○ No (2)

If you would you be happy to share your local guidance with the us, please email an electronic copy to smwhite1@sheffield.ac.uk (please note that your answers to the survey will remain anonymous)

The following questions focus on the process of making decisions about gastrostomy placement with people with MND.

Which of the following options do you routinely present to people with MND when discussing gastrostomy placement? (tick all that apply)

☐ They can have a gastrostomy placed (1)

☐ They can continue without a gastrostomy but can still choose to have a gastrostomy at a later date (2)

☐ They can decline to have a gastrostomy placed ever (3)

☐ I do not present any options (4)

☐ Other (please specify) (5)

☐

Which aspects of the procedure to place a gastrostomy do you routinely discuss with people with MND? (tick all that apply)

☐ The impact of respiratory failure on the risks associated with the procedure to place a gastrostomy (1)

☐ Detail about the procedure to place the gastrostomy (10)

☐ The sedation required during the procedure (11)

☐ Length of hospital stay (3)

☐ Risk of aspiration (4)

☐ Risk of not surviving gastrostomy placement (5)

☐ Pain post gastrostomy placement (6)

☐ Risk of infection post gastrostomy placement (7)

☐ Other (8) ____

☐ I do not discuss the procedure to place a gastrostomy with people with MND (9)

Which of the following issues relating to living with a gastrostomy tube do you routinely discuss with people with MND? (tick all that apply)

☐ Care of the gastrostomy (1)

☐ Care of the gastrostomy stoma site (2)

☐ Impact gastrostomy may have on body image (3)

☐ Later presenting tube complications and how they are managed e.g. blocked, damaged or displaced tubes (4)

☐ Later presenting stoma complications and how they are managed e.g. infection, overgranulation or leakage (14)

☐ Gastrointestinal side effects of gastrostomy feeding e.g. diarrhoea, constipation, bloating (5)

☐ The healthcare professional support they will receive once home with gastrostomy tube (7)

☐ Enteral feed methods available e.g. bolus and pump feeding (8)

☐ The impact gastrostomy feeding will have on caregivers (9)

☐ Identifying who will use and care for the tube post placement (10)

☐ The option to withdraw enteral feeding in the future (11)

☐ Other (12) ____

☐ I do not discuss living with a gastrostomy tube with people with MND (13)

Which outcomes of **COMMENCING** gastrostomy feeding do you routinely discuss with people with MND? (tick all that apply)

☐ Impact on the person's weight (1)

☐ Risk of aspiration (2)

☐ Risk of choking (3)

☐ Impact on quality of life (4)

☐ Time taken to finish meals (8)

☐ Estimated length of life remaining (prognosis) (5)

☐ Impact on caregivers (e.g. family) (9)

☐ Other (6) ____

☐ I do not discuss outcomes of commencing gastrostomy feeding with people with MND (7)

Which outcomes of **DECLINING** gastrostomy feeding do you routinely discuss with people with MND? (tick all that apply)

☐ Impact on the person's weight (1)

☐ Risk of aspiration (2)

☐ Risk of choking (3)

☐ Impact on quality of life (4)

☐ Time taken to finish meals (8)

☐ Estimated length of life remaining (prognosis) (5)

☐ Impact on caregivers (e.g. family) (9)

☐ Other (6) ____

☐ I do not discuss outcomes of not commencing gastrostomy feeding with people with MND (7)

What goals of gastrostomy feeding do you typically discuss with people with MND during decision making? (tick all that apply)

☐ Weight maintenance/increase (1)

☐ Reduce risk of aspiration (2)

☐ Prevent choking (3)

☐ Provide an alternative route for medication administration (4)

☐ Reduce fatigue (5)

☐ Prolong life (6)

☐ Prevent/slow muscle wasting (7)

☐ Improve quality of life (8)

☐ Reduce caregiver burden (9)

☐ Improve caregivers quality of life (10)

☐ Reduce stress/burden at mealtimes for person with MND (11)

☐ Other (12) ____

How long do you tell people with MND they will typically have to wait to have a gastrostomy placed following referral? (tick one option only)

○ 1 week (1)

○ 2 weeks (2)

○ 3 weeks (3)

○ 4 weeks (4)

○ 5 weeks (5)

○ 6 weeks (6)

○ 7 weeks (7)

○ 8 weeks (8)

○ If more than 8 weeks please state (9)

____

○ I do not inform people with MND about how long they will have to wait to have the gastrostomy placed (10)

What is the average period of time you inform people with MND they will be in hospital for while having their gastrostomy placed? (tick one option only)

○ 0 days (i.e. gastrostomy placed as a day-case) (1)

○ 1 day (2)

○ 2 days (3)

○ 3 days (4)

○ 4 days (5)

○ 5 days (6)

○ 6 days (7)

○ 7 days (8)

○ >7 days (please state average number of days you give) (9)

____

○ I do not discuss length of hospital stay (10)

What information sources about the decision to have a gastrostomy placed do you direct people with MND to? (tick all that apply)

☐ Locally produced patient information leaflet about gastrostomy placement (1)

☐ Locally produced patient information leaflet about home enteral feeding (2)

☐ Mytube website (3)

☐ MND Association eating and drinking guide (4)

☐ MND Association tube feeding leaflet (5)

☐ Parenteral and Enteral Nutrition Specialist Group of the British Dietetic Association (PENG) enteral feeding leaflet: 'Enteral tube feeding – your questions answered' (6)

☐ PENG leaflet: 'Having an enteral feeding tube – further questions to ask (7)

☐ Patients on Intravenous and Naso-gastric Nutrition Treatment (PINNT) (8)

☐ I do not provide any information resources (9)

☐ Other (10) ____

Do you practically demonstrate the following equipment to people with MND when discussing gastrostomy placement? (tick all that apply)

☐ The gastrostomy tube (1)

☐ Enteral feeding syringes (2)

☐ Enteral feeding pump, stand and giving sets (3)

☐ Other (please specify) (4) ____

☐ I do not demonstrate any of the above equipment (5)

☐ I refer to another healthcare professional or service to demonstrate the equipment to people with MND (please specify the professional you refer the patient to) (6) ____

How often do you encourage people with MND considering gastrostomy placement to meet other people with MND?

○ Never (6)

○ Rarely (7)

○ Sometimes (8)

○ Often (9)

○ Always (10)

The following questions focus on healthcare professionals' recommendations to people with MND. For the purposes of these questions, 'recommendations' are defined as:
‘when healthcare professionals give opinions to people with MND about whether or not they should have a gastrostomy placed.’

In your opinion, do healthcare professionals have a responsibility to give recommendations to people with MND about whether or not to have a gastrostomy placed? (tick one option only)

○ Yes (1)

○ No (2)

Do you give recommendations to people with MND about whether or not they should have a gastrostomy placed? (tick one option only)

- ○ Never (35)
- ○ Rarely (36)
- ○ Sometimes (37)
- ○ Often (38)
- ○ Always (39)

Do people with MND ask you to give a recommendation about whether or not they should have a gastrostomy tube placed? (tick one option only)

- ○ Never (11)
- ○ Rarely (12)
- ○ Sometimes (13)
- ○ Often (14)
- ○ Always (15)

When you believe the following decisions are best for a person with MND do you give recommendations that they should...

	Never (6)	Rarely (7)	Sometimes (8)	Often (9)	Always (10)
...have a gastrostomy placed? (1)	○	○	○	○	○

...continue without a gastrostomy tube? (2)	○	○	○	○	○
...not ever have a gastrostomy placed? (3)	○	○	○	○	○

Do you discuss the future withdrawal of enteral feeding during decision making discussions about gastrostomy placement? (tick on option only)

- ○ Never (11)
- ○ Rarely (12)
- ○ Sometimes (13)
- ○ Often (14)
- ○ Always (15)

Have you attended training or study events on the following topics? (tick all that apply)

- ☐ MND disease course (1)
- ☐ Supporting people making decisions about interventions (3)
- ☐ Nutritional management of people with MND (4)
- ☐ Gastrostomy placement procedures (5)
- ☐ Communication skills (6)

On average how long will you typically spend discussing gastrostomy placement with a person with MND? (tick one option only)

○ 0-15 mins (1)

○ 16-30 mins (2)

○ 31-45 mins (3)

○ 46-60 mins (4)

○ >60 mins (5)

Do you feel you have enough time to discuss gastrostomy placement during consultations with people with MND? (tick one option only)

○ Yes (3)

○ No (4)

Please explain why you feel you do not have enough time to discuss gastrostomy placement during consultations with people with MND

____

____

The following 3 questions relate to the timing of gastrostomy placement i.e. when you feel people with MND **should have had a gastrostomy tube placed by**, in relation to their weight, respiratory function and swallowing ability. Please assume when answering these questions that the person with MND has agreed to gastrostomy placement and is now deciding **WHEN** they should have it placed.

Ideally, by **what percentage (%) weight loss from symptom onset** do you feel people with MND should have had a gastrostomy placed? (choose from drop down list)

▼ 1 (1) ... 100 (100)

Ideally, in relation to their respiratory function, when do you feel people with MND should have a gastrostomy placed? (tick one option only)

○ Prior to presenting with any respiratory symptoms or objective signs of respiratory failure (1)

○ When they have symptoms of respiratory failure but objective respiratory measures remain in normal range (2)

○ When they have symptoms of respiratory failure and objective respiratory measures are outside normal range (3)

○ When initiated on non-invasive ventilation (NIV) (4)

○ When they are using non-invasive ventilation (NIV) for more than 8 hours per 24 hour period (5)

○ When they are using non-invasive ventilation (NIV) for more than 16 hours per 24 hour period (6)

○ I do not refer to markers of respiratory failure when making decisions regarding gastrostomy insertion (7) Q128 Ideally, in relation to their swallowing function, when do you feel people with MND should have a gastrostomy placed? (tick one option only)

○ Prior to presenting with any swallowing difficulties (1)

○ When they are noticing swallowing difficulty but not needing to modify the texture of their diet or fluids (2)

○ When they need to modify the texture of diet or fluids but continue to manage adequate amounts (3)

○ When they are presenting with adverse consequences of swallowing difficulties e.g. chest infections, choking episodes, reduced intake, prolonged mealtimes (4)

○ When the consequences of swallowing difficulties present a significant risk to the respiratory health or nutritional intake of the person with MND (5)

○ I do not refer to markers of swallow function when making decisions (6)

In your opinion do most people with MND agree to gastrostomy placement:

○ Too early (1)

○ About the right time (2)

○ Too late (3)

In your opinion, to what extent are people with MND placed under pressure to agree to gastrostomy placement by:

	Never (11)	Rarely (12)	Sometimes (13)	Often (14)	Always (15)
Healthcare professionals (1)	○	○	○	○	○
Family members (2)	○	○	○	○	○

The following question focuses on how different healthcare professionals work together to support people with MND making decisions about gastrostomy placement. When questions state **'in your local area'**, this refers to the other healthcare professionals who are involved in discussions with people with MND that you also support including those in community teams and MND clinics.

Which healthcare professionals are involved in supporting people with MND to make decisions about gastrostomy placement in your local area (tick all that apply)?

☐ Neurologist (1)

☐ Palliative care doctor (2)

☐ Palliative care nurse (3)

☐ Gastroenterologist (4)

☐ Radiologist (5)

☐ Anaesthetist (6)

☐ Respiratory doctor (7)

☐ Respiratory physiotherapist (11)

☐ Respiratory nurse (21)

☐ Physiotherapist (non-respiratory specialist) (18)

☐ MND/neurology Specialist Nurse (8)

☐ MND coordinator (19)

☐ Community neurology matron (22)

☐ Home Enteral Feed Dietitian (10)

☐ Dietitian (non-home enteral feeding) (9)

☐ Speech and language therapist (12)

☐ Nutrition/gastrostomy nurse or advanced clinical practitioner (13)

☐ Occupational therapist (15)

☐ Psychologist (16)

☐ Social worker (17)

☐ General practitioner (GP) (14)

☐ Other (please specify) (20)

____

What guidance do you refer to for supporting people with MND to make decisions about gastrostomy placement? (tick all that apply)

☐ NICE MND guideline [NG42] (1)

☐ European Federation of Neurological Societies (EFNS) guidelines (2)

☐ American Academy of Neurology (AAN) 'Care of patient with ALS' guideline (3)

☐ NICE nutrition support clinical guideline [CG32] (4)

☐ ESPEN guideline clinical nutrition in neurology (5)

☐ NICE shared decision making guideline [NG197] (6)

☐ Locally developed guideline (7)

☐ Other (please specify) (8)

____

☐ I do not refer to any guidance (14)

If you would you be happy to share your locally developed guideline with the us, please email an electronic copy to smwhite1@sheffield.ac.uk (please note that your answers to the survey will remain anonymous)

How useful is the available guidance at informing your practice when supporting people with MND to make decisions about gastrostomy placement? (tick one option only)

○ Extremely useful (1)

○ Very useful (2)

○ Moderately useful (3)

○ Slightly useful (4)

○ Not at all useful (5)

How often are you aware of the discussions other healthcare professionals have had with people with MND about gastrostomy placement? (tick one option only)

○ Never (6)

○ Rarely (7)

○ Sometimes (8)

○ Often (9)

○ Always (10)

Do you communicate with other healthcare professionals **AFTER** having discussions with people with MND about gastrostomy placement? (tick one option only)

○ Never (10)

○ Rarely (11)

○ Sometimes (12)

○ Often (13)

○ Always (14)

How do you communicate with other healthcare professionals? (tick all that apply)

☐ Letter (1)

☐ Email (2)

☐ Telephone (3)

☐ In MDT meetings (4)

☐ Via message function in electronic patient records (5)

☐ Other (6) ____

☐ I do not communicate with other healthcare professionals after discussions with people with MND about gastrostomy placement (7)

In your opinion, how important is it for healthcare professionals to communicate with other members of the care team **AFTER** they have had discussions about gastrostomy placement with people with MND? (tick one option only)

○ Extremely important (1)

○ Very important (2)

○ Moderately important (3)

○ Slightly important (4)

○ Not at all important (5)

What communication do you send to the person with MND following discussions about gastrostomy placement? (tick all that apply)

☐ Copy person with MND into letter sent to healthcare professionals (1)

☐ Copy person with MND into emails sent to healthcare professionals (2)

☐ Send individual letter or email to person with MND summarising content of discussion and decisions made (3)

☐ Update a patient held record (4)

☐ I do not communicate with the person with MND outside of direct contact e.g. clinic, home visit, telephone or video call (5)

☐ Other (6) ____

Do you encounter any challenges communicating with any members of the care team in your local area when supporting people with MND to make decisions about gastrostomy placement?

○ Yes (85)

○ No (86)

Do you experience these challenges specifically with any of the following professionals? (tick all that apply)

☐ Neurologist (1)

☐ Palliative care doctor (2)

☐ Palliative care nurse (3)

☐ Gastroenterologist (4)

☐ Radiologist (5)

☐ Anaesthetist (6)

☐ Respiratory doctor (7)

☐ Respiratory Physiotherapist (11)

☐ Respiratory nurse (21)

☐ Physiotherapist (non-respiratory specialist) (22)

☐ MND/neurology Specialist Nurse (8)

☐ MND coordinator (23)

☐ Community neurology matron (24)

☐ Home Enteral Feed Dietitian (10)

☐ Dietitian (non-home enteral feeding) (9)

☐ Speech and language therapist (12)

☐ Nutrition/gastrostomy nurse or advanced clinical practitioner (13)

☐ Occupational therapist (15)

☐ Psychologist (16)

☐ Social worker (17)

☐ General Practitioner (GP) (19)

☐ Other (please specify) (20) ____

Please describe any challenges you experience.

____

How effective do you believe the communication is between healthcare professionals in your local area when supporting people with MND to make decisions about gastrostomy placement? (tick one option only)

○ Not effective at all (1)

○ Slightly effective (2)

○ Moderately effective (3)

○ Very effective (4)

○ Extremely effective (6)

Do you attend meetings (e.g. multidisciplinary team (MDT) meetings) with other healthcare professionals where you discuss people with MND making decisions about gastrostomy? (tick one option only)

○ Yes (3)

○ No (4)

Are there any professionals or professional teams that you feel are missing from these meetings who are involved in discussions about gastrostomy placement? (tick one option only)

○ Yes (3)

○ No (4)

In your opinion which professionals or teams are missing from your MDT meetings? (tick all that apply)

☐ Neurologist (1)

☐ Palliative care team (2)

☐ Gastrostomy placement team (4)

☐ Anaesthetist (6)

☐ Respiratory team (7)

☐ MND/neurology Specialist Nurse or coordinator (8)

☐ Home Enteral Feed Team (10)

☐ Dietitian (non-home enteral feeding) (13)

☐ Physiotherapist (non-respiratory specialist) (11)

☐ Speech and language therapist (12)

☐ Enteral feeding advanced clinical practitioner (14)

☐ Occupational therapist (15)

☐ Psychologist (16)

☐ Social worker (17)

☐ General practitioner (GP) (19)

☐ Ward nurse (18)

☐ Other (please specify) (20)

____

How often do the MDT meetings take place? (tick one option only)

○ Weekly (1)

○ Every two weeks (2)

○ Every 3 weeks (3)

○ Every month (4)

○ Every 2 months (5)

○ Every 3 months (6)

○ Other (please specify) (7) ____

In your opinion, how consistent is the information given to people with MND about gastrostomy placement, by different members of your local MND care team?

○ Extremely consistent (1)

○ Very consistent (2)

○ Moderately consistent (3)

○ Slightly consistent (4)

○ Not at all consistent (5)

What is your sex?

○ Male (1)

○ Female (2)

○ Non-binary / third gender (3)

○ Prefer not to say (4)

○ Other (please specify) (5) ____

What is your profession? (tick one option only)

○ Doctor (1)

○ Nurse (2)

○ Dietitian (3)

○ Speech and language therapist (4)

○ Occupational therapist (5)

○ Physiotherapist (6)

○ Social worker (7)

○ Psychologist (8)

○ Other (9) ____

What is your professional role?

○ Neurologist (1)

○ Palliative care doctor (2)

○ Palliative care nurse (3)

○ Gastroenterologist (4)

○ Radiologist (5)

○ Anaesthetist (6)

○ Respiratory doctor (7)

○ Respiratory physiotherapist (11)

○ Respiratory nurse (21)

○ Physiotherapist (non-respiratory specialist) (22)

○ MND/neurology Specialist Nurse (8)

○ MND coordinator (23)

○ Community neurology matron (24)

○ Home Enteral Feed Dietitian (10)

○ Dietitian (non-home enteral feeding) (9)

○ Speech and language therapist (12)

○ Nutrition/gastrostomy nurse (13)

○ Enteral feeding advanced clinical practitioner (14)

○ Occupational therapist (15)

○ Psychologist (16)

○ Social worker (17)

○ Ward nurse (18)

○ Other (please specify) (20)

____

○ General practitioner (GP) (19)

During a typical working week, how much of your time involves supporting people with MND? (tick one option only)

○ None of your time (1)

○ Very little of your time (2)

○ Some of your time (3)

○ The majority of your time (4)

○ All of your time (5)

Approximately, how many people with MND do you currently have on your caseload? (tick one option only)

○ 0-10 (1)

○ 11-20 (2)

○ 21-30 (3)

○ 31-40 (4)

○ 41-50 (5)

○ 51-60 (6)

○ 61-70 (7)

○ 71-80 (8)

○ 81-90 (9)

○ 91-100 (10)

○ >100 (11)

What percentage of your current clinical caseload is people with MND?

○ 0-19% (1)

○ 20-39% (2)

○ 40-59% (3)

○ 60-79% (4)

○ 80-99% (5)

○ 100% (6)

How many years experience do you have caring for people with MND? (choose number from drop down box)

▼ 1 (1) ... >60 (61)

In what setting do you see people with MND when having discussions about gastrostomy placement? (tick all that apply)

☐ Hospital ward (1)

☐ Hospital based out-patient clinic (2)

☐ Hospice based out-patient clinic (3)

☐ Community based out-patient clinic (4)

☐ Domiciliary visit (5)

☐ Telephone calls (6)

☐ Video call (7)

☐ Other (8) ____

We are planning a subsequent study that will involve observing how people with MND are supported to make decisions about gastrostomy placement by their healthcare professionals, including interviews of people with MND, caregivers and healthcare professionals. If you are interested in being involved in this study please email the lead researcher at smwhite1@sheffield.ac.uk.

By clicking on the 'Submit' button, your survey will be submitted and you will no longer be able to change your answers.

Appendix 15 Evidence for how the study met the requirements of the CHERRIES checklist (Eysenbach 2004).

Supplementary information 1 from Paper 2.

Item category	*Checklist item*	*Explanation*
Design	Describe survey design	A rigorous approach was taken to developing and testing the survey prior to launch. Target population clearly identified as any UK HCP who supports pwMND to make decisions about gastrostomy placement. In the absence of a defined sampling frame a convenience sample was obtained using a snowball sampling strategy.
Ethics	Ethics board approval	Ethical approval was granted by the University of Sheffield ethics board and approval also given by HRA
	Informed consent	The participant information sheet (PIS), screening questions and consent form were embedded into the online survey. The PIS included information about the study, how long it would take to complete and how their data would be used. Potential participants were required to complete a screening question to confirm they met the inclusion criteria for

		the study. Finally, the participants were required to confirm they agreed with statements within a consent form prior to entering the survey. Participants were made aware that only submitted surveys would be included in the analysis.
	Data protection	No participant identifiable information was collected within the survey.
Development and pre-testing	Development and testing	The draft survey instrument was uploaded onto Qualtrics and piloted with 9 HCPs from the target population. The pilot participants completed a structured feedback form which informed the final changes made to the survey prior to launch.
Recruitment process and description of the sample having access to the questionnaire	Open survey versus closed survey	The survey was open to anyone who had the link to the Qualtrics survey.
	Contact mode	A snowball sampling strategy was used. This included gatekeeper organisations and groups forwarding an email including a brief summary of the study and the link to the survey to their members. The gatekeepers and lead researcher also shared the survey link using social media.

	Advertising the survey	The link to the survey was shared by gatekeepers and the lead researcher via email and social media. The MND Association, British Association of Parenteral and Enteral Nutrition (BAPEN) and the Parenteral and Enteral Specialist Group of the British Dietetic Association (PENG) also shared the link to the survey on their websites.
Survey administration	Web/email	Qualtrics online survey software was used to host the survey.
	Context	The link to the survey was disseminated via email and social media, and via news stories on MND Association, BAPEN and PENG websites.
	Mandatory/voluntary	Potential participants were made explicitly aware that it was completely voluntary for them to complete the survey. Participants did not have to answer every question.
	Incentives	No incentives were offered to complete the survey.
	Time/date	The original plan was for the survey to remain open for 8 weeks. However, the closing date was extended by 2 weeks as the target sample size had not been achieved by the end of the 8 weeks. The survey was open between 13/6/2022 and 30/8/2022.

	Randomisation of items or questionnaires	No randomisation of items or questionnaires was used in this survey.
	Adaptive questioning	Some conditional branching was used within the survey to only show certain questions dependent on answers to previous questions (e.g. questions about initiating discussions with pwMND about gastrostomy were only displayed to those participants who stated start such discussions). The main purpose of this was to reduce the number of questions being asked and to support validity of the study.
	Number of items	Only one question was asked per page of the survey. The survey included a maximum of 56 questions in total.
	Number of screens (pages)	The survey was distributed over 56 pages (screens).
	Completeness check	The survey did not include any completeness check allowing participants to not complete questions if they did not wish to rather than including an option to state they do not wish to answer the question. Participants were made aware in the PIS that they did not have to provide a response to every question. No questions were mandatory.

	Review step	Respondents were able to move backwards through their survey and alter their responses if they wished to.
Response rates	Unique site visitor	No steps were made to determine if participants completing the survey were unique. This would raise the risk of the same participant completing the survey more than once.
	View rate	Unique site visitors not recorded
	Participation rate	The participants were required to reach the end of the survey and submit their survey to be included in the study
	Completion rate	139 participants submitted the survey out of a total of 212 who confirmed that they met the inclusion criteria for the study.
Preventing multiple entries from the same individual		No steps were taken to ensure that multiple entries were recorded from the same individual.

Analysis	Handling of incomplete questionnaires	Only questionnaires that were submitted by the participant were included in the analysis. Within the reporting it is made clear how many participants answered each question.
	Questionnaires submitted with an atypical timestamp	No timeframe was implemented for which participants should take to complete the questionnaire.
	Statistical correction	No weighting or propensity scores were used to adjust for a non-representative sample.

Appendix 16. List of gatekeepers for distributing the survey link.

Supplementary information 3 from paper 2.

Below is a list of the organisations who assisted in disseminating the link to their members who may have met the inclusion criteria for the study:

- MND Clinical Studies Group (MNDCSG)
- Association of palliative medicine
- British Association of parenteral and enteral nutrition (BAPEN)
- British Dietetic Association (BDA)
- Parenteral and enteral specialist group of the BDA (PENG)
- Neurosciences specialist group of the BDA (NSG)
- PENG Virtual HEF Group
- MND Association
- National Nurse Nutrition Group (NNNG)

Appendix 17. Characteristics of survey participants.

Supplementary information 4 from Paper 2.

Question	N (%)
What is your sex? (n=139)	
Male	14 (10)
Female	123 (89)
Prefer not to say	2 (1)
Other (please specify)	0 (0)
What is your profession? (n=139)	

Doctor	17 (12)
Nurse	23 (17)
Dietitian	73 (53)
Speech and language therapist	19 (14)
Occupational therapist	3 (2)
Physiotherapist	4 (3)
What is your professional role? (n=138)	
Neurologist	4 (3)
Palliative care doctor	7 (5)
Gastroenterologist	2 (1)
Respiratory doctor	4 (3)
Respiratory physiotherapist	1 (1)
Respiratory nurse	1 (1)
Physiotherapist (non-respiratory specialist)	3 (2)
MND/neurology Specialist Nurse	12 (9)
MND coordinator	2 (1)
Home Enteral Feed Dietitian	42 (30)
Dietitian (non-home enteral feeding)	30 (22)
Speech and language therapist	18 (13)
Nutrition/gastrostomy nurse	8 (6)
Enteral feeding advanced clinical practitioner	2 (1)
Occupational therapist	2 (1)

During a typical working week, how much of your time involves supporting people with MND? (n=139)	
None of your time	3 (2)
Very little of your time	19 (14)
Some of your time	88 (63)
The majority of your time	14 (10)
All of your time	15 (11)
Approximately, how many people with MND do you currently have on your caseload? (n=138)	
0-10	61 (44)
11-20	30 (22)
21-30	16 (12)
31-40	6 (4)
41-50	2 (1)
51-60	3 (2)
61-70	0 (0)
71-80	3 (2)
81-90	2 (1)
91-100	2 (1)
>100	13 (9)
What percentage of your current clinical caseload is people with MND? (n=138)	
0-19%	86 (62)

20-39%	22 (16)
40-59%	10 (7)
60-79%	1 (1)
80-99%	5 (4)
100%	14 (10)
How many years experience do you have caring for people with MND? (n=83)	
Mean 11.1 years (SD 8.3) Range 1-39 year	
In what setting do you see people with MND when having discussions about gastrostomy placement? (n=139)	
Hospital ward	59 (42)
Hospital based out-patient clinic	60 (43)
Hospice in-patient	3 (2)
Hospice based out-patient clinic	20 (14)
Community based out-patient clinic	36 (26)
Domiciliary visit	92 (66)
Telephone calls	69 (50)
Video call	39 (28)
Other	2 (1)

Appendix 18. Formation of new categories about the timing of gastrostomy placement in relation to severity of dysphagia and respiratory failure.

Supplementary information 6 from the published Paper 2.

The categories to two questions about participants beliefs about when pwMND should have a gastrostomy placed in relation to the severity of their respiratory failure and dysphagia were combined to allow for the statistical comparisons to be made. The original question and answer categories are displayed in supplementary table 2 below.

Original data collected.

Ideally, in relation to their respiratory function, when do you feel people with MND should have a gastrostomy placed? (tick one option only) (N=138)	n (%)
Prior to presenting with any respiratory symptoms or objective signs of respiratory failure	63 (46)
When they have symptoms of respiratory failure but objective respiratory measures remain in normal range	39 (28)
When they have symptoms of respiratory failure and objective respiratory measures are outside normal range	6 (4)
When initiated on non-invasive ventilation (NIV)	2 (1)
When they are using non-invasive ventilation (NIV) for more than 8 hours per 24 hour period	1 (1)
When they are using non-invasive ventilation (NIV) for more than 16 hours per 24 hour period	0 (0)
I do not refer to markers of respiratory failure when making decisions regarding gastrostomy insertion	27 (20)
Ideally, in relation to their swallowing function, when do you feel people with MND should have a gastrostomy placed? (tick one option only) (N=138)	
Prior to presenting with any swallowing difficulties	15 (11)

When they are noticing swallowing difficulty but not needing to modify the texture of their diet or fluids	50 (36)
When they need to modify the texture of diet or fluids but continue to manage adequate amounts	61 (44)
When they are presenting with adverse consequences of swallowing difficulties e.g. chest infections, choking episodes, reduced intake, prolonged mealtimes	8 (6)
When the consequences of swallowing difficulties present a significant risk to the respiratory health or nutritional intake of the person with MND	2 (1)
I do not refer to markers of swallow function when making decisions	2 (1)

The new categories formed to allow the comparative statistics to be undertaken.

Question category from original survey	New category
When participants believed gastrostomy tubes should be placed in relation to respiratory function	
Prior to presenting with any respiratory symptoms or objective signs of respiratory failure	Prior to any respiratory symptoms
When they have symptoms of respiratory failure but objective respiratory measures remain in normal range AND When they have symptoms of respiratory failure and objective respiratory measures are outside normal range	Compensating for early respiratory symptoms i.e. pre-NIV
When initiated on non-invasive ventilation (NIV) AND When they are using non-invasive ventilation (NIV) for more than 8 hours per 24 hour period AND When they are using non-invasive ventilation (NIV) for more than 16 hours per 24 hour period	Experiencing significant consequences i.e. on NIV
When participants believed gastrostomy tubes should be placed in relation to swallowing function	

Prior to presenting with any swallowing difficulties	Prior to any swallowing problems
When they are noticing swallowing difficulty but not needing to modify the texture of their diet or fluids AND When they need to modify the texture of diet or fluids but continue to manage adequate amounts	Compensating for impact of dysphagia e.g. through texture modified diet
When they are presenting with adverse consequences of swallowing difficulties e.g. chest infections, choking episodes, reduced intake, prolonged mealtimes AND When the consequences of swallowing difficulties present a significant risk to the respiratory health or nutritional intake of the person with MND	Experiencing significant consequences e.g. chest infections, choking episodes

Appendix 19. The topics discussed by the different professions in relation to the placement of a gastrostomy and life on enteral feeding.

Supplementary information 7 from Paper 2.

The topics discussed by the different professions in relation to the placement of a gastrostomy and life on enteral feeding. * = indicates a significant difference ($p \leqslant 0.05$) between the responses of different professions. The professional role most reporting a topic is indicated in **bold**.

Topic discussed with pwMND during decision making…	**Percentage of HCP discipline stating they discuss the topic (n (%)) N=132**				
	Nurse (N=23)	Dietitian (N=73)	Doctor (N=17)	SLT (N=19)	p
…in relation to the placement of a gastrostomy					

The impact of respiratory failure on the procedural risks	18(78)	49(67)	**16(94)**	8(42)	0.006*
Detail about the procedure to place the gastrostomy	19(83)	**61(84)**	13(77)	11(58)	0.102
The sedation required during the procedure	**18(78)**	42(58)	9(53)	6(32)	0.025*
The length of hospital stay	**22(96)**	55(75)	11(65)	9(47)	0.004*
The risk of aspiration during the procedure	**19(83)**	41(56)	6(35)	13(68)	0.017*
The risk of not surviving the procedure	12(52)	28(38)	**10(59)**	2(11)	0.012*
Pain post gastrostomy placement	**20(87)**	50(69)	10(59)	4(21)	<0.001*
Risk of infection post gastrostomy placement	**18(78)**	50(69)	7(41)	11(58)	0.077
…in relation to living with a gastrostomy					
Care of the gastrostomy	21(91)	**66(90)**	5(29)	9(47)	<0.001*
Care of gastrostomy stoma site	19(83)	**62(85)**	4(24)	3(16)	<0.001*

Impact gastrostomy may have on body image	**18(78)**	33(45)	5(29)	3(16)	<0.001*
Tube complications and how they are managed	13(57)	**59(81)**	3(18)	5(26)	<0.001*
Stoma complications and how they are managed	13(57)	**59(81)**	3(18)	5(26)	<0.001*
Gastrointestinal side effects of gastrostomy feeding	11(48)	**41(56)**	2(12)	5(26)	<0.001*
The HCP support for pwMND at home	**22(96)**	68(93)	9(53)	14(74)	<0.001*
Enteral feeding methods	22(96)	**71(97)**	10(59)	12(63)	<0.001*
The impact gastrostomy feeding will have on caregivers	**19(83)**	54(74)	4(24)	9(47)	<0.001*
Identifying who will use and care for the tube post placement	21(91)	**70(96)**	8(47)	11(58)	<0.001*
The option to withdraw enteral feeding in the future	14(61)	**48(66)**	8(47)	6(32)	0.043*

The outcomes discussed by different professions in relation to when discussing **accepting** gastrostomy placement. * = indicates a significant difference ($p \leqslant 0.05$) between the responses of different professions. The professional role most reporting a topic is indicated in **bold**.

Outcome of gastrostomy feeding discussed	**Discussed in relation to accepting gastrostomy (n (%)) N=132**				**p**
	Nurses	Dietitians	Doctors	SLT	
Impact on the person's weight	18(78)	**65(89)**	11(65)	12(63)	0.021*
Risk of aspiration	**17(74)**	51(70)	10(59)	12(63)	0.717
Risk of choking	**16(70)**	40(55)	10(59)	10(53)	0.618
Impact on quality of life	**20(87)**	62(85)	12(71)	14(74)	0.366
Time taken to finish meals	15(65)	**52(71)**	11(65)	13(68)	0.925
Estimated length of life remaining (prognosis)	5(22)	10(14)	**8(47)**	5(26)	0.023*
Impact on caregivers (e.g. family)	15(65)	**50(69)**	8(47)	10(53)	0.294
Do not discuss outcomes of accepting gastrostomy	1(4)	2(3)	1(6)	1(5)	0.906

The outcomes discussed by different professions in relation to when discussing **declining** gastrostomy placement. * = indicates a significant difference (p⩽0.05) between the responses of different professions. The professional role most reporting a topic is indicated in **bold**.

Outcome of gastrostomy feeding discussed	**Discussed in relation to declining gastrostomy (n (%)) N=132**				**p**
	Nurses	Dietitians	Doctors	SLT	
Impact on the person's weight	**19(83)**	58(80)	11(65)	14(74)	0.524
Risk of aspiration	**21(91)**	55(75)	12(71)	17(90)	0.192
Risk of choking	**20(87)**	53(73)	12(71)	16(84)	0.395
Impact on quality of life	**21(91)**	50(69)	12(71)	14(74)	0.191
Time taken to finish meals	13(57)	**49(67)**	8(47)	12(63)	0.435
Estimated length of life remaining (prognosis)	11(48)	17(23)	**12(71)**	6(32)	0.001*
Impact on caregivers (e.g. family)	**14(61)**	32(44)	9(53)	11(58)	0.435
Do not discuss outcomes of declining gastrostomy	2(8.7)	5(7)	1(5.9)	0(0)	0.622

This answers to each question are represented in the main manuscript as table 3 (repeated below), indicating that severity of respiratory failure is a greater driver of participants beliefs about gastrostomy timing than severity of dysphagia. The data in the shaded boxes include those participants who believed that pwMND should have a gastrostomy placed at a similar severity stage i.e. 13 participants believed that gastrostomy should be placed prior to any respiratory symptoms and that gastrostomy should be placed prior to any swallowing problems. The data under the shaded rows includes those participants who believed that pwMND should have gastrostomy tubes placed earlier in relation to respiratory failure than in relation to dysphagia e.g. 48 participants believed that pwMND should have gastrostomy tubes removed prior to any respiratory symptoms but once having to compensate for the impact of dysphagia.

Appendix 20. Medical note review proforma.

Study ID:	Date of entry: / /
Date of data collection: / /	Date of documentation entry: / /
Data source	
Profession documenting entry	
Employer of healthcare professional	
Nature of documentation (e.g. patient/HCP interaction; administration)	
Location of interaction (e.g. clinic, ward, telephone, video call)	
Data extract	

Appendix 21. Scoping email sent to potential cases.

Subject heading: Expressions of interest for a study about gastrostomy decision making in MND care

Hi

We are contacting you to ask if you would like to express an interest in becoming a study site for research we are planning. The research aims to understand how healthcare professionals, as individuals and part of multidisciplinary teams, support people with motor neuron disease (MND) to make decisions about gastrostomy feeding tube placement in real-world practice. The research is part of the my NIHR funded Clinical Doctoral Research Fellowship. The research will involve me recruiting people with MND, their caregivers and supporting healthcare professionals to take part in a variety of research methods, focusing on how decisions about gastrostomy placement are made. I have attached a brief summary of what would be involved during study. This research will be repeated in three different study sites in the UK.

Key requirements to become a study site are that you have a MND clinic hosted within the organisation and there is a healthcare professional who is willing to act as a gatekeeper (and principal investigator) for the study site. This person would be my main link with the study site and help approach other healthcare professionals who are involved in supporting people with MND to make decisions about gastrostomy to take part in the research. If you feel this may not be a role you could take on personally, you are welcome to forward this email and attached information to the person who could take on this role e.g. the local MND coordinator or nurse.

If you wish to express an interest in being a study site please reply to me at smwhite1@sheffield.ac.uk. I will be in touch shortly to have further discussions with those expressing an interest in becoming involved in the study. Please get back to me if you require any further information or clarifications. Thank you for your time reading this information.

Kind regards

Sean

Sean White

NIHR Doctoral Fellow

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Appendix 22. Screening email sent to potential cases during case selection process.

Background

People with MND can experience many challenges taking adequate oral diet and fluids explaining the high incidence of malnutrition. Placement of a gastrostomy feeding tube is routinely offered to people with MND who may become unable to meet their nutritional requirements via the oral route. Published research describes the challenges associated with making the decision to have a gastrostomy placed and when to have it, from the perspective of people with MND, their caregivers and supporting healthcare professionals.

The Chief Investigator is undertaking research as part of his NIHR doctoral fellowship. The research plan has been informed by a systematic review of qualitative research understanding the factors that influence decisions about invasive interventions in MND care and a survey of the practice and attitudes of UK healthcare professionals towards supporting people with MND with making the decision to have a gastrostomy placed. While developing the research plan the lead researcher has collaborated with an experienced supervisory team, an expert panel of healthcare professionals and academics and a panel of people with MND and caregivers with experience of making the decision to have a gastrostomy placed.

What is the aim of the doctoral research

To understand how healthcare professionals, as individuals and part of multidisciplinary teams, support people with MND to make decisions about gastrostomy feeding tube placement.

Multiple case study

This research will use a multiple case study design. Case study research involves researching a 'phenomenon (the "case") in depth and within its real-world context, when the boundaries between phenomenon and context may not be clearly evident'. In relation to the proposed research the 'case/phenomenon' are the decisions people with MND make about gastrostomy placement, and the 'context' is the decision support provided by the MND multidisciplinary team (MDT). Data is planned to be collected from three different case study sites, each centred around their local MND service.

What would be involved if we agreed to be a case study site?

Firstly, a healthcare professional would be identified who would act as the Principal Investigator (PI) and gatekeeper for the case study site. The PI would have the following responsibilities:

- Be the Chief Investigators main link with the case study site.
- Join a one hour telephone or online call with the Chief Investigator to discuss the study protocol, recruitment plan and discuss the structure and location of the local MND service.
- Forwarding an email provided by the Chief Investigator, to healthcare professionals who are known to support people with MND making decisions about gastrostomy placement.
- Assist in identifying people with MND who are considering or have made a recent decision about gastrostomy placement (up to a month post gastrostomy placement)
- Maintain an electronic site study file.

The Chief Investigator will offer to deliver an online presentation, including a question and answer session, for healthcare professionals who wish to learn more about the study. This session will be recorded and made available to any healthcare professionals who are unable to attend the 'live session'.

The healthcare professionals on the case study site will be asked to inform potential participants (i.e. people with MND and other healthcare professionals) about the opportunity to take part in the study using information provided by the lead researcher. The aim is to recruit healthcare professionals who work in different settings (e.g. community, hospice) and/or may be employed by other organisations. The Chief Investigator would be responsible for obtaining written consent from all participants wishing to take part in the study.

Healthcare professionals can consent to one, a few or all of the following research activities:

- A one hour interview with the Chief Investigator that can be held face-to-face, on the telephone or online (up to 6 healthcare professionals will be interviewed)
- A 90 minute focus group involving up to 8 mixed role healthcare professionals
- Being observed by the Chief Investigator during discussions with people with MND and caregivers (e.g. in clinic or home visits) and other healthcare professionals (e.g. in multidisciplinary meetings) about the decision to have a gastrostomy placed
- The Chief Investigator will ask healthcare professionals to share any guidance, care pathways, or policies they refer to that are relevant to the research.

The Chief Investigator will recruit 3-5 people with MND to take part in:

- A single one hour interview with the Chief Investigator
- A review of their medical notes
- The caregivers (where present) will also be recruited to take part in an interview

The data collection on each site is expected to take place over a 6-9 month period.

Following the completion of the study participants will be invited to attend a workshop where the findings will be presented and discussion to inform the development of healthcare professional guidance about supporting people with MND to make decisions about gastrostomy placement.

The study will have obtained approval from the Health Research Authority, NHS and University Ethics and sponsor (Sheffield Teaching Hospitals NHS Foundation Trust) prior to its commencing.

What are the risks, burdens and benefits of being a case study site?

The main burden will be the time it takes for healthcare professionals to inform people with MND about the opportunity to take part in the study and participate in the research activities e.g. focus group, interview and workshop. The Chief Investigator will be flexible with regards when research activities take place and there will be the option to do them online.

The benefits will include the Chief Investigator reporting back the findings from each case study site, in addition to the combined analysis from all three sites. These findings could be used by sites to evaluate their approach to supporting the decision making of people with MND and inform local service improvements. The collaboration between the Chief Investigator and case study participants during the workshops will inform the development of guidance for healthcare professionals

supporting people with MND making decisions about gastrostomy placement, with the aim of improving practice nationally.

What should we do if we wish to express an interest in being a case study site?

Please can you complete the short questionnaire by following the link below (also included in the email sent to you).

https://forms.gle/kU3pB5R3GRWppRy97

The questionnaire will enable the Chief Investigator to understand your local MND service and inform the selection of the case study sites. The Chief Investigator is happy to share further information about the study or arrange a time to discuss. Please contact the lead researcher at smwhite1@sheffield.ac.uk if you require further information about the study or wish to arrange a time to discuss.

The Chief Investigator will seek to have further discussions with the sites selected for the study.

Many thanks for reading this information

Sean

Sean White

Chief Investigator

NIHR Clinical Doctoral Research Fellow

University of Sheffield

Sheffield Teaching Hospitals NHS Foundation Trust

Email: smwhite1@sheffield.ac.uk

Appendix 23. Screening questionnaire sent to potential cases

Section 1 of 3

Case study site screening questionnaire

These questions are being sent out to people who have expressed an interest in taking part in the study titled: 'Multiple case study exploring how people with motor neuron disease (MND) are supported by their clinical teams to make decisions about gastrostomy feeding tube'.

You can learn more about what will be involved by taking part in this study by reading the information attached to the email you have received.

Your answers to these questions will help inform the selection of the three case study sites to be included in the study.

What is your name?

What is your professional role?

What is the name of your organisation?

Section 2 of 3

Your local MND service

The following questions are aimed at understanding your local MND service

Where do your MND clinic(s) take place? (tick all the apply)

- Hospital out-patient clinic
- Community based clinic
- Hospice out-patient clinic
- Other…

How often are MND clinic's held (in any location)?

- Every week
- Every 2 weeks
- Every 3 weeks
- Every 4 weeks
- Other…

What is the professional role of the clinical lead for your MND service?

- Neurologist
- Palliative care consultant
- Respiratory consultant
- MND nurse or coordinator
- Other…

Which of the following statements best describes your local MND service? (if neither of these options describe your service then please provide more information in 'other')

- Tertiary service (i.e. have people with MND on your caseload from outside your local area)
- Non-tertiary service (i.e. only see people with MND from within your local area)
- Other…

How would you describe the model of MND care that you deliver?

- MND care centre
- MND care network
- Neurology clinic (non- MND care centre or network)
- Hospice/palliative care clinic (non- MND care centre or network)
- Other…

Which healthcare professionals are employed by your organisation and may have discussions with people with MND about gastrostomy placement? (tick all that apply)

- Neurologist
- Respiratory physician
- Respiratory physiotherapist or nurse
- Palliative care physician
- Palliative care nurse
- MND nurse
- MND Coordinator
- Dietitian
- Speech and language therapist
- Occupational therapist
- Physiotherapist (non-respiratory specialist)
- Psychologist
- Other…

Do healthcare professionals outside your organisation have discussions with people with MND about gastrostomy placement?

- Yes
- No

Can you estimate how many people are diagnosed with MND per year in your MND service? (it is not essential you provide this information now if you do not have it easily to hand)

Can you estimate how many patients are currently on your MND service caseload? (it is not essential you provide this information now if you do not have it easily to hand)

Section 3 of 3

Principal Investigator Role

The following questions relate to the Principal Investigator role for the study.

Having read the information provided can you confirm that you have an individual who could take on the role of principal investigator (PI)? The PI will be the local lead for the study and assist the Chief Investigator with recruitment of participants.

- Yes
- No

Can you give the name and role of the person who would be principal investigator.

Would you like to be considered as a case study site for the proposed research?

- Yes
- No

The Chief Investigator (Sean White) will be happy to discuss the study further with you. Please contact him at smwhite1@sheffield.ac.uk. If you have any further questions or wish to provide more information please write these below.

Please click on 'Submit' below to submit your answers.

Thank you for completing this questionnaire. The Chief Investigator will be in contact with you soon.

Appendix 24. Characteristics of the MND services on Cases 1, 2 and 3.

Characteristic	Case 1	Case 2	Case 3
MND caseload	255	130	171
New diagnoses per year (approximate)	100	37	70
MND Clinic	Weekly MND clinics hosted in a large teaching hospital out-patient department.	Monthly MND clinics hosted in each of two different hospital out-patient departments in the region.	Monthly MND clinics hosted on two sites in the region (one hospital outpatients and another in an old GP surgery).
Professions present in MND clinic	Neurologists, MND CNS and respiratory technician. Bimonthly	Neurologist, MND nurses, MND coordinator, respiratory	Neurologist, respiratory consultant, respiratory nurse, palliative care

	MDT clinics are also attended by dietitian, SLT, physio, OT and social worker. Respiratory consultant and nurse/physiotherapist attends clinic monthly. Palliative Care Consultant attends clinic once a month.	clinicians, respiratory nurse and palliative care consultant.	consultant, dietitian, SLT, respiratory physiotherapist, research nurse and MND coordinator
MDT meetings	Bi-monthly hospital-based MDT meeting attended by the care centre MDT. Monthly MDT meeting in respiratory centre attended by ventilation CNS, neurologists, palliative care consultant and MND CNS. Monthly MDT meetings in each community locality attended by range of community HCPs and MND CNS from MND centre.	Monthly MDT meetings attended by MND clinic team and HCPs from the acute service. Periodic (usually monthly) MND MDT meetings in each community locality attended by range of community HCPs and MND CNS from MND centre.	Face to face site-based MDT meeting before and after each MND clinic. 3 monthly regional MDT meeting attended by the team.
Community services	Fourteen community localities across the region belonging to different NHS Trusts include cross-section of AHP and nursing roles.	A large number of community localities across the region belonging to different NHS Trusts and	Most HCPs in clinic also can visit pwMND at home between clinic appointments. There were no separate community therapy

		including variety of nursing and AHP roles.	teams except for community palliative care team and occupational therapy (see below)
Respiratory services	Respiratory service hosted in different hospital and NHS Trust. Respiratory consultant attend monthly MND clinics in the MND centre. Respiratory centre runs its own respiratory out-patient clinics. Respiratory CNS team provide out-patient clinic and community out-reach services.	Respiratory consultants and nurses core part of MND MDT, attending all MND clinics and MDT meetings.	Respiratory consultants and nurses core part of MND MDT, attending all MND clinics and MDT meetings.
Palliative care services	Palliative care consultant attends a monthly MND centre clinic. MND clinics also run in two regional hospices, attended by palliative care, neurologist and MND CNS.	Palliative care consultant attends MND clinics. pwMND can also be referred to their local hospice or community palliative care teams.	Palliative care consultant core member of MND clinic MDT, seeing all pwMND from diagnosis. Can also refer into the community palliative care team.

Appendix 25. pwMND interview participant characteristics.

	P1	P2	P3	P4	P5	P6
Gender	Male	Male	Female	Male	Male	Male
Time since diagnosis	75 months	2 months	9 months	4 months	12 months	8 months
Speech	No speech problems	Significant dysarthria. Communicated by writing on a pad.	Unable to speak. Communicated using I-Pad.	No speech problems	No speech problems.	No speech problems
Observations involved in	Observed in clinic with neurologist and MND CNS.	Not observed.	Observed in clinic with neurologist and dietitian.	Observed discussing gastrostomy in home with dietitian and nurse	Not observed	Not observed
HCPs discussed gastrostomy with	Neurologist, MND CNS, palliative care consultant, respiratory CNS.	Ward doctor (not a neurologist), MND CNS, SLT, Dietitian	Neurologist, dietitian, respiratory centre,	Dietitian, community neurology nurse	Neurologist, respiratory consultant	MND CNS, dietitian, neurologist, respiratory consultant.
First discussion	~ 3½ years after diagnosis	At diagnosis	2 months after diagnosis.	3 months after being diagnosis.	2-3 months after diagnosis.	MND nurses discussed gastrostomy 3 months after diagnosis.

Current decision	Declined gastrostomy.	Agreed to gastrostomy soon after diagnosis.	Agreed to gastrostomy 7 months after diagnosis.	Is open to gastrostomy placement when experiencing dysphagia.	Agreed to gastrostomy 10 months after diagnosis.	Agreed to gastrostomy 7 months after diagnosis.
Gastrostomy placed	No	Awaiting gastrostomy placement.	Gastrostomy placed 3 weeks ago.	Deferred decision	Awaiting gastrostomy placement	Awaiting placement.
Caregiver interviewed?	Yes (separately)	Yes (separately)	Yes (separately)	Yes (separately)	Yes (with pwMND)	Yes (with pwMND)

	P7	P8	P9	P10	P11	P12
Gender	Female	Male	Male	Male	Male	Male
Time since diagnosis	9 months	4 months	21 months	2 years	20 months	3 years
Speech	Unable to speak. Communicated through writing.	No speech problems	No speech problems	No speech problems	No speech problems	No speech problems
Observations involved in	Observed in clinic with neurologist and respiratory consultant	Not observed	Observed in clinic with neurologist, MND CNS, palliative care consultant and respiratory consultant	Observed in clinic with respiratory consultant and physiotherapist.	Observed in clinic with dietitian, SLT, palliative care consultant, respiratory consultant, physiotherapist and nurse. Observed at home with dietitian.	Not observed

HCPs discussed gastrostomy with	Respiratory consultant, two neurologists, SLT, MND coordinator and MND CNS.	MND nurse, dietitian, neurologist.	Neurologist, MND CNS, respiratory consultant, palliative care consultant.	Dietitian.	Dietitian, SLT.	Dietitian, palliative care consultant, MND coordinator.
First discussion	Neurologist discussed gastrostomy 3 months after diagnosis.	Neurologist discussed gastrostomy at diagnosis	Early after diagnosis as part of advanced care planning.	Neurologist discussed gastrostomy at diagnosis.	Dietitian discussed 19 months after diagnosis.	Unsure. Thinks it was mentioned early by palliative care consultant.
Current decision	Uncertain	Agreed to gastrostomy shortly after being diagnosed	Declined gastrostomy	Agreed to gastrostomy 16 months after diagnosis.	Agreed to gastrostomy	Agreed to gastrostomy 2 years and 10 months after diagnosis.
Gastrostomy placed	No	Awaiting placement	No.	Yes	Awaiting placement.	Awaiting placement.
Caregiver interviewed?	Yes (with pwMND)	Yes (separately)	No caregiver	No (declined)	Yes (separately)	Yes (with pwMND)