CORA Cork Open Research Archive
Cartlann Taighde Oscailte Chorcaí

Title	Living with motor neurone disease (MND) and dysphagia – the personal experiences of people with MND and their caregivers
Authors	Lisiecka, Dominika
Publication date	2018
Original Citation	Lisiecka, D. 2018. Living with motor neurone disease (MND) and dysphagia – the personal experiences of people with MND and their caregivers. PhD Thesis, University College Cork.
Type of publication	Doctoral thesis
Rights	© 2018, Dominika Lisiecka. - http://creativecommons.org/licenses/by-nc-nd/3.0/
Download date	2026-09-14 21:04:58
Item downloaded from	https://hdl.handle.net/10468/5471

UCC
University College Cork, Ireland
Coláiste na hOllscoile Corcaigh

Ollscoil na hÉireann, Corcaigh

National University of Ireland, Cork

Living with Motor Neurone Disease (MND) and Dysphagia – the personal experiences of people with MND and their caregivers

Volume 1(2)

Thesis presented by

Dominika Lisiecka, MSc. in Speech & Language Therapy

For the Degree of

Doctor of Philosophy

IN MEDICINE AND HEALTH

SCHOOL OF CLINICAL THERAPIES — DEPARTMENT OF SPEECH AND HEARING SCIENCES

Head of Department: Prof. Nicole Muller

Supervisors: Dr. Helen Kelly, Prof. Jeanne Jackson

2018

Table of Contents

DECLARATION ..... **ix**
List of Tables ..... **x**
Dedication ..... **xiv**
Acknowledgments ..... **xv**
Abstract ..... **xvii**
PREFACE AND RATIONALE FOR THIS THESIS **1**
Margaret *1*
Background to the Thesis *2*
Statement of the Problem *3*
Research Aims and Questions *5*
Overview of chapters *5*
CHAPTER 1: LITERATURE REVIEW - MOTOR NEURONE DISEASE (MND) AND DYSPHAGIA **8**
Introduction **8**
1.1 An Introduction to MND **8**
1.1.1 The Diagnosis of MND *10*
1.1.2 The Prognosis of MND *11*
1.1.3 The Management of MND *13*
1.1.4 MND in the Irish Context *15*
1.2. Dysphagia Associated with MND **17**
1.2.1 The Symptoms of Dysphagia *18*
1.2.2 The Consequences of Dysphagia *20*
1.2.3 The Diagnosis of Dysphagia *22*
1.2.4 The Management of Dysphagia *23*
1.2.4. a) Rehabilitation and Compensatory Techniques *25*
1.2.4. b) Non - oral Feeding *28*
The Types of Non - oral Feeding in MND *29*
Factors Influencing Non - oral Feeding in MND *30*
The Impact of Non - oral Feeding on Survival in MND *32*
Non - oral Feeding and Cognitive Impairment in MND *34*
1.3. The Experiences of Dysphagia **35**

1.3.1 The Experiences of Dysphagia Related to MND *39*
1.3.1. a) Non - oral Feeding and the Decision Making Process *40*
1.3.1. b) Living with Non - oral Feeding *42*
1.4 Chapter Summary **44**
CHAPTER 2: RESEARCH PROCEDURE **47**
Introduction **47**
2.1 Methodological Underpinnings **47**
2.1.1 Rationale for a Qualitative Research Design *47*
2.1.2 Epistemology *48*
2.1.3 Interpretative Phenomenological Analysis (IPA) - An Introduction *50*
2.1.4 Theoretical Underpinnings of IPA *52*
2.1.4. a) Phenomenology *52*
Edmund Husserl *53*
Martin Heidegger *54*
Maurice Merleau-Ponty *56*
Jean-Paul Sartre *56*
2.1.4. b) Hermeneutic *57*
Friedrich Schleiermacher *57*
Martin Heidegger *58*
Hans-Georg Gadamer *59*
2.1.4. c) Idiography *60*
2.1.5 Relating the Philosophy of IPA to This Project *61*
2.2 Research Methods **61**
2.2.1 Research Design *62*
2.2.2 Study Sites *62*
2.2.3 Population and Sample *62*
2.2.3. a) Sample Size *63*
2.2.3. b) Eligibility Criteria *64*
The People with MND *64*
The Caregivers *66*
2.2.3. c) Recruitment Procedure *66*
2.3 Data Collection **68**
2.3.1 Multiple Interviews *69*
2.3.1. a) Indirect Interviews *70*
2.3.1. b) Duration of Interviews *72*

2.3.1. c) Gap between Interviews *72*
2.3.2 Observations *76*
2.4 Research Ethics and Consent **77**
2.5 Data Analysis **79**
2.6 Pilot Study **85**
2.7 Assessing Quality **88**
2.7.1 Sensitivity to Context *88*
2.7.2 Rigour and Commitment *91*
2.7.3 Transparency and Coherence *92*
2.7.4 Impact and Importance *92*
2.8 Researcher Positionality **93**
2.8.1 Potential Issues that Concerned Me *94*
2.8.2 My Presuppositions *95*
2.8.3 The Journey through this Research *96*
CHAPTER 3: PARTICIPANTS **99**
Introduction **99**
3.1 The People with MND **99**
3.1.1 Introducing the People with MND: Short Stories *102*
Bob *102*
Carmel *103*
Eileen *104*
Fiona *105*
Joseph *105*
Mike *106*
Sally *107*
Sheila *108*
Ted *110*
Terence *111*
3.2 The Caregivers **112**
3.2.1 Introducing the Caregivers: Short Stories *114*
Angela *114*
Helen *115*
Jack *116*
Jackie *116*
Kay *117*

Mary…… *118*
Martha…… *119*
Sophia…… *120*
Tom…… *120*
Una…… *121*
CHAPTER 4 RESULTS: THE PEOPLE WITH MND 122
Introduction 122
Presentation of Findings 125
Over-arching Theme: Living in the Here and Now 135
4.1 Superordinate Theme 1: MND as a Life Changing Disease 136
4.1.1 Subordinate Theme 1: Changed Flow of Life *137*
Emergent Theme 1: Awareness of Upcoming Deterioration *137*
Emergent Theme 2: Lost Skills and Independence *139*
Emergent Theme 3: Changed Rhythm of Life *141*
Emergent Theme 4: Fragility of Life *141*
Emergent Theme 5: Ruined Plans *142*
4.1.2 Subordinate Theme 2: Coping *142*
Emergent Theme 1: Maintaining Previous Routines *143*
Emergent Theme 2: Finding Solution to Manage *145*
Emergent Theme 3: Family Support *145*
Emergent Theme 4: Avoidance *146*
Emergent Theme 5: Making Sense of MND *147*
Emergent Theme 6: Maintaining a Positive Attitude *148*
4.1.3 Subordinate Theme 3: Emotions and Feelings *149*
Emergent Theme 1: Loss and Grief *149*
Emergent Theme 2: Nervousness and Uncertainty *149*
Emergent Theme 3: Frustration *150*
Emergent Theme 4: Happiness *151*
Emergent Theme 5: Hatred *151*
Emergent Theme 6: Diagnosis as a Relief *152*
4.2 Superordinate Theme 2: Living with Dysphagia 153
4.2.1 Subordinate Theme 1: Consequences of Dysphagia *155*
Emergent Theme 1: Eating as Risk *155*
Emergent Theme 2: Changed Role of Food and Diminished Pleasure from Eating and Drinking ……*156*

Emergent Theme 3: Changes in the Social Aspect of Meals *157*
Emergent Theme 4: Repetitive and Limited Diet *158*
Emergent Theme 5: Inability to Eat Preferred Food *159*
Emergent Theme 6: Emotional Impact of Dysphagia *160*
Emergent Theme 7: Changed Participation in Celebrations *161*
4.2.2 Subordinate Theme 2: Self-management Techniques *162*
Emergent Theme 1: Noticing Physical Symptoms of Dysphagia *163*
Emergent Theme 2: Paying Special Attention during Meals and Modifying Eating Environment *166*
Emergent Theme 3: Changed Preparation of Food *167*
Emergent Theme 4: Excluding Risky Food *168*
Emergent Theme 5: Eating, Drinking, and Swallowing Routines *169*
4.3 Superordinate Theme 3: Professional Management of Dysphagia 170
4.3.1 Subordinate Theme 1: Recommendations to Manage Dysphagia *171*
Emergent Theme 1: Non - oral Feeding *171*
Emergent Theme 2: Modification of Fluid Consistency and Textures of Food *180*
Emergent Theme 3: Adherence and Adjustment *181*
4.3.2 Subordinate Theme 2: Service Provision *185*
Emergent Theme 1: General Satisfaction *185*
Emergent Theme 2: Associated Emotions *186*
Emergent Theme 3: Suggestions for Change *188*
4.4 Chapter Summary 189
CHAPTER 5 RESULTS: THE CAREGIVERS OF PEOPLE WITH MND 192
Introduction 192
Presentation of Findings 195
The Overarching Theme: Transformation 207
5.1 Superordinate Theme 1: Immersion in the Caregiving as an Act of Slacklining 208
5.1.1 Subordinate Theme 1: Changed Flow of Life *209*
Emergent Theme 1: Unpredictability *210*
Emergent Theme 2: Changed Plans *211*
Emergent Theme 3: Living by Routines *211*
Emergent Theme 4: Having no Break *212*
Emergent Theme 5: Disturbed Sleep *213*
Emergent Theme 6: Neglecting Self *214*
5.1.2 Subordinate Theme 2: Caring as Balancing *215*

Emergent Theme 1: Having to Cope with Life on Top of MND *215*
Emergent Theme 2: Attempting to Protect Own Needs *216*
Emergent Theme 3: Adjusting Level of Care to Promote Independence of the PwMND *217*
Emergent Theme 4: Quality of Life (QOL) Versus Professional Recommendations *218*
Emergent Theme 5: Intermittent Desire to Know the Prognosis *218*
5.1.3 Subordinate Theme 3: Emotions and Feelings *219*
Emergent Theme 1: Shock of the Diagnosis *219*
Emergent Theme 2: Frustration *221*
Emergent Theme 3: Loneliness *221*
Emergent Theme 4: Lack of Appreciation *222*
5.1.4 Subordinate Theme 4: Coping *223*
Emergent Theme 1: Avoidance *224*
Emergent Theme 2: Positive Thinking *224*
Emergent Theme 3: Focus on the Here and Now *225*
Emergent Theme 4: Reciprocal Relationship with the Person with MND *227*
5.2 Superordinate Theme 2: Transformation of Mealtime Experience 228
5.2.1 Subordinate Theme 1: Eating as a Risk of Choking *230*
Emergent Theme 1: Visibility of Dysphagia *230*
Emergent Theme 2: Food as Danger *231*
Emergent Theme 3: Emotional Impact of Dysphagia *232*
5.2.2 Subordinate Theme 2: Focus on Safety *241*
Emergent Theme 1: Diet Modifications *241*
Emergent Theme 2: Supervision *242*
Emergent Theme 3: Changed Approach to Food *243*
5.3 Superordinate Theme 3: Professional Management of Dysphagia 245
5.3.1 Subordinate Theme 1: Professional Recommendations to Manage Dysphagia *245*
Emergent Theme 1: Consistency Modifications *245*
Emergent Theme 2: Non - oral Feeding *248*
Emergent Theme 3: Adjustment and Adherence *252*
Emergent Theme 4: Contradictory Recommendations *254*
5.3.2 Subordinate Theme 2: Service Provision *255*
Emergent Theme 1: Accessibility *256*
Emergent Theme 2: MND Clinic *257*
Emergent Theme 3: Involvement of the Caregiver *258*

Emergent Theme 4: Professional Approach *259*
Emergent Theme 5: Domiciliary Service *263*
Emergent Theme 6: Training *264*
5.4 Chapter Summary **265**
CHAPTER 6: DISCUSSION **268**
Introduction **268**
6.1 The Symbiotic Relationship between a Person with MND and their Caregiver **269**
6.2 Was Dysphagia an Issue for People Living with MND? **271**
6.2.1 What People Living with MND Wished to Talk about - People Diagnosed with MND *274*
6.2.2 What People Living with MND Wished to Talk About - The Caregivers *277*
6.2.3 How People Living with MND Understood the Term Dysphagia? *280*
6.3 How People Living with MND Wished to Manage Dysphagia? **282**
6.3.1 Managing Alone – People Diagnosed with MND *282*
6.3.2 Wishing for More Professional Help – The Caregivers *283*
6.3.3 Approaching Professional Recommendations to Manage Dysphagia *286*
6.3.3a) Texture Modifications *289*
6.3.3b) Non - oral Feeding *292*
6.4 The Impact of Dysphagia **295**
6.4.1 The Social Aspects of Meals *295*
6.4.2 Transformed Relationship with Food *299*
6.4.3 Nutrition and Weight *300*
6.4.4 Eating as a Struggle *302*
6.4.5 Readiness to Accept Non - oral Feeding *303*
6.5 Professional Management of Dysphagia **305**
6.5.1 MND Clinics *305*
6.5.2 Wishing for a Positive Experience *307*
6.5.3 Suggestions for Change *309*
6.6 Reflections for Professionals Providing Dysphagia - related Services in MND **310**
6.7 Chapter Summary **316**
CHAPTER 7: FINAL REFLECTION – STUDY CONTRIBUTION TO RESEARCH AND PRACTICE **317**
7.1 Study Contribution **317**
7.2 Implications for Future Research **320**

DECLARATION

Candidate's Declaration

This is to certify that the work I am submitting is my own and has not been submitted for another degree, either at University College Cork or elsewhere. All external references and sources are clearly acknowledged and identified within the contents. I have read and understood the regulations of University College Cork concerning plagiarism.

Candidate's Signature:..... Date:.....

List of Tables

Table 2.1 Multiple Interviews: A Summary 75

Table 3.1 Demographic Characteristics of the People with MND 101

Table 3.2 Demographic Characteristics of the Caregivers 113

Table 4.1 Hierarchy of Themes: The People with MND..... 127

Table 4.2 Presentation of Themes Supported by Chosen Quotations from Transcripts: The People with MND 128

Table 4.3 Dysphagia Related Status of the People with MND during the Data Collecion Period..... 154

Table 5.1 Hierarchy of Themes: The Caregivers..... 196

Table 5.2 Presentation of Themes Supported by Chosen Quotations from Transcripts: The Caregivers 197

Table 5.3 Dysphagia - related Status of the PwMND under the Care of the Participants 229

List of Figures

Figure 4.1 Hierarchy of Themes: The People with MND 124

Figure 4.2 Terence's Approach to a Professional Recommendation to Thicken Fluids 184

Figure 5.1 Hierarchy of Themes: The Caregivers 194

List of Abbreviations

A

AAC
Augmentative or Alternative Communication 72
ALS
Amyotrophic Lateral Sclerosis 8
ASHA
American Speech-Language-Hearing Association 25

C

CTPA
Communication and Treatment Preference Assessment Instrument....16

E

ECAS
Edinburgh Cognitive and Behavioural ALS Screen....71
EFNS
The European Federation of Neurological Societies 33

F

FDA
Food and Drug Administration 14
FOIS
Functional Oral Intake Scale....71
FTD
Frontotemporal Dementia 10
FVC
Forced Vital Capacity....12

I

IASLT
Irish Association of Speech and Language Therapists 24
ICF
International Classification of Functioning, Disability and Health....42
IMNDA
Irish Motor Neurone Disease Association 2
IPA
Interpretative Phenomenological Analysis....6

M

MDT
Multi Disciplinary Team....14
MND
Motor Neurone Disease xviii

N

NICE
National Institute for Health and Care Excellence 10

P

PEG

Percutaneous Endoscopic Gastrostomy 32
PwMND
People with Motor Neurone Disease xviii

Q

QOL
Quality of Life 3

R

RCSLT
Royal College of Speech & Language Therapists 24
RIG
Radiologically Inserted percutaneous endoscopic Gastrostomy 32

S

SLT
Speech and Language Therapy/ Therapist 5

Dedication

To my daughters Maya and Livia: Follow your dreams little ones! It will happen!

Acknowledgments

First of all, I wish to express my most sincere gratitude to all of my participants. Thank you for accommodating me during such a difficult time in your life. Thank you for being so honest and genuine. Thank you for being so inspirational. I will never forget any of you.

I would also like to sincerely thank my Supervisors, Dr. Helen Kelly and Prof. Jeanne Jackson, thank you for allowing me to develop this study and for your patience, guidance, and ongoing support. I thoroughly enjoyed our meetings and discussions.

Dr. Arlene McCurtin, my Mentor, thank you for your consistently good advice and unrelenting support and encouragement during this research.

Dr. Martina Carroll, thank you sharing your expertise of IPA with me throughout this study and for facilitating IPA research groups in Ireland.

To my Official Collaborators:

Prof. Orla Hardiman, and her team in Beaumont Hospital, thank you for your advice and for allowing me to shadow your work during the initial stages of this research;

The Irish MND Association, and Ms. Katie Kinsella in particular, many thanks for your invaluable assistance with recruitment;

and Dr. Helena Moore, a big thank you for your assistance with recruitment.

Ms. Leslie Doyle and Aideen O' Riordan, thank you for facilitating my attendance at MND Clinics and for sharing your expertise with me.

The Health Research Board, thank you for awarding me the Research Training Fellowship for Therapy Professionals 2015 (HPF-2015-993) and for your generous funding.

Áine Kearns, another HRB Fellow and PhD candidate, thank you so much for our discussions. You brought a lot of sunshine into my fellowship!

Aislinn Radwanski, my friend, thank you for your patience!

Last but not least, heartfelt thanks to my husband, other family members, and friends, who listened to me and supported me during the most intensive years of my life. An especially big thank you goes to my mother, who moved to Ireland for three years, despite her love of the sun, and who helped look after my children when I was away from home with this research.

Thank you all from the bottom of my heart!

Dziekuje Wam z Całego Serca!

Abstract

Living with Motor Neurone Disease (MND) and Dysphagia – the personal experiences of people with MND and their caregivers.

Introduction

MND is a rare progressive neurodegenerative illness for which there is no cure. There are approximately 350 people diagnosed with MND in Ireland at any one time. Dysphagia frequently occurs in neurodegenerative diseases such as MND, and if the onset of MND is in the bulbar region dysphagia may be the first symptom of this disease. Dysphagia is generally reported to interfere with the quality of life of people with MND (PwMND); however, little is known about how people living with MND understand and experience dysphagia.

Research aims

To explore the experiences of dysphagia in MND from the perspectives of PwMND and their caregivers in order to investigate: (1) how they understand dysphagia, (2) how dysphagia impacts their lives, (3) their coping strategies in relation to dysphagia, and (4) their experiences of professional services received to manage dysphagia.

Methods

Multiple in - depth interviews (supported by observations) were conducted with 10 PwMND and 10 caregivers from Ireland. Data was analysed utilising Interpretative Phenomenological Analysis whereby an idiographic approach was followed by a cross - case analysis of the two groups of participants. The findings were interpreted from a Speech and Language Therapy perspective.

Results

Two overarching themes were derived from the data: (1) *Living in the here and now* for the PwMND and (2) *Transformation* for the caregivers. It emerged that both groups approached dysphagia in a different manner. PwMND aimed to manage dysphagia on their own; however, the caregivers wished for increased professional support in specific areas, such as the management of choking. A changed perception of food and diminished eating - related pleasure was observed in both groups. Also, a difference was noted between participants' perception of their dysphagia and their clinical presentation. Participants expressed their views in relation to professional services received for dysphagia which could potentially motivate the professionals to reflect on the services currently provided for people living with MND and dysphagia.

Conclusion

This study suggests that the experience of dysphagia in MND is complex and should not be investigated / managed in isolation. The recognition of swallowing difficulties in oneself seems to be a lengthy process which does not always mirror the process of professional diagnosis. This may influence individuals' approach to professional recommendations and their engagement with health services. PwMND and their caregivers understand dysphagia differently and may have different expectations regarding dysphagia management.

“Feet, what do I need you for when I have wings to fly?”

- Frida Kahlo

PREFACE AND RATIONALE FOR THIS THESIS

Margaret

Margaret was the person who sparked my interest in MND and started me on a journey of many years, which culminated in this research. She was the individual who provided me with my first insight into what it was like to live with MND. I met Margaret shortly after I began working in an Irish hospital in 2006. Her husband wheeled her into my office on a Friday at 4pm. When I saw Margaret I felt completely overwhelmed. There were so many issues, so many disabilities, yet her mind was sharp and she knew what she wanted and what she did not. During the clinical dysphagia assessment Margaret showed signs of aspiration on every swallow, which worried me considerably. I could not think beyond the medical risks associated with dysphagia. I was even considering preventing her from returning home by admitting her to the hospital; such was the depth of my concern.

Margaret, on the other hand, simply smiled when I spoke about non - oral feeding and shook her head to refuse. She was not interested. "How are you managing the meals at home?" I asked her husband, hoping he would admit the struggle. "The meals are fine, no problem"- he answered. I realised I was alone in my dysphagia - related panic. I was unsure what to do.

Margaret was the first of many people with MND, who together with their families, have taught me many valuable lessons and provoked me to think "outside the box". Each person with MND brings their unique life story, which appears to influence

the manner in which they approach dysphagia. I owe a great deal to Margaret and the inspirational individuals I have met in the course of this research.

Background to the thesis

MND is a rapidly progressing, fatal and relatively rare neurodegenerative disease of unknown aetiology (Beresford, 1995; Boillee, Velde, & Cleveland., 2006; Chiò et al., 2009, 2013; Ferguson & Elman, 2007; Lee, 2012; Mitchell & Borasio, 2007). Those diagnosed with MND experience increased fatigue and develop progressive muscle atrophy and weakness, dysphagia and respiratory failure (Rowland, 1994; Sejvar, Holman, Breses, Kochanek, & Schonberger, 2005). The median survival time ranges from 20-48 months, with only 10-20% surviving longer than 10 years (Chiò et al., 2009). The median survival of Irish patients is 16.4 months (O'Toole et al., 2008). According to the Irish Motor Neurone Disease Association (IMNDA) there are approximately 350 people living with MND in Ireland at any one time (IMNDA, 2017). Currently, there is no cure for MND and treatment is largely symptomatic (Ferguson & Elman 2007).

Swallowing disorders (dysphagia) often occur as a result of progressive neurological conditions such as MND (Cichero & Murdoch, 2006; Grocher & Crary, 2010; Logemann, 1998; Mitsumoto, Chad, & Pioro, 1998; Murry, Carrau, & Chan, 2016). Dysphagia can be life threatening (Logemann, 1998; Kindell, 2002; Heiss, Goldberg, & Dzarnoshi, 2010; Rofes et al., 2011) and have a severe medical, social and psychological impact on those affected (Paterson, 1996). It is estimated that up to 90% of people with MND will experience dysphagia at some stage of the disease (Oliver, 1996).

Statement of the Problem

Living with a life-threatening condition such as MND can be challenging, overwhelming, and stressful (Brown, 2003; Blackhall, 2012; Foley, Timonen, & Hardiman, 2012; Kurt, Njboer, Matuz, & Kübler, 2007; Palmieri et al. 2010; Whitehead, O'Brien, Jack, & Mitchell, 2011). Having to cope with progressive weakness resulting in multiple disabilities requires a high level of support for both people with MND and their caregivers. Although the appropriate management of dysphagia has been shown to have significant prognostic implications in MND, obvious gaps in our knowledge remain (Cameron & Rosenfield, 2002; Simmons, 2005), for example, in relation to the benefits of non - oral feeding (Katzberg and Benatar, 2011). It is clear that health professionals require a thorough understanding of dysphagia associated with MND and would benefit from clear clinical guidelines in this area.

In addition, the personal experiences of those living with dysphagia in MND are currently largely unknown. As MND cannot be cured the primary goal of care is the optimization of quality of life (QOL) (Simmons, 2005). Interestingly, a number of studies have reported that QOL in people with MND does not correlate with physical strength and function and does not necessarily decline over time despite the progressive deterioration of physical functions (Simmons, Bremer, Robbins, Walsh, & Fischer, 2000; Robbins, Simmons, Bremer, Walsh, & Fischer, 2001; Bromerg & Forshew, 2002). Therefore, we cannot assume that when a person's swallow declines, so too does their QOL. There is also little currently known regarding how people with MND perceive their swallowing impairment and how they cope with it at home.

Finally, gaps also exist in our knowledge of the lived experiences of caregivers of people with MND in relation to the management of dysphagia. Delivering care to someone with a progressive neurological condition, such as MND (Galvin et al., 2016, Kaub-Wittemer, Steinbűchel, Wasner, Laier-Groeneveld, & Borasio, 2003; O'Connor & McCabe, 2011), Multiple Sclerosis (Aronson 1997; Cheung & Hocking, 2004 a, b; Knight, Devereux, & Godfrey, 1997; McCabe, Firth, & O'Connor, 2009; McKeown, Porter-Armstrong, & Baxter, 2003, 2004; O'Connor & McCabe, 2011), Huntington's disease (McCabe, Firth, & O'Connor, 2009; O'Connor & McCabe, 2011; Røthing, Malterud, & Frich, 2014), or Parkinson's disease (Bhimani, 2014; Jones et al., 2017; McCabe, Firth, & O'Connor, 2009; O'Connor & McCabe, 2011) has long since been recognized as very challenging. It is also known that in MND there is a significant association between caregiver and patient distress (Boerner & Mock, 2011). Some caregivers can spend over 11 hours a day looking after a person with MND (Krivicas, Shockley, Mitsumoto, 1997) and this can obviously include duties in relation to the management of dysphagia.

The literature review (presented in Chapter 1) revealed that little is known in relation to the impact of dysphagia on the lived experiences of both adults with MND and their caregivers. This knowledge is urgently required in order to support these two groups in the most efficient way, by ensuring that health resources are allocated based on identified need. Therefore, the current study aims to investigate the impact of dysphagia on the lives of both people with MND and their caregivers, thus reflecting one of the top research priorities identified for therapy professionals in Ireland:

Top research priorities for SLTs involve investigation of SLTs service provision in Ireland in terms of identified needs of service users and the extent to which they are

met and research that seeks the views of clients / carers with regard to experiences of living with swallowing difficulties (Health Research Board, 2010, pg.37).

Research Aims and Questions

This study aims to gain further insight into the lives of people living with dysphagia and MND and attempts to understand dysphagia in MND from the perspective of both those diagnosed with MND and their caregivers. It was envisaged that this knowledge could contribute to the professional management of dysphagia in MND and to the organisation of service provision.

The main research question is: How do people living with MND (both people diagnosed with MND and their caregivers) experience and understand dysphagia?

The secondary research questions are:

1. How does dysphagia impact on the lives of people with MND and their caregivers?
2. What are the main difficulties experienced in relation to dysphagia?
3. What are the ways of coping with these difficulties?
4. Are there ways to improve services for dysphagia emerging from this investigation of the lived experiences of people with MND and their caregivers?

Overview of chapters

This thesis consists of two volumes. Volume one consists of seven chapters and volume two includes all appendices and references. Chapter 1 presents a review of the

current literature focused on selected aspects of MND and dysphagia. The chapter begins by summarising the main characteristics of MND and how this disease is diagnosed. It then presents an overview of the evidence related to the management of MND, before focusing on MND in the Irish context. Subsequently, the topic of dysphagia is introduced by describing the main symptoms, consequences, and management options in dysphagia and relating these findings to MND. Lastly, studies investigating the experiences of living with dysphagia are included. As the number of studies in this field was limited, findings pertaining to experiences of dysphagia of any aetiology are summarised before focusing on MND.

Chapter 2 presents the research procedures. It explains the main features of Interpretative Phenomenological Analysis (IPA) and provides the justification for using this approach in the current study. It then presents the details of how the study was conducted and how the quality standards were maintained. Finally, the researcher's background and positionality are described.

Chapter 3 presents both the demographic characteristics and a summary of the personal story of each participant. This chapter is divided into two subsections: (1) people with MND and (2) caregivers.

Chapter 4 and 5 present the findings of this study. Chapter 4 is dedicated to the people with MND, and Chapter 5 to the caregivers. Chapter 4 and 5 are divided into subsections corresponding to the main themes identified through the process of data analysis. At the beginning of the chapter an overarching theme is presented: Living in the here and now (for people with MND) and Transformation (for the caregivers).

Chapter 6 discusses the main findings of this research and how they relate to current body of knowledge.

Chapter 7 highlights the relevance of this study in relation to professional practices. In addition, it identifies further gaps in our knowledge and suggests areas requiring future research.

CHAPTER 1: LITERATURE REVIEW - MOTOR NEURONE DISEASE (MND) AND DYSPHAGIA

Introduction

This chapter aims to orientate the reader to MND and dysphagia. It presents an up to date critical analysis of work that has been conducted in this field, which provides context to the current study. Chapter 1 begins with a presentation of the basic knowledge related to MND by summarizing the literature related to diagnosis, prognosis, and the management of this disease. A section focused on MND in the Irish context is also provided to situate participants' experiences, as the current study took place in Ireland. Subsequently, the topic of dysphagia is presented, in particular dysphagia associated with MND. Studies investigating the experiences of living with dysphagia are also included to outline work which has already been carried out and to identify gaps in the current knowledge in this field. The structure of this literature review was guided by the central focus of this study i.e. revealing an understanding of dysphagia in MND by investigating lived experiences.

1.1 An Introduction to MND

There are four principal types of MND: Amyotrophic Lateral Sclerosis (ALS), Progressive Bulbar Palsy, Progressive Muscular Atrophy and Primary Lateral Sclerosis (Armon, 2012; Mardsen, 2011). The majority (~ 70%) of patients with typical MND (ALS) present with spinal (limb) onset of the disease and others develop bulbar onset of MND leading to dysarthria and dysphagia (Wijesekera & Leigh, 2009). Although each type of MND has a unique presentation, symptoms may merge over time (Brown & Addington-Hall, 2008; Wijesekera & Leigh, 2009).

Although ALS is a subtype of MND the terms ALS and MND are often used interchangeably or in combination "ALS / MND" to cover the spectrum of progressive neurodegenerative syndromes affecting motor neurones (Armon, 2013). A recent international survey of 72 clinicians specialising in the management of MND confirmed that a broad number of terms were used for diagnostic and clinical descriptions of MND with no significant differences found between European, Australian, and North American practice (Rutter-Locher, Turner, Leigh, & Al-Chalabi, 2016). The term MND will be used throughout this thesis in line with the terminology employed in Ireland.

MND is understood to be a multisystem degenerative disorder not restricted to the motor system (van der Graaff, De Jong, Baas, & de Visser, 2009). Extra-motor dysfunctions are not uncommon, particularly in the area of the prefrontal cortex (Abrahams et al., 1997; Ludolph et al., 1992). These dysfunctions can include cognitive impairment (Abrahams et al, 1997; Abrahams, Leigh, & Goldstein, 2005; Murphy, Henry & Lomen-Hoerth, 2007; McCombe, Wray, & Henderson, 2017; Schreiber, Gaigalat, Wiedemuth-Catrinescu , Graf, Uttner, Muche & Ludolph, 2005; van der Hulst, Bak & Abrahams, 2015), emotional lability or pseudobulbar affect (Abrahams, Leigh, & Goldstein, 2005), sensory symptoms or neuropathies such as pain (Handy, Krudy, Boulis, & Federci, 2011), and abnormal autonomic functions such as fatigue or heart rate control (Merico & Cavinato, 2011). Language deficits, psychotic symptoms, changes in personality and behaviour can also be present in MND (van der Hulst, Bak, & Abrahams, 2015). Not every person with MND will develop cognitive or behavioural symptoms, about 50% will present with subtle changes whereas 10-15% will develop frontal dysfunction severe enough to reach the diagnosis of frontotemporal dementia (FTD) (Abrahams, 2013; Elamin et al., 2013;

Lilo & Hodges, 2010; Phukan et al., 2012; Ringholz et al., 2005; Taylor et al., 2013). Cognitive impairments of frontal type are more predominant in people with pronounced bulbar syndrome (Abrahams et al., 1997; Schreiber et al., 2005). The literature indicates differences between FTD and FTD related to MND (FTD-MND) such as the presence of aphasia (a language impairment) in FDT-MND not typically seen in pure FTD (Kamminga et al., 2016; Taylor et al., 2013).

1.1.1 The Diagnosis of MND

The diagnosis of MND is primarily based on the interpretation of clinical symptoms and investigations to exclude other causes (Andersen et al., 2012). According to the National Institute for Health and Care Excellence [NICE] (2016) the diagnosis should be given by a consultant neurologist with knowledge and expertise in MND and the level of information provided at the time of the diagnosis should be guided by the wishes of the person with MND. The "El Escorial" diagnostic criteria were originally developed in 1994 (Brooks, 1994) and revised in 2000 as "Airlie House" (modified) criteria (Brooks, Miller, Swash, & Munsat, 2000; Turner et al., 2013) to aid the diagnosis of MND. Based on the revised El Escorial criteria patients can be classified into four categories: "Clinically definite", "Clinically probable", "Clinically probable - Laboratory supported" and "Clinically possible" MND. However, some concerns have arisen in recent years in relation to employing the El Escorial criteria as a diagnostic tool in clinical practice due to more advanced knowledge of MND such as, for example, the importance of extra-motor manifestations (Agosta et al., 2014). The specificity of the "Possible" category has also been questioned (Ludolph et al., 2015). The World Federation of Neurology Research Group on MND / ALS proposed another revision of the El Escorial criteria,

which led to the modification of the original diagnostic categories and meant that a diagnosis of MND could be made if previous "Possible" criteria are fulfilled (Ludolph et al., 2015). It is now proposed that to receive a diagnosis of MND a person must present with progressive upper and lower motor neuron deficits in at least one region of the body (previous "Possible" category), or lower motor neuron deficits in one region observed clinically and / or in two regions if confirmed by electromyography, in the absence of evidence of other diseases which could explain the above symptoms (Lee, 2012; Turner et al., 2013).

It has been reported that the general public have poor awareness of MND which can result in a delay in medical advice being sought (Davies & Turner, 2010). The diagnosis is often made at the midpoint of the disease (Mitchell et al., 2010), yet it is recognised that timeliness is an important element of high quality health care (Institute of Medicine (US) Committee on Quality of Health Care in America, 2001).

1.1.2 The Prognosis of MND

A systematic review of survival in MND indicates that both the El Escorial criteria "Clinically definite" and bulbar onset of the disease are associated with a significantly poorer prognosis than all other types (Chiò et al., 2009; Moura et al., 2015). Poorer prognosis is also associated with a more advanced age at onset of MND (Chiò, 2009; Logroscino et al., 2010; Magnus, Giess, Puls, Naumann, & Toyka, 2002; Moura et al., 2015; Rooney, 2013), a faster early symptoms progression rate (Tortelli et al., 2016), and a quicker baseline Forced Vital Capacity[1] (FVC) decline (Logroscino et al., 2010; Magnus et al., 2002). In addition, a Body Mass Index [BMI] of less than

[1] FVC refers to total amount of air exhaled during the forced expiratory volume test.

25kg/m² (Moura, et al., 2015), the particular site of onset (Chiò et al., 2002, Magnus et al., 2002), and the presence of comorbid executive dysfunction or FTD (Chiò et al., 2009; Elamin et al., 2011; Rooney et al., 2013) all have an impact on prognosis. Although a bulbar onset of MND is more common in women, gender is not considered to be of prognostic significance (Chiò et al., 2009). The vast majority of people (90-95%) develop a sporadic (random) form of MND with the mean age of onset being approximately 60 years with a slight male prevalence (M:F ratio ~1.5:1) (Wijesekera & Leigh, 2009). Although the progression of MND appears to be variable and unpredictable, symptoms generally increase in their severity and distribution. However, recent evidence suggests that it is not uncommon for people with MND to experience periods of stability or slight reversals of the disease. In the recent study (PRO-ACT), 25% of participants did not decline over a 6 month period. This percentage diminished with time, for example, over a 12 month period only 16% did not decline (Bedlack et al., 2016). Also, some examples of MND reversals have been reported in the recent literature. Bedlack (2017) identified 23 cases of MND reversal with some people recovering most of their motor functioning. Bedlack provided a number of possible hypotheses which may explain this unusual occurrence, such as a diagnosis of MND-mimic disease rather than MND and a genetic resistance to the disease (Bedlack referred to a similar genetic abnormality leading to resistance to the HIV virus in some people). Other possible factors included the positive effects of some alternative therapies, and the removal of environmental triggers (Bedlack, 2017). However, Bedlack admits that large sustained MND reversals are rare and established as less than 1% (Bedlack et al., 2016). A study of MND reversals (St.A.R.) is currently being undertaken in America (www.alsreversals.com).

The majority of people with MND survive 2-3 years post diagnosis and only 10 – 20 % survive longer than 10 years (Chiò et al., 2009; Bouteloup, et al., 2009; Harwood et al., 2012). Respiratory issues are responsible for the majority of deaths in MND (Boarsio, Gelinas, & Yanagisawa, 1998; Wijesekera & Leigh, 2009).

1.1.3 The Management of MND

At present, there is no cure for MND and treatment is largely symptomatic (Ferguson & Elman, 2007). A palliative approach from the moment of diagnosis is recognised as the optimal management of MND (Bede et al., 2011). Riluzole was the sole disease-modifying medication approved since 1995 (Bensimon, Lacomblez & Meininger, 1994; Bensimon et al., 2002; Cleveland & Rothstein, 2001; Meininger, Lacomblez & Salachas, 2000; Mitchell et al., 2006; NICE, 2016) until the 5th of May 2017, when the U.S. Food and Drug Administration [FDA] approved Radicava (edaravone) to treat people with MND in America (FDA, 2017). It appears that both Riluzole and Radicava can be temporarily effective for people presenting with a specific form of MND. Riluzole (100 mg daily) "probably" extends the median survival by 2–3 months when taken for 18 months in patients diagnosed with Clinically Probable or Definite El Escorial MND and presenting with symptoms for 5 years or less, have a FVC greater than 60%, and be less than 75 years of age (Miller, Mitchell, Lyon, & Moore, 2012, pg. 14). The efficacy of the new drug (edaravone) was confirmed in Japan only for people diagnosed with "Clinically definite" or "Clinically probable" MND, with onset of the disease not extending 2 years; who were independent for activities of daily living, and presented with a progression of MND during the 12 weeks prior to the administration of edaravone (Tanaka, Sakata,

Palumbo, & Akimoto, 2016). Currently edaravone is not licenced in Europe for the management of MND.

The NICE guidelines (2016) recommend that care should be provided by a clinic-based MND multidisciplinary team (MND MDT), in a hospital or in the community. It is recognised that attendance at a MND clinic improves survival (Chiò et al., 2009; Hardiman, van den Berg, & Kiernan, 2011; Higo, Tayama, & Nito, 2004; Oliveira & Pereire, 2009; Rooney et al., 2013; Traynor, Alexander, Corr, Frost, & Hardiman, 2003). Ronney et al. (2015) proposed that multiple decision-making processes within a MND clinic utilising a MDT approach improves clinical outcomes for the people with MND.

The MND MDT should include health and social care professionals with expertise in MND along with domiciliary input, such as a neurologist, specialist MND nurse, dietitian, physiotherapist, occupational therapist, speech & language therapist, respiratory healthcare professionals, and a person with palliative care expertise (NICE, 2016). This team should have access to other services such as social care, neuropsychology / clinical psychology, counselling, respiratory / ventilation services, orthotics, gastroenterology, assistive technology services, wheelchair services, community neurology teams, and specialist palliative care (NICE, 2016). The NICE guidelines (2016) stress the importance of adequate communication between all professionals, people with MND, their caregivers and families in order to ensure all relevant people are informed of key decisions. The MND MDT should provide regular assessments and reviews at an estimated frequency of 2-3 months. However, it is acknowledged that some people would benefit from more or less frequent contacts depending on their individual needs. In addition, people with MND should not be discharged from the caseload even if their condition deteriorates, in order to ensure

continuity of care. Discussions with regard to end of life should be carried out whenever the person with MND wishes and support and advice on advance care planning should be provided (NICE, 2016). It is recognised that as the MND progresses the goals of care may change. In acknowledgement of this on-going change, a Communication and Treatment Preference Assessment Instrument (CTPA) was developed to provide structure for regular discussions with regard to treatment options and goals of care, and to inquire about legal documents (Morris, Walsh, Raheja, Kovacik-Eicher, & Simmons, 2016). The CTPA provides a framework to the MND clinics, it ascertains whether the person with MND has completed legal documentation, and assists in ensuring that management goals are consistent with the wishes of the person with MND.

1.1.4 MND in the Irish Context

Ireland is considered to be one of the leading countries engaged in researching MND and one of the key centres for new clinical trials for MND (Hardiman, 2014). The Irish MND Research Group was founded in 2007 to facilitate research into the aetiology and treatment of MND as well as to increase the awareness of this disease (Research Motor Neurone, 2017). The group maintains a national register of people with MND, which dates back to 1995. In addition, a DNA bank containing blood samples from people with MND attending Beaumont clinic in Dublin has been maintained since 1998 and is considered to be one of the best in the world (Hardiman, 2007). Irish people with MND are offered support from the Irish MND Association (IMNDA), which was set up in May 1985. The IMNDA's key service includes domiciliary visits by nurses specialising in MND, who in addition liaise with local

services to ensure the best possible care (www.imnda.ie/about-imnda/). Currently, there are three MND nurses providing services to the people with MND in Ireland.

According to O'Toole et al. (2008) the average annual incidence rate of MND in Ireland between 1995 and 2004 for the population older than 15 years was 2.6 per 100 000. There is a general agreement that the incidence of MND is homogeneous across Europe (Logroscino et al., 2010). O'Toole (2011) reported that there are 200-240 people living with MND in Ireland and 80 people are diagnosed annually. These figures seem to have increased (potentially reflecting the growth of the Irish population in general), as in 2015 Rooney et al. stated that approximately 110 patients are diagnosed annually with MND in Ireland (Rooney et al., 2015). Although recent reports by the IMNDA (IMNDA, 2017) state that there are approximately 350 people living with MND in Ireland at any one time, 382 people with MND were registered with IMNDA on the 3rd of April 2017 (IMNDA, personal communication, 03.04.2017).

A Report on the National Survey of Neurology Services (2015) confirmed significant deficits in the number of Neurologists across all neurology centres in Ireland (Neurological Alliance of Ireland, 2015). Although this report stated that the recommended ratio is one consultant Neurologist per 70,000 population, this population is exceeded in every Irish hospital (Baxter, 2010). The Irish ratio of consultant Neurologists is considered to be the lowest in Europe (Baxter, 2010). Moreover, there appears to be a significant delay in physicians referring patients to a neurologist (Baxter, 2010). According to Donaghy, Dick, Hardiman and Patterson (2008) people with MND in Ireland may spend 4.8 months under a physician's care before a referral to neurology is made. Donaghy et al., (2008) also established that the median time from the onset of MND symptoms to diagnosis was 15.6 months.

Considering the median survival time of people with MND in Ireland is reported to be between 14.7 months (Rooney et al. 2013) and 16.4 months (O'Toole et al., 2008) this time delay between symptom onset and diagnosis appears to be too long.

The rate of familiar (genetic) MND appears to be higher in Ireland (established at 16% by Byrne et al., 2013) in comparison to international rates (reported between 5-10% by Armon, 2013). Spatial cluster analysis of the MND population in Ireland revealed two significant areas of lower MND risk with the larger area corresponding to Co. Kilkenny and Co. Carlow, and a smaller low-risk region in Co. Clare and Galway (Ronney et al., 2015). Although Rooney et al. (2014) identified a slightly higher relative risk in the North-East coast (Co. Louth, Meath and North Dublin), Cork city, the Dingle peninsula in Co. Kerry, and the Western part of Co. Clare, their study from 2015 did not reveal any areas of increased risk.

Currently, there are 3 MND clinics in Ireland providing multidisciplinary care to people diagnosed with this disease. Professor Hardiman established the first clinic in Beaumont Hospital in Dublin in 1994 in collaboration with IMNDA. This clinic provides multidisciplinary care to any person with MND in Ireland (Hardiman, 2007). There are also MND Clinics in Galway and Cork. Irish studies were the first to report that multidisciplinary care delivered by MND MDT improves outcomes for people with MND (Traynor et al., 2003).

1.2. Dysphagia Associated with MND

Swallowing disorders often occur as a result of progressive neurological conditions such as MND (Carrau & Murry, 1999; Cichero & Murdoch, 2006; Grocher & Crary, 2010; Logemann, 1998; Mitsumoto et al., 1998) and if the onset of MND is in the bulbar region dysphagia may be the first symptom of this disease (Grocher &

Crary, 2010). The exact incidence of dysphagia in the population of people with MND is unknown (Ball et al., 2012). At least 73% of people with MND have dysphagia prior to requiring a ventilator and it is estimated that an even higher percentage present with dysphagia subsequently (Yorkston, Strand, Miller, Hiller, & Smith, 1993). According to Oliver (1996) 90% of people with MND will experience dysphagia during the progression of the disease. The correlation between bulbar onset of MND and the presence of dysphagia has been well established (Luchesi, Kitamua, & Mourão, 2014, Ruoppolo, et al., 2013). There is a significant body of knowledge related to the physical characteristics of swallow impairments in MND and it is relatively well understood how the physiology of swallowing can change during the course of this disease. The following section provides a summary of the literature concerned with the main characteristics of dysphagia associated with MND and how dysphagia is typically diagnosed and managed.

1.2.1 The Symptoms of Dysphagia

The term dysphagia refers to a cluster of swallowing impairments caused by an underlying disease or disorder (Reilly & Ward, 2005). These impairments can relate to any phase of the swallow: anticipatory (pre-oral stage involving environmental factors and eating behaviours that occur before the food enters oral cavity) (Leopold & Kagel, 1997), oral preparatory (mastication), oral (propulsion of the bolus), pharyngeal (bolus transfer through the pharynx), and oesophageal (bolus transfer through the oesophagus) (Cichero & Murdoch, 2006; Grocher & Crary, 2010; Logemann, 1998; Morgan & Ward, 2001; Murry et al., 2016; Schindler & Kelly, 2002; Seikel, King & Drumright, 2010). The symptoms of dysphagia can be subtle (Grocher

& Crary, 2010; Logemann, 1998) or there may not be any overt signs (Cichero & Altman, 2012).

Dysphagia in MND can manifest in reduced tongue mobility leading to difficulties with chewing and controlling food in the oral cavity (Dworkin & Hartman, 1979; Graner & Strand, 2010; Kawai, et al. 2003). Labial closure can be reduced leading to drooling or spillage of the food (Graner & Strand, 2010; Logemann, 1998). Velar function can also be affected causing nasal regurgitation of the food (Graner & Strand, 2010; Regan & Walshe, 2012; Robbins, Logemann, & Kirshner, 1982). Laryngeal elevation can be decreased placing a person with MND at risk of aspiration from their own secretions, liquids, and food (Leder, Novella, & Patwa, 2003; Logemann, 1998). A delay in initiating the pharyngeal swallow is also reported (Graner & Strand, 2010). Aspiration can occur before, during, and after swallow (Graner & Strand, 2010; Kawai, et al, 2003; Leder et al., 2003, Regan & Walshe, 2012). Gender differences have been reported placing women with bulbar onset of MND at a higher risk of developing severe dysphagia in comparison with men (Luchesi et al., 2014). One potential explanation for this phenomenon is that males have a significantly higher maximum tongue pressure capability (Stierwalt & Youmans, 2006) and lingual pressure is considered to be an important factor contributing to the presence or absence of dysphagia (Carrau & Murry, 2006; Kawai et al., 2003; Logemann, 1998). In addition, there are reports that the glossopharyngeal muscle is more susceptible to neurodegeneration in women (McCombe & Henderson, 2010). Interestingly, as MND has a limited impact on sensory neuron function, people with MND are reported to swallow relatively safely in the earlier stages of dysphagia, as they appear to adapt and compensate for the impaired ability to swallow (Ball et al., 2012).

1.2.2 The Consequences of Dysphagia

It is known that dysphagia can affect oral intake and it is estimated that 70% of people with neurogenic dysphagia present with malnutrition and dehydration (Almirall, Cabre, & Clave, 2007). Weight loss is considered to be a poor prognostic factor in MND (Desport et al., 1999; Jawaid, Murthy, Wilson, Qureshi, Amro, & Wheaton, 2008; Körner et al., 2003; Lacomblez, et al., 1996; Limousin et al., 2010; Stambler, Charatan, & Cedarbaum, 1998). In addition, weight loss appears to have a negative impact on QOL as it contributes to decreased energy levels (Körner et al., 2013). Although the exact prevalence of malnutrition is not known (Leah et al., 2003), it is estimated that 15-55% of people with MND present with clinically severe weight loss (Holm et al., 2013).

Interestingly, weight loss in MND is not always attributed to dysphagia (Körner, et al., 2013). The main confounding factors leading to malnutrition in MND are: hypermetabolism (Bouteloup et al., 2009; Desport et al., 2001; Desport, Torny, Lacoste, Preux, & Couratier, 2005a; Funalot, Desport, Sturtz, Camu, & Couratier, 2009; Limousin et al., 2010) and reduced caloric intake (Kühnlein et al., 2008; Limousin et al., 2010; Spataro et al., 2011). In addition, the literature suggests that dysphagia may not be the leading or only cause of loss of appetite in MND (Holm et al., 2013; Leigh et al., 2003). In MND, there appears to be a correlation between appetite loss and dyspnoea (Holm et al. 2013).

Other consequences associated with dysphagia may include choking, aspiration, pneumonia, and death (Cichero & Murdoch, 2006, Groher & Crary, 2009; Heiss, Goldberg, & Dzarnoshi, 2010; Kindell, 2002; Logeman, 1998; Marik & Kaplan, 2003; Murry et al., 2016; Rofes et al., 2011). Aspiration refers to material, such as oro-

pharyngeal secretions, food, pills or fluids entering the airway below the level of the true vocal folds (Murry et al., 2016). Aspiration pneumonia is a lung infection primarily caused by the aspiration of food, liquids or saliva during swallowing, the aspiration of post-swallow residue where a person is unable to clear residue from the oral cavity or pharynx, or the aspiration of gastroesophageal contents, for example, reflux (M. Groher & T. Groher, 2012). The risk factors of aspiration pneumonia include, for example, dependence on others for feeding and oral care, tube feeding, use of suctioning, and decreased mobility (Langmore et al., 1998; Langmore, Skarupski, Parks, & Fries, 2002). Other risk factors include dependence on a ventilator (Davis & Thompson Stanton, 2004), impaired or absent cough or decreased consciousness (Masiero, Pierobon, Previato, & Gomiero, 2008). Despite reports that aspiration pneumonia is relatively uncommon in MND, it is associated with high mortality rates (Sorenson, Crum & Stevens, 2007a). The current prevalence rate of aspiration pneumonia in MND is not known.

The social and psychological impact of dysphagia has also been recognised in various populations, such as elderly (Ekberg, Hamdy, & Woisard, 2002; Paterson, 1996) or people with head and neck cancer (Nguyen et al., 2005). Dysphagia is generally reported to affect the perceived QOL of people with MND (Franceschini & Mourão, 2015; McHorney, Robbins, Lomax, Rosenbek, Chignell, Kramer, & Bricker, 2002; Paris et al., 2013; Tabor, Gaziano, Watts, Robison, & Plowman, 2016). Although some studies report that this impact may not be related to disease duration (Franceschini & Mourão, 2015; Schindler et al., 2010), others suggest that people at a more advanced stage of MND experience poorer QOL related to swallowing (Tabor, et al., 2016). The most affected domains associated with swallow-related QOL appeared to be fatigue and eating duration (Franceschini & Mourão, 2015; Paris et al.,

2013; Tabor et al., 2016) contributing to decreased desire to eat (Paris et al., 2013). In addition, Tabor et al. (2016) reported a relationship between airway protection during swallowing, disease progression, and swallow related QOL in MND. For example, people with MND who aspirate were reported to present with an increased level of fear and reduced socialisation (Tabor et al., 2016).

1.2.3 The Diagnosis of Dysphagia

The assessment of dysphagia is usually conducted by a Speech & Language Therapist [SLT] and involves taking a full case history followed by a clinical and / or instrumental assessment (IASLT, 2015; Royal College of Speech & Language Therapists [RCSLT], 2014). During clinical (bedside) dysphagia assessments an oro-motor exam is typically conducted (to examine the structure and functioning of the oro-motor apparatus), followed by testing of the swallowing on appropriate consistencies of liquid and food (Logeman, 1998). A clinical assessment, as described above, can often identify the signs of dysphagia, for example, decreased laryngeal elevation or pharyngeal swallow delay (Cichero & Murdoch, 2006; Logeman, 1998; Murry et al., 2016). It may not, however, sufficiently identify the severity of dysphagia or the extent of aspiration and therefore an instrumental assessment of swallowing (such as a videofluoroscopy or fibre-optic endoscopic evaluation) may be indicated. Instrumental assessments are often required in order to guide a decision regarding oral intake, judge the extent of aspiration and the effectiveness of compensatory strategies (Clark, 2003; Daniels & Huckabee, 2008; Logemann, 1993, 1998).

SLTs may also be responsible for training other professionals, such as nursing staff or doctors, to carry out a screening assessment for dysphagia (Alberta College of

Speech-Language Pathologists and Audiologists, 2015; IASLT, 2016; RCSLT, 2007, 2010). The main purpose of this screening assessment is to establish the presence of dysphagia and the need to refer the individual for a full clinical assessment by a SLT (IASLT, 2016; RCSLT, 2014). According to Perry (2001a, 2001b) screening and detailed clinical assessments are two distinct procedures, which are conducted by different professionals, at different time points and for different reasons. Screening alone does not identify the severity of dysphagia and it does not inform the treatment of dysphagia (IASLT, 2016). Studies have identified benefits related to the screening of dysphagia in people post cerebrovascular accident (Bravata et al., 2009; Hinchey, Shephard, Furie, Smith, Wang, & Tonn, 2005; NICE, 2010; RCSLT, 2007). These findings have not been yet replicated with any other population (IASLT 2016). The IASLT therefore recommends that the screening assessment for dysphagia should be used only with people who have had a cerebrovascular accident (IASLT, 2016). This recommendation implies that if dysphagia is related to MND a full swallowing assessment carried out by a SLT is indicated, rather than a screening assessment for dysphagia only.

1.2.4 The Management of Dysphagia

It has been recognised that a team approach in dysphagia is essential in order to ensure a holistic service and that a multidisciplinary approach is considered best practice in the management of dysphagia (ASHA, 2002; Carrau & Murry, 2006; Leonard & Kendall, 2007; IASLT, 2015; Logemann, 1998; RCSLT, 2014). The crucial role that SLTs have in the management of dysphagia has also been recognised (American Speech-Language-Hearing Association [ASHA], 2004; IASLT, 2015;

RCSLT, 2014). According to Speech Pathology Australia (2004) the main aims in relation to the effective management of dysphagia include increasing the efficiency and safety of swallowing, recommending the most appropriate type of nutrition and hydration (such as oral versus non - oral), establishing the specific types of textures of food and fluid consistencies to be taken by a person with dysphagia, and maximising the social aspect of eating and drinking. The management of dysphagia depends on the underlying diagnosis (cause of the swallowing impairment) and expected prognosis (Logemann, 1998). In relation to MND, Tabor et al. (2016) strongly advocate for the multidisciplinary management of dysphagia. It has been established that while the provision of multidisciplinary care does not alter the neurological course of MND, it does significantly contribute to increased survival (Chiò, Bottacchi, Buaffa, Mutani, & Mora, 2006; de Rivera, et al., 2011; Traynor, et al., 2003; Sorenson, Mandrekar, Crum, & Stevens, 2007) and QOL (Jiménez, Sala, Riera, Herrera, Povedano, & Virgili, 2007; van den Berg et al., 2005). Despite these positive outcomes some studies reported dissatisfaction with health services in MND (Brown, Lattimer, & Tudball, 2005; Hugel, Grundy, Rigby, & Young, 2006; Hughes, Sinha, Higginson, Down, & Leigh, 2005; McCabe, Roberts, & Firth, 2008; van Teijlingen, Friend, & Kamal, 2011).

The NICE guidelines (2016) recommend monitoring weight, nutrition, and swallow throughout the course of MND from the initial diagnosis. Advice should be provided in relation to specialised eating and drinking utensils, food and liquid preparation, maintaining optimal posture during meals, and coping with social situations. The guidelines suggest that discussions regarding non - oral feeding should take place early and regularly. It is not known, however, what type of professional services aimed at the management of dysphagia are preferred by people with MND

and whether they have any suggestions in relation to potential modifications to the way these services are currently provided in Ireland.

1.2.4. a) Rehabilitation and Compensatory Techniques

The management of dysphagia can generally be divided into rehabilitation (therapy techniques) and compensation (compensatory techniques). Therapy techniques aim to improve the mechanism of the swallow itself (Rosenbek & Jones, 2009), for example, the super-supraglottic swallow, which aims to close the airway entrance (Logemann, 1983; Martin, Logemann, Shaker, & Dodds, 1993; Ohmae, Logemann, Kaiser, Hanson, Kahrilas, 1996) and improve the laryngeal elevation (Ohmae, et al., 1996). In order to be carried out correctly the majority of therapy techniques for dysphagia demand a certain level of stamina and cognitive status. During super-supraglottic swallow, for example, a person is required to hold a breath during swallowing and cough immediately after the swallow while simultaneously bearing down (Logemann, 1998). Therefore for some populations, including people with neurodegenerative conditions such as MND, the implementation of therapy techniques for dysphagia is typically not indicated and compensatory management is more frequently applied. In addition, the literature reports that therapy exercises should be used with caution in MND, as they can lead to swallow deterioration by increasing the level of fatigue (Walshe, 2014). However, Plowman (2015) stated that mild form of exercises should possibly be considered as part of dysphagia management in MND, although we do not have sufficient evidence in this regard yet.

Compensatory management is focused on techniques to compensate for lost functions and aims to immediately improve the safety of the swallow whilst optimising the level of nutrition and hydration (Rosenbek & Jones, 2009). There are a number of

compensatory techniques recommended for dysphagia. These include postural techniques related to altering the position of the body or head during eating and drinking; the provision of external labial, chin, or buccal support and the application of techniques to improve oral sensory input, such as introducing stronger taste or temperature. Additional compensatory techniques include changes in the feeding or eating process, for example a modified rate of eating; multiple swallows in order to clear oro-pharynegal residue; changes in the bolus size, and finally modification of the texture of food and fluids (Cichero & Murdoch, 2006; Groher & Crary, 2009; Lazzara, Lazarus, & Logeman, 1986; Logeman, 1992, 1998; Loret, 2015). In MND the compensatory strategies are typically prescribed to prevent aspiration (Palovcak, Mancinelli, Elman, & McCluskey, 2007), nutritional deficiency, and dehydration (Strand, Miller, Yourkston, & Hillel, 1996). It is recognised, however, that complete elimination of aspiration may not be achieved (Ball, Wright, & Lewis, 2013). The following paragraph presents the main characteristics of the texture modified diet. This compensatory strategy has become one of the most commonly recommended to manage dysphagia associated with various illnesses, as it is considered to contribute to safer swallowing (Cichero & Murdoch, 2006; Groher & Crary, 2009; Logeman, 1998; Murry et al., 2016; RCSLT, 2014; Steele et al., 2015).

A texture modified diet involves the alteration of fluid viscosity or food texture in order to optimise swallow safety or minimise the risk of aspiration. In Ireland, the IASLT and the Irish Nutrition and Dietetics Institute developed a consensus document outlining national terminology to be applied to texture modifications (2009). They distinguished four stages of liquid consistency (very mildly thick, mildly thick, moderately thick, and extremely thick) and four levels of modified food consistency

(soft, minced and moist, smooth pureed, and liquidised)[2] (IASLT, 2009). In order to alter liquid consistency, people with dysphagia are prescribed a thickening powder which is mixed with the fluids in specified amounts and impacts on their flow which ultimately provides the person with dysphagia more time to coordinate their swallow and achieve airway closure before the swallow is triggered (Clave et al., 2006; Logemann, 1993). There are multiple commercial types of thickeners currently used in Ireland. A recent systematic review (Steele et al., 2015) confirmed the benefits of thickened liquids in relation to reducing aspiration, however, a risk of post-swallow residue with thicker consistencies was also reported. Pharyngeal residue has been recognised as a potential risk of aspiration (Oliveira, Moreira, de Freitas, Gonçalves, Furkim, & Clavé, 2017). Moreover, the changes of liquid consistency are reported as being disliked by the patients (Chadwick, Jolliffe, Goldbart, & Burton, 2006; Colodny, 2005; Garcia, Chambers, & Molander, 2005; King & Ligman, 2011; McCurtin, Healy, Kelly, Murphy, Ryan, & Walsh, 2017; Macqueen, Taubert, Cotter, Stevens, & Frost 2003), which should also be taken in consideration when establishing dysphagia management plan.

In order to alter food consistency, people with dysphagia are often encouraged to eliminate unsafe types of food, to cut food into smaller pieces and to mash or blend it. These strategies are aimed at adjusting the cohesiveness, hardness, and slipperiness of the bolus. However, Steele et al. (2015) stated that the evidence to guide clinical practice in relation to the different levels of food texture modification for people with dysphagia is not sufficient and more research is required in this regard.

[2] Appendix 1 (for all appendices refer to volume two)

1.2.4. b) Non - oral Feeding

Non - oral feeding may be introduced in the management of dysphagia as a short or long term option (Holmes, 2004). Non - oral feeding is typically recommended for three main reasons: (1) if a person is unable to sustain oral nutrition (even if dysphagia is absent), (2) if the person requires additional nutrition or hydration on a short term basis, for example, to overcome an acute medical issue, or (3) if a person is at risk of aspiration related to dysphagia (M. Groher & T. Groher, 2012). Non - oral feeding is generally considered as a reliable route for administering medication, hydration, and nutrition (Russ, Phillips, Mel Wilcox, & Peter, 2015). If non - oral feeding is required for more than 30 days a gastrostomy is typically performed (Holmes, 2004). In general terms, gastrostomy refers to the surgical procedure of creating an external opening in the abdomen leading to the stomach. Commercially available high-caloric formulas are delivered through a catheter into the stomach using a syringe, a plastic bag hanging above the level of the tube, or a mechanical pump (M. Groher & T. Groher, 2012). However, in many developing countries these formulas may be replaced with blended food due to financial reasons (Pearce & Duncan, 2002). There are two main methods of non - oral feeding: a bolus feeding and a continuous feeding. A bolus feeding is given in larger amounts a few times a day, typically with a syringe. Alternatively, a continuous feed is given at a slow rate over an extended time period by a pump.

In order to prevent malnutrition and weight loss in people with MND the placement of a gastrostomy tube is generally recommended (Andersen et al., 2012; Katzberg & Benatar, 2011; R. Miller et al., 2009). Some authors, however, suggest that a trial of various compensatory strategies should be implemented in the first instance, leaving the discussion of non - oral feeding until after the compensatory strategies are found to be ineffective (Ball et al., 2012; Palovcak et al., 2007). The

international rates of gastrostomy insertion in MND vary from up to 60% in Japan (Seki, Kameya, & Kimura, 2000; Yanagisawa, 1996 as cited by Katzberg & Benatar, 2011) to up to 24% in Italy (Boitano, 2001; Chiò, 2002, Mazzini, 1995 as cited by Katzberg & Benatar 2011), Norway (Bak, 1994 as cited by Katzberg & Benatar, 2011), and Canada (Strong, 1999 as cited by Katzberg & Benatar, 2011).

The presence of a gastrostomy tube does not necessarily prevent a person from also receiving an oral diet, especially when palliative care is provided or as a means of improving QOL. It is often recommended to people with MND that they eat and drink orally in addition to using a non - oral route of feeding to enhance their QOL (Ball et al., 2012).

The Types of Non - oral Feeding in MND

Although there are various types of non - oral feeding, the percutaneous endoscopic gastrostomy (PEG) and the radiologically inserted percutaneous endoscopic gastrostomy (RIG) are the most commonly prescribed in MND (Moran & O'Mahony, 2015; Russ et al., 2015). According to Katzberg and Benatar's Cochrane review (2011) both options appear to be equally effective and have similar complication rates. However, more recent reports suggest that the evidence regarding which method is safer for the population of people with MND is inconclusive (ProGas Study Group, 2015; Stavroulakis, Walsh, Shaw, & McDermott, 2013). The insertion of a PEG tube can result in increased patient distress in comparison to a RIG, however, in the same study the RIG was reported to have an increased rate of post-procedural complications (ProGas Study Group, 2015). These concerns are not surprising considering the specifics of PEG tube insertion, such as the necessity to undergo intubation with an endoscope and the subsequent passing of the gastrostomy tube

through the oral cavity and into the placement site (Laasch et al., 2003). The complications following RIG insertion can be explained by understanding the characteristics of the RIG, which include a narrow tube (risk of blockage) and a balloon retention system (risk of deflation or displacement) which can result in the RIG not being fixed as securely against the abdominal wall as the PEG (ProGas Study Group, 2015). In other sources, however, the RIG was shown to have a higher success rate and a lower rate of complications compared with the PEG therefore it was recommended over the PEG especially when the FVC was reduced below 50% (Russ et al., 2015, Thornton et al., 2002). It has been reported, however, that the RIG is not widely available internationally, for example in Canada (Benstead, Jackson-Tartlon, & Leddin, 2016). In a study by Benstead et al. (2016) only 30% of responding clinics offered RIG insertion for people with MND.

Factors Influencing Non - oral Feeding in MND

Many factors appear to be taken into consideration when a recommendation of non - oral feeding is made in the population of people with MND. The evidence appears to support early tube placement i.e. before FVC falls below 50% of its predicted level, due to the risk of post-procedural respiratory complications (Andersen et al., 2007; Mathus -Vliegen, Louwerse, Merkus, Tytgat, & Vianney de Jong, 1994; Mazzini et al., 1995; Miller et al., 1999; R. Miller et al., 2009; Nunes, Santos, Grunho, & Fonseca, 2016; Russ et al., 2015; Sorenson et al., 2007a). The European Federation of Neurological Societies (EFNS) recommend considering non - oral feeding when weight loss is greater than 10% of the persons' premorbid weight, while taking into consideration the bulbar functions, respiratory functions, and the general condition of

the person with MND (Andersen et al., 2007). Although it has been reported that non - oral feeding may prevent the deterioration of nutritional status in MND (Nunes et al., 2016), a Cochrane Review concluded that the evidence supporting the nutritional benefits of non - oral feeding in this population is still weak (Katzberg & Benatar, 2011). A more recent study (ProGas Study Group, 2015) suggested considering non - oral feeding when: (1) the weight loss is approximately 5% and (2) the person with MND is not meeting their daily nutritional requirements. According to the ProGas Study Group (2015) non - oral feeding succeeds in preventing further weight loss in approximately 50% of people with MND. However, the authors highlighted that the greater the weight loss prior to the introduction of non - oral feeding the smaller the likelihood of recovery from this loss (ProGas Study Group, 2015).

Interestingly, some studies (Czell, Bauer, Binek, Schoch, & Weber, 2013; Kak et al., 2015; Kak et al., 2017; Sarfaty, Nefussy, Gross, Shapira, Vaisman, & Drory, 2013; Spataro et al., 2011) have reported that gastrostomy tubes are safe regardless of the FVC status, suggesting that even people with more significantly compromised respiratory functioning should still be considered for gastrostomy tube placement, especially if non-invasive ventilation is used during the procedure (Katzberg & Benatar, 2011). In addition, Lyall, Donaldson, Polkey, Leigh, & Moxam, (2001) did not consider FVC to be an accurate measure of predicting respiratory failure post gastrostomy tube insertion in MND as, according to the authors, patients with FVC as high as 75% may also develop respiratory failure.

The issue of non - oral feeding in MND is complex although a number of clinical indicators have already been established (Katzberg & Banatar, 2011), Plowman (2014) highlighted that the placement of a gastrostomy tube is a highly personal decision. Psychological readiness to accept non - oral feeding is a factor which should be

considered (Benstead et al., 2016). Johnson et al. (2012) emphasised that the loss of eating-derived pleasure can be a powerful indicator of a person with MND's readiness to accept non - oral feeding. Therefore, the authors proposed using a simple visual analogue scale to assess pleasure related to eating. Oliver and Turner (2010) advocated for the introduction of the topic of non - oral feeding shortly after the onset of bulbar symptoms in order to enable the patients to be actively engaged in the decision-making, rather than feeling under pressure when their respiratory function deteriorates. The NICE guidelines (2016) recommend discussing non - oral feeding at an early stage and ensuring that this discussion takes place at regular intervals "taking into account the person's preferences and issues, such as ability to swallow, weight loss, respiratory function, effort of feeding and drinking and risk of choking" (p. 21). The NICE guidelines (2016) acknowledge that not all people with MND will accept undergoing a gastrostomy; however, once a person decides to opt for non - oral feeding the procedure should be carried out without delay. The late insertion of a gastrostomy tube can contribute to an increased risk of mortality and procedural complications (NICE, 2016). There is no consensus to date with regard to the best timing for insertion of a gastrostomy tube (Katzberg & Benatar, 2011; Stavroulakis et al., 2013).

The Impact of Non - oral Feeding on Survival in MND

The existing literature investigating the benefits of non - oral feeding in MND is complex (Katzberg & Benatar, 2011) and the impact of artificial feeding on life prolongation is questionable (Simmons, 2005). Although non - oral feeding was reported to increase life expectancy by a few months in some studies (Chiò et al., 1999; Mathus-Vliegen et al., 1994; Pena et al., 2012; Plowman, 2014; Safarty et al., 2013;

Spataro et al., 2011), others reported no increase in survival (Russ, et al., 2015; Lu & Johnson, 2016).

The type of MND (bulbar versus spinal) seems to be considered an important factor by researchers concerned with measuring the survival rates post commencement of non - oral feeding, however, the correlation is not clear. The largest dataset to date (n = 2172), aimed at determining the impact of non - oral feeding on life prolongation in MND, revealed two main conclusions (Plowman, 2014). Firstly, the length of survival increased by 107 days on average in the group of people with MND who accepted non - oral feeding. Secondly, people presenting with spinal onset of MND had a longer survival time in comparison with people with bulbar onset of MND. In contrast, a study by Pena et al. (2012) concluded that although there were no differences in the length of survival of people with bulbar as opposed to spinal onset of MND, older age was, however, associated with poorer outcomes in bulbar onset MND. According to Pena et al. (2012) younger bulbar onset patients benefited most from non - oral feeding. The significance of age when establishing the relationship between life prolongation and non - oral feeding was also supported by the ProGas Study Group (2015). In addition, it is acknowledged, that non - oral feeding does not prevent aspiration pneumonia (Sorensen et al., 2007).

Jiménez et al. (2015) viewed non - oral feeding as an option which can potentially improve the QOL and well-being of a person with MND rather than increasing their survival. Oliver and Turner (2010) referred to a gastrostomy as "primarily a comfort measure" (p.3). The impact of artificial feeding on QOL in MND will be discussed in the next section (1.3.1).

Non - oral Feeding and Cognitive Impairment in MND

Although the literature exploring the benefits of non - oral feeding in the population of people with MND considered many relevant factors, such as the onset of the disease, nutritional status, respiratory function, and age, little emphasis was placed on the cognitive functioning of the person with MND. In relation to non - oral feeding, the NICE guidelines (2016) recommend that a person with MND and dementia undergoes a cognitive assessment by a neurologist in order to establish "ability to make decisions and to give consent; the severity of frontotemporal dementia (FTD) and cognitive problems; whether the person is likely to accept and cope with treatment [non - oral feeding]" (p. 21). To date, the benefits of non - oral feeding in the population of people with MND and FTD, in comparison to people with MND without any cognitive impairment, have not been investigated. These investigations appear to be important considering the existing literature focusing on non - oral feeding in dementia. The literature suggest that non - oral feeding in dementia is unlikely to positively impact on important clinical outcomes, such as a reduced risk of aspiration pneumonia or improved nutritional status (Finucane & Bynum, 1996; Finucane, Christmas, & Travis, 1999; Finucane, Christmas, & Leff, 2007; Kaw & Sekas, 1994), improved QOL (Gillick, 2000), and prolonged survival in this population (Finucane & Christmas, 2007; Mitchell, Kiely, & Lipsitz, 1997). Therefore, non - oral feeding in dementia should not be generally recommended (Murphy & Lipman, 2003; Gillick, 2000). It is not clear at present whether the process of recommending non - oral feeding for people with MND and FTD should be the same as that for people with MND without any cognitive impairment.

1.3. The Experiences of Dysphagia

In this section, the findings of the literature review pertaining to the experiences of dysphagia are presented. Studies involving both people diagnosed with dysphagia and their caregivers were reviewed. As the body of literature related to this topic is relatively limited, publications investigating the experiences of adults with dysphagia of various aetiology and their caregivers are included initially in this review, before focusing on MND in section 1.3.1.

The majority of publications identified investigating the impact of dysphagia were focused on the population of people with a carcinoma (Grant & Rovera, 1995; Penner, McClement, Lobchuk, & Daeninck, 2012; Roe, Leslie, & Drinnan, 2007; Tong, Lee, Yuen, & Lo, 2011; Watt & Whyte, 2003; Patterson et al., 2013; Nund, Ward, Scarinci, Cartmill, Kuipers, & Porceddu, 2014a, 2014c, 2014d, 2016). There appears to be a general consensus in the literature that dysphagia leads to psychosocial consequences and impacts on QOL (Balandin, Hemsley, Hanley, & Sheppard, 2009; Ekberg, Hamdy, Woisard, Wuttge-Hannig, & Ortega, 2002; Jacobsson, Axelsson, Osterlind, & Norberg, 2000; Klinke, Hafsteinsdóttir, Thorsteinsson, & Jónsdóttir, 2013; McCurtin, et al., 2017; Murry et al., 2016; Nund et al., 2014a, 2014b, 2014c, 2014d, 2016; Parker, Power, Hamdy, Bowen, Tyrrell, & Thompson, 2004; Roe et al., 2007; Swan, Speyer, Heijen, Wagg, & Cordier, 2015; Watt & Whyte, 2003; Tong et al., 2011). A range of negative emotions have been documented in these studies, such as fear (Jacobsson et al., 2000; Nund et al., 2014d; Watt & Whyte, 2003;); shame (Jacobsson et al., 2000); hopelessness (Bailey, 2004; Watt & Whyte, 2003;); embarrassment (Nund, 2014d; LaDonna et al., 2016); dissatisfaction about changed physical and social appearance (Jacobsson et al., 2000); anxiety (Ekberg et al., 2002;

Nund 2014d); frustration, loss, depression, disappointment, anger, and envy (Nund 2014d). Dysphagia has also been found to negatively influence dietary intake and reduce mealtime related pleasure often leading to weight loss (Ekberg et al., 2002; Watt & Whyte, 2003). Although non - oral feeding may improve a person's sense of control, it may also increase their anxiety (O'Farrell, Strong, Zou, & Rowe, 2005) and provoke feelings of lost independence (Holmes, 2004). Texture modifications, another common management option in dysphagia (RCSLT, 2014), were associated with a poorer QOL (Low, Wyles, Wilkinson, & Sainsbury, 2001; McCurtin et al., 2017; Swan et al., 2015), especially if they were more radical (Swan et al., 2015). Interestingly, one study reported that participants with dysphagia related to non-head and neck cancer self-selected softer food textures in response to their symptoms of dysphagia (Roe et al., 2007). Murry et al. (2016) described the impacts of dysphagia on QOL and they summarise their findings into three categories: (1) Functional limitations, such as limitations to the food types which a person with dysphagia can swallow safely, (2) Activities and participation, such as the inability to eat in a group setting, and (3) Environmental factors, such as eating in public, which can result in unwanted attention.

In order to understand people' experiences of dysphagia it appears important to acknowledge their perceptions of having to live with a swallowing impairment. Klinke et al. (2013) highlighted that living with dysphagia post a cerebrovascular accident requires constant readjustments. Although the literature researching perceptions of dysphagia is limited, it does suggest that people's attitude towards own dysphagia is individualised and may be disease specific. For example, the caregivers of people with myotonic dystrophy did not consider dysphagia to be problematic or have a big impact on the QOL of the people under their care. Instead, they reported fatigue or weakness

to be a more debilitating symptom of the disease with a significant impact on QOL (LaDonna et al., 2016). In comparison, both people with carcinoma (Watt & White, 2003) and their caregivers (Penner, McClement, Lobchuk, & Daeninck, 2012) found dysphagia to have a significantly negative influence on their lives.

Interestingly, one study investigating dysphagia related to nasopharyngeal carcinoma, highlighted differences in the participants' interpretation of dysphagia in comparison to the medical definition of dysphagia (Tong, Lee, Yuen, & Lo, 2011). Participants associated dysphagia with pharyngeal phase difficulties only while ignoring symptoms of oral stage dysphagia. In addition, the risk of aspiration was neglected and participants maintained their focus on cancer treatment options rather than their own dysphagia (Tong et al., 2011). The authors concluded that not having sufficient knowledge and awareness of dysphagia may lead people to underestimate their own swallowing impairment.

One study evaluated the perceptions of people with head and neck cancer in relation to dysphagia - related services (Nund et al., 2014c). This study highlighted the need for increased support with regard to alternations to food textures and the preparation of meals, guidance in selecting suitable foods, and practical tips for managing dysphagia. Nund et al. (2014c) recommended the development of new models of care which would address not only the physical but also the psychological and social impact of dysphagia following head and neck cancer.

Studies exploring the experiences of caregivers clearly suggest that providing care to someone with dysphagia alters their lifestyle, brings significant challenges, and greatly influences the social aspect of eating including dining out (Arai, 2005; Chadwick et al., 2006; Nund et al., 2014b; Patterson et al., 2013; Penner et al., 2012). The experience of caregiving for someone with dysphagia has been compared to a

journey: from the initiation of caregiving duties, through the acceptance of reality, the experiencing of the inner struggle related to the provision of food for a person with dysphagia, to the restructuring of life, and finally to a phase of finding a balance between the caregiving duties and own personal life (Arai, 2005). Although the caregivers are reported to adapt to a family member's dysphagia (Chadwick et al., 2006; Nund et al., 2014b), the emotional consequences of caregiving, such as frustration, sadness and worry, are frequently reported (Arai, 2005; A. Johansson, & U. Johansson, 2009; Nund et al., 2014b). The caregivers' role may include motivating the persons with dysphagia to eat (Patterson, et al., 2013) and the refusal to eat may be interpreted by the caregivers as a rejection of their support (Reid, McKenna, Fitzsimons, & Mc Cance, 2009). Food preparation for someone with dysphagia has also been found to be demanding (Chadwick et al., 2006; A. Johansson, & U. Johansson, 2009) and attempts to make modified food textures look appetising found to be frustrating (Patterson et al., 2013). Reid et al. (2009) reported on the food-related tension which could be present between people with cachexia (due to advanced cancer) and their caregivers. Penner et al (2012) highlighted changes in lifestyle and role within the family due to providing care to someone with dysphagia. Nund et al. (2014a) mapped the findings from their study investigating the impact of dysphagia on the caregivers of people with head and neck cancer to the International Classification of Functioning, Disability and Health (ICF). The ICF provides a well-established classification system for the description of health and health-related status (World Health Organisation, 2001). The results of this study by Nund et al. (2014a) revealed that dysphagia can be considered as an environmental factor impacting on the functioning of the caregivers. Nund et al. (2014a) reported that the caregivers most frequently expressed issues associated with the activities and participation domains of

ICF, for example, dysphagia was perceived to influence interpersonal relations between the caregivers and the person with dysphagia and it caused an increase in the caregivers' domestic responsibilities. The caregivers in the Nund et al. (2014a) study also reported that in order to manage dysphagia they had to learn and apply new knowledge and restrict their previous activities. In addition, dysphagia also appeared to influence the diet of the caregivers, as they avoided eating food which was unsuitable for the person with dysphagia and often chose to consume a modified diet. Similar findings were reported by Patterson et al. (2013), who investigated the population of caregivers managing dysphagia in people with head and neck cancer.

Finally, the positive association between the psychological health of caregivers and the outcomes for the person under their care has been recognised (Porter, et al., 2011). A holistic, family-centred approach to the management of dysphagia can therefore contribute to the improved well-being of the caregiver and more positive outcomes for the person with dysphagia (Nund et al., 2014a).

1.3.1 The Experiences of Dysphagia related to MND

This section focuses on the literature concerned with the lived experiences of dysphagia associated with MND. The literature published to date appears to be primarily focused on the experiences of non - oral feeding as an option to manage dysphagia in MND. No studies focusing on the experiences of other aspects of dysphagia in MND were found, for example, in relation to food texture modifications. There appears to be a paucity of research focused on the experiences of caregiving for someone with MND and dysphagia, although Trail, Nelson, Van, Appel, & Lai (2003) reported that dysphagia is one of the three top stressors for the caregivers of people

with MND. In the next sections, the literature describing the experiences of non - oral feeding and dysphagia - related health services in MND is presented.

1.3.1. a) Non - oral Feeding and the Decision Making Process

Decision making process in relation to non - oral feeding appears to be complex in MND. Greenaway et al. (2015) identified three key factors influencing a patients' decision to accept or reject non - oral feeding (or ventilation). These included (1) the guidance and information received from professionals, (2) the level of trust in the experts, and (3) the level of pressure from the professionals. Stavroulakis et al. (2014) suggested that an individual with MND may delay accepting a gastrostomy for a number of reasons, such as reluctance to give up oral feeding, uncertainty over disease trajectory, lack of awareness of potential benefits, and a negative perception of gastrostomy. These findings highlight the ability of professionals to significantly influence the decision of people with MND with regard to non - oral feeding. Other studies have also shown that decision-making related to the management options in MND generally depends on the relationship with health professionals and the support offered by them (Chiò, et al., 2012; Forbes, Colville, & Swingler, 2004; Hodgen, Greenfield, Nugus, & Kiernan, 2012a; van Resendaal, Verhoef, & Kinsella, 1999).

Greenaway et al. (2015) when investigating decision making process in relation to non - oral feeding, distinguished between: (1) patient-centred factors, such as perceptions of choice and control, acceptance and need, and aspects of fear; (2) external factors, such as the influence of health care professionals and family; and (3) the concept of time, for example, living in the moment and attempting to do the "right thing at the right time". Greenaway et al., (2015) reported that people with MND

attempt to live in the now and they appear to confront issues if they notice an obvious need to do so. Living in the moment and enjoying each day were the main coping strategies described by Gale (2015). This approach, however, curtails the ability to make decisions regarding the future, especially when that future is as unpredictable as it is for someone with MND (Greenaway et al., 2015). This may explain why in the study by Stavroulakis' et al. (2014) none of the participants chose to undergo an early gastrostomy, despite professional recommendations, as they were still managing their eating and drinking relatively well.

The participants in the study by Stavroulakis et al. (2014) may have benefited from the provision of increased guidance, information, and direction from their health professionals in order to understand the potential benefits of non - oral feeding. On the other hand, Stavroulakis et al. (2014) highlighted the challenge for health professionals involved in the decision making process regarding non - oral feeding in MND due to the absence of robust evidence regarding the benefits of gastrostomy in this population.

Martin et al. (2016) investigated the decision-making process of people with MND regarding non - oral feeding and non-invasive ventilation from the perspective of the health care professionals. The professionals highlighted a number of factors which they perceived as influencing a patients decision to accept or decline these interventions, with the most important being the perceived effect on QOL and the desire to prolong life. The perception of physical need was also reported as being important with some people with MND agreeing to an intervention when they were already experiencing significant symptoms of respiratory compromise or dysphagia (Martin et al., 2016). In the same study, acceptance of the disease was considered a prerequisite for patients' acceptance of interventions including non - oral feeding. This

finding was also reported by Greenaway et al. (2015) suggesting that the perceptions of people with MND and their health care professionals may be similar in this regard. The influence of third parties, such as caregivers, family, and health care professionals, on the person with MND's decision to consent to non - oral feeding or non-invasive ventilation was also reported by Martin et al. (2016). In some circumstances, both the person with MND and their caregiver were perceived to be equally involved in the decision making process. In addition, the people with MND appeared to consider the impact of the interventions on their family.

The professionals (Martin et al., 2016) reported feeling responsible for providing guidance regarding interventions and they recognised the need for an individualised approach which would take into account patients' and families' coping strategies, emotional state, level of acceptance of the disease as well as the physical condition and the level of progression of MND. However, the issue of timing was reported to be extremely challenging in relation to when to initiate the discussion about interventions and when to make professional recommendations for the intervention itself.

1.3.1. b) Living with Non - oral Feeding

Non - oral feeding is a frequently recommended dysphagia management option in MND (R. Miller et al., 2009; Katzberg & Benatar, 2011) as it contributes to stabilising body weight (Kasarskis, Scarlata, Hill, Fuller, Stambler, & Cedarbaum, 1999; R. Miller et al., 2009; Katzberg & Benatar, 2011) and improving QOL for some people with MND (Körner et al., 2013; Rickman, 1998; Simmons, 2005). However, significant challenges related to non - oral feeding, which could potentially decrease

the QOL of a person with MND, have also been reported (Stavroulakis et al., 2016). According to Stavroulakis (2016) these complications are multiple and include clinical complications, psychological impact, time burden, difficulties with administering feeds, handling gastrostomy-related equipment, supplies and storage, dressing, bathing, and tube discomfort In Stavroulakis' et al., (2016) study, participants reported the positives related to non - oral feeding including a reduction in anxiety related to prolonged meals, the stabilisation of weight, and perceived life prolongation. However, participants also reported the negative aspects such as increased anxiety related to keeping the tube intact, the loss of the social aspects of meals, such as eating with a person with MND, and social isolation. Interestingly, although Stavroulakis et al. (2016) identified more challenges than benefits related to non - oral feeding in MND, the study concluded that the establishment of a new and safer alternative route for eating, drinking, and taking medications, was perceived by both people with MND and their caregivers as outweighing the issues. According to the authors, positive perceptions of non - oral feeding, despite so many challenges reported, could be influenced by participants' hope of increased survival "in the absence of alternatives" (pg.59).

Another study (ProGas Study Group, 2015) reported that non - oral feeding had no effect on the QOL of the person with MND, however, it caused a significant increase in caregiving duties. The authors concluded that the health services should offer support before and after gastrostomy not only to the people with MND, but also to their caregivers. Olsson, Markhede, Strang, & Persson (2010) stated that in order to improve the care of both the person with MND and their caregiver it is crucial to examine them in parallel over time (Olsson et al., 2010b). Stavroulakis et al. (2016) suggested training should be provided: (1) at the time of referral for a gastrostomy, (2)

post-gastrostomy procedure in hospital and (3) after discharge home to recap on the training provided in hospital.

Overall, there appears to be limited evidence in relation to QOL and non - oral feeding in MND. Katzberg and Benatar (2011) in their Cochrane review concluded that the effects of non - oral feeding on the QOL of people with MND and their caregivers needed to be researched in controlled studies, as it is a critical area of importance when considering non - oral feeding. Knowledge of patients' experiences prior to, during, and post insertion of a gastrostomy tube was reported to be essential for the development of guidelines in relation to the commencement of non - oral feeding in MND (Benstead et al., 2016) and the relationship between non - oral feeding and QOL in MND was considered to be a crucial argument for undergoing a gastrostomy (Katzberg & Benatar, 2011).

1.4 Chapter Summary

This literature review aimed to provide up-to-date evidence in relation to selected aspects of MND and dysphagia. Currently, there is no cure for MND and professional input is focused on symptom management. Dysphagia is recognised as one of the most common consequences of MND. Moreover, the impact of developing dysphagia associated with MND extends beyond serious medical complications, as there are reports of negative psycho-social influences, strong emotions, and deterioration in swallowing-related QOL. These consequences of dysphagia appear to affect not only people diagnosed with dysphagia, but also their caregivers. Surprisingly, studies researching the experiences of dysphagia in MND from the caregivers' perspective are limited. This appears to be significant considering that

many people with MND require a high level of care. The caregivers can be the key person responsible for the implementation of specific dysphagia management techniques, such as appropriate diet preparation or administration of non - oral feeding.

The current management of dysphagia in MND seems to be primarily compensatory, although this area appears to be significantly under-researched. Although previous research has focused on investigating experiences related to specific dysphagia management options (such as gastrostomy), there are no reports focusing on other common management techniques, for example, the experiences and perceptions of specific dysphagia compensatory management techniques (such as changes in food textures, modified postures and positions during meals, or the use of specialised utensils for eating and drinking). Moreover, the clinical relevance of therapy for dysphagia in MND is yet to be established.

As MND progresses non - oral feeding may be recommended mainly as a solution for weight loss. However, the literature exploring the experiences of non - oral feeding in MND highlights the strain related to managing the gastrostomy tube at home. The evidence clearly indicates that there is a need for a greater amount of direction and support from health professionals in relation to non - oral feeding and that the relationship between people with MND and their health care professionals is very important during the decision-making process regarding gastrostomy.

Although the experiences of dysphagia in other populations (such as those with head and neck cancer) are more frequently reported in comparison to MND, it would not be appropriate to translate those findings to the population of people with MND, as they appear to be disease specific. Therefore, there is a clear gap in the body of knowledge in relation to understanding the experiences of dysphagia in MND,

especially in relation to how dysphagia impacts on the lives of people with MND and their caregivers, how they perceive dysphagia, and what their main coping strategies are in relation to the swallowing impairment. Moreover, an investigation into the experiences of dysphagia - related health services could potentially influence clinical practice and adherence to the professional management of dysphagia.

CHAPTER 2: RESEARCH PROCEDURE

Introduction

This chapter presents the methodological considerations of the study, introduces Interpretative Phenomenological Analysis (IPA), and provides justification for choosing this approach. It then focuses on the research design including methods, the recruitment of participants, data collection, and data analysis. The pilot study and its influence on the final research design is also explained. This chapter is concluded by describing my positionality as the researcher and presenting my presuppositions and how they evolved during this project.

2.1 Methodological Underpinnings

This section presents the rationale for choosing a qualitative research design and summarises my epistemological stance. It later introduces Interpretative Phenomenological Analysis (IPA), the qualitative approach employed in this study, and explains its theoretical underpinnings.

2.1.1 Rationale for a Qualitative Research Design

In order to address my research interest (the experiences of dysphagia in MND) I decided to employ a qualitative methodology, which focuses on the exploration of social and human problems and experiences (Creswell, 2013). The strength of qualitative research derives significantly from its orientation towards the world, its concern with specific situations or people, and its emphasis on descriptions rather than numbers (Maxwell, 2013). Rather than investigating a specific hypothesis, as

quantitative research does, qualitative methods generally ask broad questions (Fossey, Harvey, McDermott, & Davidson, 2002). In this project the broad question was: How do people living with MND[3] experience and understand dysphagia? As qualitative methodology provides scope for the generation of contextual and explanatory data (Ritchi & Lewis, 2003), it was considered appropriate to address the above research question.

As a novice qualitative researcher I firstly engaged in building the theoretical base by reviewing the literature pertaining to qualitative methodologies and research design, attending relevant training courses, and generating and exchanging ideas with the Supervisory Team. The initial step of acquiring knowledge related to qualitative research involved the establishment of my epistemological stance.

2.1.2 Epistemology

Epistemology can be defined as the relationship between the researcher and reality (Carson, Gilmore, Perry, & Gronhaug, 2001) and how this reality is understood. A subjective epistemology was employed whereby the reality is believed to be constructed between the researcher and the participants and shaped by individual experiences (Creswell, 2013). Therefore, the reality is understood to be multiple and relative (Hudson & Ozzane, 1988, Neuman, 2000). I accept (in line with an ontological perspective) that reality is constructed through our lived experiences and interactions with others (Robson, 2011). I do not believe that it would ever be achievable to remove ourselves, our perceptions, thoughts, and experiences, from the world, in order to

[3] The term: "People living with MND" refers to both people diagnosed with MND and their caregivers throughout this thesis

reveal some definitive "reality". I do, however, also recognise that I am not a pure relativist (i.e. I do not believe that the truth and the reality is different for different people and that universal and objective truth does not exist at all) and that in fact my personal approach is closer to being a 'minimal hermeneutic realist' (Dreyfus, 1995).

The concept of minimal hermeneutic realism was first found in 'Being and Time' (Heidegger, 1927/2017) in which Heidegger[4] stated that intelligibility of each kind of thing depends upon humans' ability to make sense of things. This theory was later summarized by Larkin, Watts, & Cliffton (2006), "What is *real* is not dependent on us, but the exact meaning and nature of *reality* is" (p. 107). Minimal hermeneutic realism recognized that some aspects of reality exist and would have existed even if humans had not. Those aspects could be revealed as 'real' only by humans, who encounter them by bringing them meaningfully into their lives (Polt, 1999). For the reasons presented above, the epistemological stance taken within this study appears to fall somewhere between constructivism [5] (which considers reality to be an invention of a particular society or culture, as the social world is created by human interactions and language; Hirschman, 1985; Berger & Luckman, 1967 in Hudsin & Ozanne, 1988; Kely, 2008), and critical realism [6] (which considers reality to be independent of human consciousness and thoughts, but that the meaning of this reality is socially constructed; Archer, 1998; Bhaskar, 1978; 1998; Easton, 2010; Fletcher, 2017; Mingers & Willcocks, 2004; Morton, 2006; Oliver, 2012). Although I believe that human experiences are grounded in a socio-cultural context, my primary interest is to investigate the unique perspective of individuals in order to understand how they each

[4] Heidegger's philosophy is further explained in the section 2.1.4 a)
[5] The terms social constructionism and constructivism appear to be used interchangeably under general term 'constructivism' (Charmaz 2000, 2006).
[6] The term critical realism originated from a combination of Bhaskar's transcendental realism and critical naturalism (Sayer 2000).

perceive and experience a phenomenon. My epistemological approach is consistent with IPA (Smith, Flower, & Larkin, 2009) which was employed to fulfill the research aim and find answers to the research questions.

2.1.3 Interpretative Phenomenological Analysis (IPA) - An Introduction

IPA was first introduced by Jonathan Smith in 1996 (Smith, 1996) as a qualitative approach concerned with the detailed exploration of personal lived experiences and the examination of how people make sense of important life experiences. IPA acknowledges that participants' interpretations of their experiences are grounded in their socio-cultural context and it aims to understand the experiences from the perspective of the participants. Therefore, IPA is considered to be phenomenological, as its central focus is a detailed examination of an individual's experiences as they emerge in their own terms, as opposed to examining the experiences using "predefined category systems" (Smith et al., 2009, pg. 32). IPA is also idiographic as it maintains the focus on individual participants even if the study involves a group of participants. IPA is concerned with an individual in terms of the level of detail and depth of analysis. According to Smith et al. (2009) IPA studies offer "detailed, nuanced analyses of particular instances of lived experience" (p.37). In IPA the analytical process moves from single cases to cross case analysis to reveal overarching statements.

IPA has a clear methodological stance and according to Shinebourne (2011) it "proposes a middle way between different qualitative methods" (p.45). IPA is considered to be a distinctive qualitative psychological methodology (Shinebourne, 2011a). While it allows for flexibility and creativity it also proposes a framework for

the process of data analysis. Therefore, I understand IPA as both a qualitative methodology and a method in itself. Smith, Jarman, and Osborn (1999), in one of their articles referred to IPA as a "qualitative approach", "not a prescriptive methodology", and "a qualitative method" (p.239), which supports my understanding of IPA as being both a qualitative methodology and a method.

IPA is particularly suitable for investigating unique, meaningful, important, and complex lived experiences, and was originally used in health psychology. IPA has gained significant interest internationally within other disciplines, for example, nursing (Carradice, Shankland, & Beail, 2002), midwifery (Charlick, Pincombe, McKellar & Fielder, 2016), mental health (Todd, Simpson, & Murray, 2010), education (Jeong & Othman, 2016), and sports studies (Callary, Rathwell &Young, 2015).

The decision to implement IPA took a substantial amount of time and consideration. Initially, I engaged in the process of learning about various qualitative approaches and evaluating their potential suitability for this research. As this was my first experience of IPA I underwent formal training to expand my skills, and also liaised with experts in IPA both nationally and internationally.

The literature was reviewed to establish how IPA had previously been applied in the field of MND, dysphagia, caregiving, and SLT. Three IPA studies related to MND were found: information seeking behavior (O'Brien, 2004), reasons behind refusing non-invasive ventilation (Ando et al., 2015) and exploring the transitional process from diagnosis to living with MND (Mistry & Simpson, 2013). No published study was found to have used IPA in order to investigate dysphagia. There were over 50 studies which had implemented IPA in order to investigate the population of

caregivers, with the largest number of these publications being focused on mental health, dementia, and intellectual disability. No IPA study involving caregivers of people with MND was found. Three IPA studies related to SLT were revealed: two focused on aphasia (Brown, Worrall, Davidson, & Howe, 2011a, 2011b), and another on bilingualism within the deaf and the hearing world (Fort & Kent, 2013). This review concluded that IPA was a relatively new approach in the field of SLT and it did not appear to have been used to investigate dysphagia previously.

2.1.4 Theoretical Underpinnings of IPA

Phenomenology, hermeneutic, and idiography form the theoretical foundations of IPA (Smith et al., 2009). This section begins by presenting a synthesis of phenomenology and the key points from the work of Husserl, Heidegger, Merleau-Ponty, and Sartre. It then introduces hermeneutics and the main points in Schleiermacher's, Heidegger's and Gadamer's philosophy, as relevant to IPA. Finally, a short description of idiography is presented before explaining how the philosophy of IPA relates to this research.

2.1.4. a) Phenomenology

Phenomenology is concerned with the exploration, analysis, and understanding of human experiences and perceptions of phenomena, where phenomena refers to things or experiences as human beings experience them (Creswell, 2013; Finlay, 2009; Langdridge, 2007; Moran, 2000). "Things" are understood, as much as it is possible, by the way they appear (or "are given" in phenomenological terms) to the experiencing person (Zahavi, 2003). According to

Pollio, Henley, & Thompson (1997) the goal of phenomenology is to provide "a rigorous description of human life as it is lived and reflected upon in all of its first-person concreteness, urgency, and ambiguity" (p.5). Its primary position is that the most basic human truths are accessible only through inner subjectivity and consciousness and that the world and the people in it are intimately constituent (Burns & Grove, 1999; Creswell, 2013; Thorne, 1991). IPA employs the core ideas of Husserl, Heidegger, Merleau-Ponty, and Sartre (Smith et al., 2009). These core ideas are summarised in the subsections below.

Edmund Husserl

Edmund Husserl (1859 -1938) was a German philosopher, who is considered to be the founder of descriptive phenomenology. Husserl's concept of *natural attitude* implies that in order to understand phenomena the researchers have to re-evaluate their knowledge and deliberately suspend their judgements about the world around them (*époche* or *bracketing*) in order to access a phenomenon without presuppositions (Creswell, 2013; Drew, 1999; LeVasseur, 2003; Robson, 2011). Husserl (1970) introduced the idea of the *Lebenswelt* (*life-world*) which consists of specific structures. We are all body-subjects as we all have a sense of embodiment and self. *Life-world* consists of different forms of sociality and involves: a sense of spatiality as we are inserted into the world that surrounds us, and temporality as we live in the present, with a determining past and future, which is yet to be determined. Husserl's phenomenological inquiry is concerned with the experience as it is present in the consciousness of a person. In Husserl's terms the experience is always *intentional* as it is related to the object of attention (our consciousness and perception of the world are always directed to our object of attention; Dworkin, 2003).

Husserl's method of identifying the core features of human experience (the *essence*) involves a series of *reductions*. Husserl recommends that in order to investigate the human experience the researcher has to *bracket* previous judgments and presuppositions (Lewis & Staehler, 2011). The *eidetic reduction* involves repeated dwelling and immersing in the experience in order to discover its *essence* (Finlay, 2011; Smith et al., 2009). The *transcendental reduction* involves focusing on the nature of consciousness to discover what enables our consciousness of anything (Smith et al., 2009).

Husserl "sets the agenda for the attentive and systematic examination of the context of consciousness" (Smith et al., pg.16) in order to discover the *essence* of it. The focus of this study was on investigating participants' understanding of dysphagia via the interpretation of their lived experiences. Therefore, Husserl's philosophy was relevant as it established the importance of human experiences and perceptions.

Martin Heidegger

Heidegger (1889 -1976) was also a German philosopher, who worked as Husserl's assistant in the early stages of his career. Heidegger's phenomenology, which is interpretative, places a central focus on lived experiences (Creswell, 2013; Finlay, 2011; Gallagher, 2012; Heidegger, 2017; Moran, 2000). According to Heidegger, existence always refers to a person, therefore it is personal and owned (Moran, 2000). Heidegger referred to a human being as *Dasein* (or *Being-in-the-world*). *Dasein* is a *Being*, who is aware of their *Being* and the meaning of this *Being* (Heidegger, 2017). *Dasein* is existence itself and humans are thrown into the world and cannot be meaningfully separated from it (Finlay, 2011; Heidegger, 2017).

Heidegger's concept of *Geworfenheit* (*thrownness*) denotes the arbitrary nature of *Dasein*. As Heidegger was interested in the meaning of being and what it is to be human, he considered the world to consist of two parts: phenomenon and logos (Heidegger, 2017; Moran, 2000). For Heidegger, the phenomenon has two dimensions as things have both visible and concealed meanings. Heidegger interprets the phenomenon of our being as entering a new state, presenting itself to us, coming forth. Phenomenon is primarily perceptual whereas logos is analytical. The main aim of Heidegger's phenomenology is to examine things as they appear to show themselves to us. Phenomena cannot be understood without attending to their *Being-in-the-world*. Phenomenology is therefore a methodology for discovering (uncovering) phenomena in the world. Although the phenomenon appears almost spontaneously, the logos helps us to grasp this appearance in order to make sense of it (Smith et al., 2009).

For Heidegger, language and understanding cannot be separated as it is only through language that we can understand our *Being-in-the-world.* (Finlay, 2011). However, when humans make interpretations they are not free of their pre-suppositions and pre-experiences, which re-evaluates Husserl's idea of époche (Heidegger, 2017). Therefore, for Heidegger (2017) there is no presuppositionless starting point because human's interpretations are grounded upon their prior experiences (*fore-having*) and they are always in some way *fore-given* (Heidegger, 2017). As reported by Finlay (2011), we (humans) start making interpretations with a pre-understanding and "move on to being open to discovering something" (p. 53). In order to develop a deeper understanding of a phenomenon we engage in a process of reinterpreting our initial presuppositions, revising them, and then challenging the new interpretation.

Maurice Merleau-Ponty

Phenomenology evolved further through the work of the French philosopher Merleau-Ponty (1908 -1961). Merleau-Ponty suggested in his "Phenomenology of Perception" (Merleau-Ponty, 1962 as cited by Finlay, 2011) that people see themselves as different from anything else in the world. This is due to humans' engagement in looking at the world rather than being incorporated within it. Merleau-Ponty concentrated on *embodiment* as, in his view, the understanding of the world comes from one's embodied relationship with the world. The body is not considered to be an object existing in the world, but it is our means of communication with the world (Merleau-Ponty, 1967). Merleau-Ponty referred to the body as "the vehicle of being in the world" (Merleau-Ponty, 1962 pg. 82 as cited by Finlay, 2011). Therefore, experiences belong to one's own embodied position in the world and ultimately cannot be entirely shared with other person. The *subjective body* is ME, I do not possess my body, I AM my body. My experience belongs to my embodied position in the world and my body shapes my knowledge about the world. The *objective body* is the body that we can observe and objectify, for example, someone else's body or our own body at a particular time, such as during an illness (Finlay, 2011). According the Smith et al. (2009), Merleau-Ponty's concept of the body as the central element in experience is critical to IPA and has to be considered when investigating lived experiences.

Jean-Paul Sartre

Sartre (1905-1980) was a French philosopher, whose work advanced the development of existential phenomenology. Sartre emphasized that people engage in the world through their self-consciousness and that we are constantly becoming

ourselves. For Sartre, humans have the freedom to make choices and are responsible for their actions, but these actions have to be considered from the context of individual circumstances. Sartre's concept of *nothingness* implies that our being and our perception of the world are influenced not only by things that are present, but also things that are absent (Catalano, 1985). For example, when people anticipate seeing an object in a given environment, but this object is unexpectedly absent, this absence alters people's perception of the environment. People's perceptions of the world are also considerably influenced by the presence of other humans, their own experiences, and the type of relationships formed with them (Finlay, 2011; Smith et al., 2009).

2.1.4. b) Hermeneutic

Hermeneutics, which initially developed from the interpretation of biblical texts and was subsequently considered to be a philosophical theory of interpretation (Smith et al., 2009), is now introduced. Schleiermacher, Heidegger, and Gadamer are considered to be the most important hermeneutic philosophers (Smith et al., 2009).

Friedrich Schleiermacher

Friedrich Schleiermacher (1768-1834) was a German philosopher, who is considered to be the father of modern hermeneutics (Jasper, 2004) due to his work in interpretation and translation, where he transformed the traditional Biblical hermeneutic into hermeneutics of any text (Enns, 2014). Schleiermacher emphasized two aspects of interpretation: grammatical and psychological. The grammatical understanding of text is achieved when words are examined in relation to sentences,

sentences to paragraphs, and so on. This implies back and forth movements, known as a *hermeneutic circle*.

We cannot understand the meaning of the whole text apart from understanding the meaning of the individual sentences, and even words, in the text. On the other hand, we cannot properly understand the individual parts apart from some grasp of the whole (Stiver, 1996, pg. 89).

This psychological interpretation is an attempt to enter the mind of the original author or speaker in order to understand the meaning of that text to him / her, which requires the interpreter to familiarise themselves with details of the author's life (Stiver, 1996). For Schleiermacher, interpretation and translation are an art as the interpreter does not follow a set of rigid rules (Smith et al., 2009).

Schleiermacher's philosophy in the context of IPA research highlights the relationship between the author of text (or the author of the interview) and the researcher and the researcher's ability to offer their own unique perspective on the text (Smith et al., 2009).

Martin Heidegger

Although some of Heidegger's phenomenology has been already presented in relation to phenomenology, the focus of this section is on Heidegger's involvement in hermeneutics. Heidegger is reported to "link phenomenology with hermeneutics" (Moran, 2000, p.229). Heidegger further developed the concept of a *hermeneutic circle*, as he connected it to the whole process of understanding a phenomenon through interpretation (Heidegger, 2017). During a hermeneutic circle the initial understanding of human experience, described as a "rough and ready approximation" (Finlay, 2011,

pg. 53), is constantly challenged as the researcher moves between their pre-understanding and understanding. The revision of the initial understanding is interpretative and the understanding deepens as the researcher goes around in the circle repeatedly (Heidegger, 1962; Polt, 1999).

Heidegger's philosophy, where phenomenology is explicitly interpretative and connected to hermeneutics, is one of the key components of IPA. Smith (2004) referred to *double hermeneutics* as during IPA the researcher attempts to make sense of the participant's sense-making in relation to the investigated experience. The *hermeneutic cycle* forms an important part of IPA. It is understood to be a dynamic relationship between the part and the whole, for example a single word is related to the whole sentence, a single extract to the full text, a single interview to the complete research project. The process of analysis is therefore iterative (Smith & Osborn, 2003, Smith 2004, Smith et al., 2009). Being able to move back and forth within the data allows for a review of the data at different levels and the building of a deep understanding based upon multi-perspectival considerations.

Hans-Georg Gadamer

Another German philosopher, Hans-Georg Gadamer (1900-2002), built on Heidegger's work. Gadamer added the concepts of *prejudgments* or *horizons* which refer to the set of prejudices people possess (Gadamer, 2004). He pointed out that *prejudgments* should not be viewed in a negative and limiting way, as they are alterable and can promote understanding (Gadamer, 2004; Moran, 2000). In fact, understanding occurs through the development of a new *horizon* via *pre-understanding*, *prejudice* and *fore-meaning* (Gadamer, 1966); when our present

horizon moves to a different or new horizon by an encounter (termed as *fusion of horizons*); and by *universality* (a connection between people's expressions and understandings; Gadamer, 2004). According to Gadamer, (2004) understanding has an essential and fundamental connection (*belongingness*) with language and language will always be biased due to pre-existing cultural influences.

Gadamer's claim that all understanding is situated in the historical and cultural context (Gadamer, 1966) means, in the context of IPA, that interpretations are always influenced by the moment when they were made (Smith et al., 2009).

2.1.4. c) Idiography

The term idiography comes from the Greek word "idios", which means "own" or "private". The idiographic approach is concerned with an individual. Malim, Birch, & Wadeley (1992) stated that the idiographic approach (focused on an individual) allows for an in-depth exploration of a participant as it considers the uniqueness of every person. Applying idiography in research can be summarized in Skinner's words: "Instead of studying a thousand rats for one hour each or a hundred rats for ten hours each the investigator is more likely to study one rat for a thousand hours" (Skinner, 1996 p.21).

According to Smith et al. (2004, 2009) idiography had a considerable influence on IPA. IPA is committed to an in-depth focus on an individual and aims to deliver a detailed analysis of actual life and lived experiences.

2.1.5 Relating the Philosophy of IPA to this Project

I would like to synthesise the relevance of the above philosophical concepts and beliefs to this research project. First of all, my interest is phenomenological, as I aim to explore the experiences of a particular group of people (people living with dysphagia) in a particular context (MND). My aim is to reveal the *Being-in-the-world* of people living with dysphagia caused by MND (Larkin et al., 2006; Smith et al., 2009). I am interested in attending to the things that individuals are actively engaged with and how these influence or shape their experiences in the world, and the meaning they hold for them. The search for the meaning of dysphagia in MND via the process of interpretation is an important element of my research. I view the idiographic approach and the hermeneutic circle as opportunities to achieve an in-depth understanding of participants' experiences. I see language as the fundamental tool of interpretation. I believe that humans have the ability to make choices and can respond to their realities in various ways (Husserl, 1962). Marleau-Ponty's concept of experiences belonging to our own embodiment position in the world is also relevant to this study, as living with a deteriorating body forms a central element in the meaning-making experiences of the participants.

2.2 Research Methods

This section presents the recruitment procedure and the process of data collection and data analysis. It also describes the development of the pilot study and how this pilot influenced the research design. Research ethics and consent are also discussed. Finally, the criteria implemented to assess the quality of this project are presented followed by a statement of the researcher's positionality.

2.2.1 Research Design

This study employed qualitative, multiple, and in-depth interviews as its primary source of data collection. The interviews were conducted with people diagnosed with MND and dysphagia and also with family caregivers (separately) to explore their perceptions and experiences of living with dysphagia as part of MND. Although the interviews formed the basis of data collection, they were supported by field notes and observations in order to assist in the development of more detailed accounts of the phenomenon (Smith et al., 2009). This is further expanded upon in the data collection section (2.3).

2.2.2 Study Sites

The geographical boundary chosen for this study was the South-West of Ireland. The majority of participants (16/20) were recruited from the Irish Motor Neurone Association [IMNDA] and the remainder from Speech & Language Therapy [SLT] Clinics in Ireland. Most participants were interviewed in their own homes (17/20), two in a palliative care centre, and one in her workplace. A flexible approach was employed in relation to offering a choice of date, time, and venue for the meetings.

2.2.3 Population and Sample

Interviews were conducted with people diagnosed with MND and dysphagia and caregivers of people with MND and dysphagia. All those with MND had accessed SLT services in relation to their dysphagia and were experiencing changes in their eating, drinking, and swallowing in comparison to pre-MND. All caregivers were

involved in the management of dysphagia associated with MND. The enrollment of person with MND-caregiver dyads was not required, as the aim was to collect the data and analyse it for the two groups separately.

2.2.3. a) Sample Size

In adherence to the IPA approach to data collection, the aim was to purposively select participants who met the study's eligibility criteria and were willing to provide insight into their experiences of living with dysphagia and MND (Smith et al., 2009). Ten people with MND and ten caregivers were recruited for this study. This sample size allowed the idiographic focus to be preserved and prevented the analysis from being too general or descriptive. Specific factors which could have potentially influenced the data collection were accounted for, for example, obtaining limited data from some people with MND due to their poor health status or limited ability to communicate. In line with IPA's recommendations (Smith et al., 2009), the aim was to keep the sample homogeneous, and a set of eligibility criteria was therefore developed for each group.

Prior to the commencement of the study the researcher aimed to ensure the probability of finding the required number of participants (20) within the specified geographical area. At the time of research design there were 335 people with MND registered with IMNDA (as per IMNDA database accessed on the 24th of October 2013). This included 63 people within the South -West of Ireland. According to Mardsen, 70% of the MND population experience dysphagia (Mardsen, 2011). Therefore, it was anticipated that at least 44 people with MND and dysphagia could be identified within the chosen geographical area. It was also anticipated that most of

these individual's would require a caregiver. In the event of sufficient suitable participants not being found within the specified geographical area the researcher was prepared to expand the study nationally. This however, was not required.

2.2.3. b) Eligibility Criteria

A number of specific inclusion criteria were developed for both groups, such as age (over 18), willingness to participate in the research, and the ability to provide informed consent. Participants were required to consent to their data being audio recorded, summarised and analysed by the researcher in consultation with the Supervisory Team. Additional criteria were developed for the population of people with MND and the caregivers.

The People with MND

A gap of at least two months between diagnosis of MND (confirmed by a Neurologist) and the date of the initial interview was required, to ensure participants were given some time to adjust to their diagnosis.

People with MND were eligible if they presented with normal or mildly impaired cognition. This was assessed using the Edinburgh Cognitive and Behavioural ALS Screen [ECAS] (Abrahams, Newton, Niven, Foley, & Bak, 2014).

The diagnosis of dysphagia was required to have been made by a SLT at least 1 month prior to the initial interview to ensure all participants had already been informed about their dysphagia and had received professional advice in relation to

dysphagia management. In addition, the Functional Oral Intake Scale[7] [FOIS] (Crary, Carnaby Mann, Groher, 2005) was utilised and people with MND who self-reported as being at levels 1 - 5 were eligible for this project. FOIS levels 1 - 5 included people who were fully and / or partially dependent on non - oral feeding in addition to people eating a full oral diet prepared in a special way, for example, by modifying the texture of food.

In order to ensure participants' ability to take part in the interviews, functional communication skills were required. An evaluation of verbal skills was carried out using selected parts of the Sentence Intelligibility Test from *Assessment of Intelligibility of Dysarthric Speech* (Yorkston & Beukelmand, 1981). A set of ten sentences was chosen based on their phonemic variability to achieve a representable sample of speech. In addition, verbal people with MND with no AAC support were deemed eligible if rated at levels 10 – 6 of the *ALS Severity Scale: Speech* (Hillel, Miller, Yorkston, McDonald, Norris, & Konikow, 1989)[8]. If a person's voice volume was poor a voice amplifier was available. Persons with significant dysarthria (below level 6 of the *ALS Severity Scale: Speech*) were eligible if they had effective means of augmentative or alternative communication [AAC] which allowed them to express themselves at sentence level.

In order to rule out any potential medical contraindications for taking part in this research, the gatekeepers (MND Nurses, SLTs, or Neurologists) were asked to confirm participants' ability to engage in multiple interviews.

[7] Appendix 2
[8] Appendix 3

The Caregivers

Caregivers were required to regularly be providing dysphagia - related care to a person with MND, for example, diet preparation, supervision during meals, or feeding. They could not have any professional background in the management of dysphagia.

Caregivers were required to have normal cognitive status, which was assessed using the 6-Item Cognitive Impairment Test (Katzman, Brown, Fuld, Peck, Schechter, & Schimmel, 1983). This test is brief, simple, and demonstrates higher sensitivity then the Mini-Mental State Exam (Brook & Bullock, 1999).

Caregivers were not restricted to being a member of the person with MND's biological family.

2.2.3. c) Recruitment Procedure

Working within ethical guidelines participants were recruited via professionals (SLTs and MND Nurses), who handed a written invitation and information about the project to potential participants[9]. Participants were asked to contact the researcher directly regarding enrolment, or alternatively to give permission for the professional to pass on their contact details to the researcher. Six potential participants contacted the researcher directly requesting further information about the study. Twenty potential participants were contacted by the researcher to confirm their interest in taking part, provide answers to any study-related questions, and to schedule the initial meetings. There was a gap of at least 48 hours from when the person was given the

[9] Appendix 4

invitation pack to when the initial contact was made to allow them time to consider their participation.

The six people who contacted the researcher confirmed their interest in taking part in this study. The majority of people who were contacted by the researcher (18/20) also expressed their interest in participating. Two remaining people (caregivers) declined the participation on behalf of the person with MND under their care. Both expressed concerns regarding poor stamina of the persons with MND. Additionally, one person with MND who expressed an interest in taking part in the study, became very unwell and passed away prior to the first interview being scheduled.

At the initial meeting informed, witnessed consent[10] was obtained, which was recorded and kept for future reference, and an assessment of suitability took place. Twenty-three people in total were assessed against the eligibility criteria (ten caregivers and thirteen people with MND) and three people with MND were excluded due to their moderate level of cognitive impairment. The remainder were deemed eligible and invited to participate in the first interview. For the majority of caregivers (7/10) the first interview took place immediately following their assessment of eligibility, whereas three others were interviewed at subsequent meetings. Fewer people with MND expressed their readiness to participate in the first interview following the assessment of eligibility criteria (4/10) and therefore the majority (6/10) were interviewed at subsequent meetings.

When scheduling meetings with the people with MND the ALS Health State Scale (Riviere, Meininger, Zeisser, Munsat, 1998) was to be used in order to prioritise participants according to their level of impairment and starting interviews with those

[10] Appendix 5

most affected by MND. However, this was not required as the recruitment naturally followed a sequential order.

No attempt was made to recruit patient-caregiver dyads as, in accordance with the idiographic approach, the aim was to reveal individual experiences of each participant first before moving to the cross-case analysis of each group. However, for convenience reasons, the recruitment of the caregivers of people with MND who had also consented to participate in this project, was prioritized. The final sample consisted of seven caregiver-person with MND dyads and the remaining three people with MND and three caregivers were unknown to each other.

All participants were informed about their right to withdraw their participation at any stage and without any consequences, and of the possibility of having their data destroyed at their request (for up to two weeks after the final interview). However, none of the participants withdrew their wish to participate or stated their wish to have their recordings destroyed.

2.3 Data Collection

The process of data collection took 9 months (from the 10th of September 2015 until the 2nd of June 2016). The primary methods of data collection were multiple semi-structured individual interviews, supported by observations plus field notes to assist in developing more detailed accounts of the phenomenon (Smith et al., 2009).

In accordance with IPA, its idiographic focus, and the experience from the pilot study (discussed later in section 2.6), all participants were interviewed separately. Only one person with MND requested to have her daughter present during the interviews for emotional support, which was honoured.

2.3.1 Multiple Interviews

According to Smith et al. (2009) the best way to elicit people's perceptions is through face-to-face, one-to-one interviews. In addition, data collected during interviews can be immediately clarified and expanded upon (Penner & McClement, 2008; Speziale & Carpenter, 2007). In order to conduct a good quality IPA study, detailed, in-depth, and rich information must be obtained, therefore good interviewing skills are necessary (Smith 2011). Although one-off interviews seem to be the most common method of data collection in IPA (Smith et al., 2009), the use of multiple interviews has also been described. Flowers (2008) stated that multiple interviews can provide richer data (due to the established rapport) as long at the gap between them doesn't impact on the participant's ability to recall the previous interview. A decision to carry out multiple interviews as the primary method of data collection in this study was taken for a number of reasons. Firstly, the researcher had concerns regarding the medical status of the people with MND, for example, in relation to their respiratory status. It was also anticipated that dysarthria and the physical effort related to accessing a communication device could hinder the collection of rich descriptions during one interview alone. In addition, multiple contacts with the participants were considered likely to improve rapport with them which could contribute to obtaining more detailed information (Adler & Adler, 2002; Ely, Anzul, Friedman, Garner, & Steinmetz, 1991; Flowers, 2008; Seidman, 1991). Moreover, a multiple interview design allows for the clarification of the information reported during the previous interview (Knox & Burkard, 2009), thus providing an opportunity to validate the findings. In addition, having prolonged contact with the researcher provides the participants with an opportunity to reflect on their own experiences, which may potentially increase the depth of the data collected (May, 1991).

2.3.1. a) Indirect Interviews

Initially at least two (maximum of four) face-to-face interviews were planned with each participant. However, a decision was made to adapt the multiple interview design in response to the particular needs of the population under study and indirect interviews were added to supplement the face-to-face interviews.

Over half of the caregivers (6/10) continued to work in addition to their caring duties. Therefore, it was very challenging for them to find the time to schedule multiple interviews and organise care for the person with MND while they participated in the interviews. In response to their busy lifestyles (and guided by the findings of the pilot study further presented in the section 2.6) the option of scheduling one longer face-to-face interview was offered. However, the caregivers were required to consent to being contacted indirectly by the researcher should any further clarification or additional information be required. Half of the group of the caregivers preferred to participate in one longer interview, whereas the other half took part in two face – to – face interviews.

Interviewing people with MND via indirect means (such as emails) has previously been documented (O'Brien, Whitehead, Jack, & Mitchell, 2011) and this option was also applied in the current study. The decision to include indirect interviews was made during the data collection from the first participant (Joseph[11]). Joseph was observed to have great difficulty accessing his ACC device. He appeared to significantly tire after 35 minutes of his initial interview (onset of hand tremor, increased drooling, and faster respiratory rate), therefore, the meeting was concluded earlier than intended. The review of the data collected from Joseph revealed that he

[11] All names were assigned a pseudonym

was able to answer two questions in 35 minutes (generating 389 words in total). Although Joseph was keen on using technology he clearly required extra time and effort to do so. The researcher became concerned that the effort required to write the answers within the specified time frame could potentially have a negative impact on Joseph's health. In addition, the continuation of multiple face-to-face interviews was unlikely to provide rich descriptions of his experiences due to physical difficulties with accessing his communication device. As Joseph was skilled in using emails he was offered the possibility of conducting interviews via email in addition to face-to-face meetings. This modification was successful as it allowed Joseph to provide descriptions of his experiences when it was most convenient for him. Although Joseph participated in two face-to-face interviews in total, the most detailed information about his life was obtained from his emails. Subsequently, two other people with MND (Bob and Terence) supplemented their face-to-face interviews with interviews via email.

Hand-written interviews were also carried out if participants with MND were unable to use emails. Following the initial interview with Sally and Ted, a notebook was posted to them with follow-up questions. The participants were requested to write their answers in the notebook within 2-3 weeks if possible. A stamped addressed envelope was provided, however the researcher also offered to collect the notebooks in person if this was more convenient. Ted posted back the answers within 10 days and Sally misplaced the notebook at home (this is later discussed in the section 2.3.1c).

Three other participants (Terence, Fiona, and Carmel) spontaneously provided the researcher with information / their reflections written in-between the face-to face interviews, which were included as part of mega-interview for further stages of data analysis (more about mega-interview approach is explained in the section 2.3.1.c below).

Ultimately, all participants were interviewed face-to-face at least once. The total number of interviews is presented in table 2.1.

2.3.1. b) Duration of Interviews

In accordance with the project's ethical guidelines the duration of each interview was monitored and the researcher signaled the end of the interview after 45 minutes (for a person with MND) and 60 minutes (for a caregiver). However, this duration was adjusted in response to participant's individual needs. Shorter interviews were offered if there were concerns with regard to participant's safety (such as signs of fatigue) or in the event of unexpected visitors. Eight participants (three people with MND and five caregivers) requested that their interview continue for a longer time. A summary of the duration of interviews is presented in table 2.1.

2.3.1. c) Gap between Interviews

Although the time gap between the interviews was not precisely specified at the commencement of this study, the aim was to carry out the follow-up interviews within two weeks of the initial one (four weeks at the latest). This gap was deemed reasonable to ensure that participants could still recall their reports from the previous interview. However, the timing of data collection was largely influenced by the participants. Firstly, three participants spontaneously contacted the researcher following the initial interview to provide written accounts pertaining to their experiences of dysphagia. Secondly, seven interviews were postponed due to illness or participants going away on holiday. In the end, the majority of interviews (80%) were conducted within 2-4 weeks and 20% were conducted within two months. These

were all included in the final analysis as findings from the previous interviews were deemed valid by the participants enabling the researcher to implement a mega-interview approach (Flowers, 2008), where data from all interviews is collated in one document and analysed together.

One interview was excluded from the analysis as it was carried out five months post the initial one due to a misunderstanding. The participant in question (Sally) had misplaced her notebook with the written answers and therefore had not returned it. Due to the lack of contact from this participant it was assumed that Sally was unable or unwilling to continue her involvement in the study. It was not deemed appropriate for the researcher to make any direct inquiries in relation to the return of the notebook as the researcher was aware of some significant changes in the participant's circumstances. Almost five months later, the researcher was contacted by Sally's caregiver, who reported that the notebook had been misplaced at home but that Sally would still like to complete her participation in this research. Subsequently, a second interview was carried out in Sally's home, however, it's idiographic analysis revealed significant differences in her perceptions and experiences in comparison to the initial interview. This time gap of five months was considered too significant to allow for the use of a mega-interview approach (Flowers 2008), and following consultation with the researcher's Supervisory Team a decision was made to exclude the second interview from the cross-case analysis.

During the first interviews an interview guide[12] (separate one for a person with MND and for a caregiver) was used in a flexible manner in order to remain open to

[12] Appendix 6a & b

conversation with the participants on topics which were important to them. It was also referred to if necessary in order to re-direct the conversation to dysphagia.

The total number of face-to-face interviews (32) and the total number of indirect interviews or follow up contacts (36) were similar which indicated to the researcher that they were an equally important means of data collection. Although the mean duration of the interviews was comparable in both groups it needs to be acknowledged that the caregivers generated richer descriptions of their experiences in comparison to the people with MND and therefore did not require as many indirect follow up contacts.

Table 2.1 Multiple Interviews: A Summary

Sample	Total number of face-to-face interviews	Duration (range)	Duration (mean)	Duration (total time)	Number and type of indirect methods of data collection
PwMND (n=10)	17	31 mins – 61 mins	44 mins	11 hrs 49 mins	22 Emails 5 Other written accounts
Caregivers (n=10)	15	22 mins* – 71 mins	48 mins	11 hrs 56 mins	3 phone calls 2 emails 4 text messages

**The 22 minutes interview with a caregiver was terminated due to an unexpected visitor.*

2.3.2 Observations

The majority of interviews were supported by observations of a meal or meal preparation. A number of interviews (n = 3) were carried out outside of the participants' home environment, which negated the opportunity to make dysphagia - related observations. Although the main aim of including observations was to focus the interview on experiences related to dysphagia, it was also anticipated that they could increase the depth of data thus helping to capture and explore the participants' world in more detail. Natural opportunities for observation were sought, for example, while having a snack or a cup of tea with the participants. Also, when participants had difficulties communicating specific aspects of their experiences using words they were encouraged by the researcher to demonstrate their actions. For instance, if it was hard for participants to describe how non - oral feeding was managed the researcher proposed that they showed her the feeding pump. This was still in line with hermeneutics, which emphasises the role of language, as the participants were encouraged to describe their actions as they were demonstrating them. These moments appeared to be very important as they provided the opportunity to gain a more in-depth understanding of the participants' world.

Notes were taken during the interviews and observations to record specific details, such as non-verbal communication or specific actions related to food preparation. In addition, the researcher audio recorded any observations immediately following the interview. These recordings were subsequently transcribed and used during the process of data analysis.

2.4 Research Ethics and Consent

The original ethics approval was obtained from the Clinical Research Ethics Committee of the Cork Teaching Hospitals (26th of April 2013). However, this approval was amended twice: following completion of the pilot study to reflect the modifications applied to the research design (such as changed cognitive testing tools), and following recruitment of the first caregiver (as the data collected revealed potential risks for a person with MND and therefore the consent was revised to include conditions under which confidentiality may be broken).

This research was carried out in accordance with four fundamental ethical principles of justice, benevolence, non-malevolence and autonomy (Butts & Rich, 2013). The process of obtaining informed consent from the participants focused on providing detailed information about the study. This included: (1) the purpose of the study, (2) its expected duration, (3) the data collection process, (4) the right to decline or withdraw, (5) potential risks and benefits, (6) confidentiality, (7) the dissemination of findings, and finally (8) who to contact with further questions or complaints. The researcher ensured all potential participants were familiar with the content of the written consent and had sufficient time to process information and ask questions. All participants were able to sign a witnessed consent form.

In the early stages of the development of this project the researcher acknowledged that speaking about the experiences of living with MND and dysphagia may evoke feelings and emotions in both groups. A decision was made that in the event of a participant becoming significantly upset, a termination of interview would be offered. Participants registered with the IMNDA were reminded about their entitlement to avail of free counselling sessions. Non-registered participants were to

be requested to consider contacting their GP or a counsellor. However, during the course of this research none of the participants wished to terminate an interview, although many experienced emotional distress. It appeared sufficient to provide the participants with a short break until they volunteered to continue with the interview.

The well-being of the people with MND was a particular concern due to their medical diagnosis, therefore, the researcher was focused on observing for any potential changes in their status (for example, respiratory changes) or any indicators of fatigue (sleepiness, inattentiveness, deterioration of voice and speech). Although none of the participants with MND became unwell during the interviews, signs of fatigue were often observed. Any observation or concern in relation to the level of fatigue of the participants was always communicated to them and a break or termination of the interview offered.

A debriefing process followed each interview whereby the researcher thanked the participants and assured them that their participation would be kept confidential and their responses would be anonymised. Data collected during this study was treated with confidentiality during all stages of the project. Only the researcher and the Supervisory Team had access to the original data. All participants were assigned a pseudonym to ensure they could not be identified from any publications or reports arising from this study. All other specific information which could identify the person (for example, the names of their children, the place of work etc.) were also anonymised.

2.5 Data Analysis

In line with the study design, an in-depth analysis was commenced once all data was collected from a participant. Interview transcripts from each person were amalgamated into one Microsoft Word document while maintaining the chronological order of the data collection (for example, a transcript from the initial face-to-face interview was followed by copies of emails and another transcript from a subsequent face-to-face interview). The final document was treated as one comprehensive text or "mega-interview" (Flowers, 2008) during the data analysis process. Data analysis was consistent with the IPA approach and followed the steps as proposed by Smith and colleagues (Smith et al., 2009), which are now presented.

Step 1. The initial step of data analysis was considered as my great attempt to enter each participant's world. I listened to the recordings from every interview at least five times until a sense of immersion in each participant's personal experience was obtained. I read the notes taken during and after interviews prior to re-listening to any important parts of the interview (for example, long descriptive pieces which I had attempted to make sense of, or the interviews of those with dysarthric speech which were particularly difficult to understand). At times, a slow-speed mode was used to listen to the recordings to help with recognizing some words. A professional transcription service was used for all audio-recorded interviews, with a confidentiality contract in place. All transcripts were double checked against the original recordings. This revealed a number of errors in them, particularly in relation to medical terminology used by the participants.

Step 2. In preparation for step two (initial noting) each transcript was changed to landscape mode leaving wide margins on both sides to allow space for notes and

emergent themes and inserted line numbers. The exploratory comments were written in the right margin and a colour coding system was used to highlight particular areas of interest, such as parts of interview especially important for the participants, and parts directly relevant to dysphagia. The parts highlighted led to the development of both descriptive and conceptual comments. Moreover, specifics linguistic features (for example metaphors) were colour coded too, and they were used to create linguistic comments. The colour coding system was particularly useful during cross-case analysis as it allowed to, for example, quickly identify parts directly relevant to dysphagia within each transcript. In order to distinguish the type of comments within the transcript I followed another visual system (as suggested by Smith et.al 2009): descriptive comments were written in normal font, linguistic in italic, and conceptual were underlined.

I combined working on the computer with working manually. For me, using pen and paper creates almost an intimate relationship with participants' worlds and enables a deeper engagement with the data.

Step 2 was a particularly important phase of the data analysis as it led to discovering new aspects of participants' experiences thus creating a feeling of "getting closer" to understanding their world. At this stage, I continued to listen to the original recordings to maintain a loose connection with participants. Step 2 was treated as a transition to working primarily with the text at the later stages of data analysis.

Step 3. This step involved the development of emerging themes, where I simultaneously attempted to "reduce the volume of detail whilst maintaining the complexity" (Smith et al., 2009 pg. 91). This was a process of working primarily with the notes and comments created during the previous steps of data analysis. This step

brought specific difficulties as initially I felt uncomfortable about having to break the narrative flow of the interview. However, when the idiographic analysis of the first two participants was completed, I was able to see how all the parts came together again in the final write up. Although my interest was in seeking participants' perceptions and experiences of dysphagia I remained open to considering what was emerging from the data and what appeared to be of particular importance to the participants. My hypothesis was that understanding participants' lives in general would bring me closer to their perceptions of dysphagia. The final list of emergent themes consisted of both participant's original words and thoughts, and my own interpretations. At the end of this process, a table was created with all emergent themes in one column and supporting quotations in the other, which provided an opportunity to review the themes again to ensure they appropriately reflected what was emerging from the data. This table was useful when writing the final analysis: as the emergent themes were grouped into the higher themes (subordinate and superordinate) they were copied and pasted into the final data analysis table together with the corresponding quotations.

Step 4. This step involved searching for connections between themes through the implementation of the techniques of abstraction, subsumption, polarization, contextualization, numeration, and function (Smith et al., 2009). Each of these techniques is now briefly explained.

According to Smith et al., (2009) abstraction relates to identifying patterns ("putting like with like") between emergent themes and "developing a new name for the cluster" (p.96). In the process of abstraction, for example, the emergent themes "Modifying consistency of food", "Substituting risky foods with softer textures", and "Monitoring the pace of eating and drinking" became one superordinate theme: "Strategies to manage dysphagia".

Subsumption is carried out in the same way as abstraction; however, an emergent theme itself is assigned a superordinate status.

Polarization includes an examination of oppositional relationships between emergent themes. For example, an emergent theme "Quality of life considerations" was contrasted with "Professional recommendations", where the first theme related to participants' desire to continue a regular oral diet despite dysphagia for perceived quality of life reasons, and the second included professional recommendations to modify the textures of food due to concerns about the medical safety of the person with dysphagia.

Contextualization considers the connections between emergent themes from a contextual or narrative perspective to create a "more local understanding" (Smith et al., 2009, p. 98). For example, the themes related to Terence's attempts to follow a recommendation from his SLT to thicken his liquids, were dispersed across his transcript. Through the process of contextualization these themes were collated together, which allowed an understanding of Terence's approach to this professional recommendation (see Figure 4.2).

Numeration applies to considering the frequency of the appearance of emergent themes, as presented in tables 4.1. and 5.1.

Lastly, function relates to an investigation of specific functions of emergent themes within the transcripts such as, for example, particular meanings or language. In Joseph's interviews, for example, consistent writing of the word "mnd" was interpreted as him expressing his attitude towards this disease (section 4.1.2).

The first part of step 4 included printing a list of the emergent themes and cutting them so each theme was on a separate piece of paper. A large space was used

to explore the spatial representation of how the themes fitted together, for example, the related themes (representing similar understanding) were grouped together and a new name was developed for the cluster (abstraction). This stage of the analysis was particularly creative as I was aware that there were multiple possibilities in terms of organizing the themes. To document my own decisions and justifications for those decisions a reflective diary was kept with comments on my analytical work. This diary, along with the notes from observations conducted during interviews, was often reviewed during step 3 and 4 of data analysis. Although the primary focus on the research questions was maintained throughout the analytical process, I allowed the data to lead my interpretations. This resulted in the inclusion of the themes important to the participants, even if they appeared not to be related to dysphagia. At the end of this step a final table was created for each participant[13] representing higher order themes and including my comments, explanations, and interpretations. Subsequently, a mind map[14] was developed for each participant, to provide a simplified visual representation of the order themes. The mind maps proved to be particularly useful for noticing general differences and similarities between participants during the initial stages of the cross-case analysis.

Step 4 concluded the idiographic analysis for each participant.

Step 5. This step involved the completion of steps 1 - 4 for another participant following the chronological order of interviews. I focused on the analysis of one particular participant until this was completed before moving to another person.

Step 6. Looking for patterns across cases formed the final step of the data analysis. At this point the idiographic analysis had been concluded for each participant

[13] Example in Appendix 7a & 7b
[14] Example in Appendix 8a & 8b

and the data was subsequently divided into two groups: people with MND and the caregivers. The group of people with MND was analysed first. Each mind map was laid out on a large surface to identify connections and relationships, but also differences between cases, aiming to draft recurrent themes and develop a deeper level of interpretations. In order to ensure adequate interpretation of the mind maps, however, I always referred to the final analysis table for each participant (example in Appendix 7). In addition, I repeatedly referred to individual transcripts to deepen my understanding of each individual participant, and to stay close to their original words. Both similarities and divergences between the members of each group were taken into consideration. Attention was paid to the words used to describe specific experiences, and in particular to any metaphors used. The wider context of each participant's life was examined to search for connections between their experiences and personal circumstances. I reflected on my previous knowledge and presumptions, both personal and professional, to recognize them and their potential influence on the analytical process.

The establishment of the higher order themes took over two months for each group. This process was particularly complex, as while making a conclusion relevant to the whole group I did not want to lose individual stories. A decision was made that in order to define themes as being recurrent they had to be present in at least half of the group. However, in accordance with the idiographic approach, unique themes were also reported on. The final result of the cross-case data analysis for each group was a table representing the hierarchy of themes followed up by an in-depth description of the results for each group (Chapters 4 and 5).

2.6 Pilot Study

In order to test the research design and pilot the interview technique, a pilot study was completed with one person with MND (Nora) and her daughter (Natalie) in May-June 2014. The participants were enrolled from the IMNDA and three meetings were conducted in their home. The aim of the first meeting was to explain the project, obtain consent, and assess participants' eligibility for this research. Subsequently, two more meetings were carried out to pilot the interview technique (a joint interview with Natalie and Nora followed by an individual interview with Nora only). The data was analysed maintaining the idiographical focus and following the steps as suggested by Smith et al. (Smith et al., 2009). The data obtained from this pilot study was excluded from further cross-case analysis, due to modifications in the research design which were applied following completion of this pilot study.

A number of lessons were gleaned from the pilot study in relation to multiple interview design, joint interviewing, establishing ethical boundaries, and techniques to help the participants to focus their attention on dysphagia. I observed that the data obtained from Natalie (caregiver) was significantly richer in comparison to Nora's (person with MND), who had a communication disorder (dysarthria). This observation led me to anticipate that participants in each group were likely to provide a very different body of data within the same timeframe. Moreover, Natalie appeared to be under significant time pressure and in fact she was unable to participate in the second interview, as she did not have anyone to stay with Nora for that period of time. Therefore, I acknowledged that for some caregivers meeting me in person more than once may cause significant strain.

It also became apparent that joint interviews created a degree of discomfort for me, particularly when the participants voiced different opinions. I also suspected that the presence of Natalie influenced Nora's communicative efforts (she was observed to let her daughter talk for her), potentially impacting on my understanding of Nora's own experiences. These observations encouraged me to request separate individual interviews for future participants.

During the pilot interviews I was exposed to the emotional side of living with MND. Although this was anticipated in advance, witnessing participants' emotions directly was difficult. This provoked further reflection and consideration regarding how to sensitively approach and manage participants' emotions during subsequent interviews.

An ethical issue arose during the pilot study, which related to the establishment of my boundaries as a researcher. Natalie raised a number of clinical queries looking for my clinical support. This created an ethical dilemma for me, as due to my professional experience and knowledge in the field of MND I could potentially have responded to Natalie's queries. This was the moment when I had to change my stance from being a therapist to being a researcher. Further reflection on this experience and discussions carried out with my Supervisory Team ensured my ethical boundaries as a researcher were maintained throughout this research project.

For me, one of the most unexpected observations during the pilot study was that despite being diagnosed with dysphagia and taking a modified diet, Nora appeared to have difficulties reporting her experiences of dysphagia. Natalie also largely focused on aspects of living with MND other than dysphagia. I came to recognize my own presupposition that dysphagia in MND would be one of the primary concerns for

participants. I noted that the pilot interview became naturally focused on dysphagia when Natalie offered Nora something to eat and drink. While she was making tea for Nora, Natalie began describing her experiences of thickening Nora's fluids and while she was cutting a cake she recalled a recent choking episode. Having something to eat and drink appeared to also encourage Nora to express her experiences of dysphagia. Although she denied having dysphagia a few times during the interview, she began to describe her swallowing difficulties while eating the cake.

As I aimed to understand Nora's lived experiences holistically I was interested to discover issues that mattered to her. Further analysis of the interview data revealed that Nora's main concern was her fear of losing her voice. Experiencing dysphonia was more important than experiencing dysphagia for her. Therefore Nora re-directed her answers to some of my dysphagia - related questions and spoke about her difficulties with voice projection instead. She benefited from a cue (being offered some food and drink) to help her focus on the experiences of dysphagia. This situation encouraged me to include observations of eating or / and drinking in the study design, primarily to help the participants recall their experiences of dysphagia, but ultimately to improve the depth and the quality of the data collected.

Following the pilot study the interview guide was finalised applying certain modifications to its pre-pilot format. I ensured no leading questions were included and questions directly concerned with dysphagia were not asked at the early stage of the interview. Instead, I planned to spend the time attempting to get to know each participant by inquiring about their lives in general. The pilot study led to the awareness that during data collection I may have to look for ways of uncovering the experiences pertaining to dysphagia, as the participants may not be focused on them at all. This understanding led to me maintaining an open mind during the interviews

and waiting for appropriate moments to direct the conversation towards dysphagia, if necessary. The pilot study highlighted that each participant is unique, and that the interview schedule therefore needed to be used in a flexible manner.

The completion and analysis of the pilot study confirmed IPA as a suitable methodology committed to understanding the participants' perspective of an important life event (MND and dysphagia) from the third-person (researcher's) position.

2.7 Assessing Quality

Although quantitative and qualitative research employ very different conceptual frameworks, it is not uncommon for qualitative research to be judged as valid erroneously based on criteria specifically developed for quantitative research (Hollway, 2007; Yardley, 2011). To enhance and assess the validity of this project, and in accordance with Smith et al. (2009) recommendations, Yardley's (2000) criteria were implemented, which were: (1) Sensitivity to context, (2) Rigour and Commitment, (3) Transparency and coherence, and (4) Impact and importance.

2.7.1 Sensitivity to Context

Good qualitative research must represent its context sensitively including, for example, sensitivity to socio-cultural aspects of the study, relevant ethical issues, the existing literature on the topic, and the data obtained from the participants. In IPA the researcher should demonstrate their sensitivity to context from the very initial stages. Smith and colleagues (2009) stated that the choice of IPA itself can be sensitive to context "through close engagement with the idiographic and the particular" (p. 180).

In IPA, sensitivity to context should continue through all stages of the research: data collection, analysis and write up.

My sensitivity to context manifested through being observant, flexible, and responsive to the specific needs of the populations under study. The goal was to ensure that this research was as convenient as possible for the participants and that it did not create an additional burden for them.

The sensitivity was also evident in relation to my preparation for the interviews so that I could ensure the time in each interview was optimally spent. I practiced my interviewing skills with people with communication impairments prior to this study, including people post-cerebrovascular accident with dysarthria. In addition, I completed multiple workshops on qualitative interviewing. The pilot study was conducted and reflected upon and included consideration of my interviewing skills. I was also aware that my accent was different to the participants' as English is not my native language. Therefore I engaged in conversations with the participants prior to the interviews to allow them time to familiarize themselves with my accent.

As the study progressed, my sensitivity to context manifested in a number of strategies which were employed to ensure a correct understanding of participants' utterances and to minimize their efforts related to expressive communication. I reassured each person with MND, who communicated via writing, that perfect spelling was not required. If appropriate, I attempted to predict longer words to reduce participants' efforts and always offered help if someone indicated difficulties with typing a specific word. In addition, yes-no questions were included to clarify or expand on participants' reports, although I was aware that this is not typically recommended in qualitative interviewing (Creswell, 2013; Flick, 2014; Brinkmann & Kvale, 2009).

At times, participants' utterances were repeated back to them to ensure I had understood them correctly. Moreover, notes of any non-verbal communication were also taken, such as gestures and facial expressions. These were reviewed along with the interview transcripts during the analysis.

My desire to obtain intelligible information from people with MND was adjusted according to their emotional state. I accepted that some parts of the interviews might be less intelligible. No request for a message to be repeated was made if the effort related to communication appeared significant or if the participant was upset. Instead, an effort was made to review the unintelligible parts at the subsequent contact.

The individual needs of each participant were acknowledged throughout the study. During the data collection an attempt was made to create a relaxed atmosphere and maintain a natural communicative environment. This included, for example, looking at the person with MND rather than a communication device.

During the data analysis I immersed myself in the data paying detailed attention to each participant. My interpretations were grounded in the data and my analysis was carried out carefully. I consulted my Supervisory Team in the event of any uncertainty, and I also consulted with them at every stage of the data analysis.

Finally, the sensitivity to context was maintained while relating the findings to the existing literature. This manifested in ensuring regular reviews of the literature, ensuring up-to-date knowledge through attendance at conferences, and liaison with experts in the field (both nationally and internationally).

2.7.2 Rigour and Commitment

Rigour refers to conducting the study thoroughly and can be shown, for example, by a reasonably homogenous sample selected adequately to match research questions (Yardley, 2000, 2011). While designing this research, I strived to maintain rigour by the careful selection of the sample in order to answer the research questions, and the establishment of comprehensive eligibility criteria. A pilot study was completed to test the research design and changes were implemented appropriately (as reported in section 2.6). Data was recorded objectively (audio recordings) and translated using professional services. All transcripts were double checked against the original recordings by the researcher. Data analysis was systematic and followed the sequence of steps suggested by Smith et al., (2009) for novice researchers. Each theme was supported with quotations to demonstrate how the analysis was grounded in the data.

In any IPA project commitment is expected to be shown by the degree of attentiveness to each participant while collecting and analyzing the data (Yardley, 2000, 2011). My commitment manifested in all stages of this study, from establishing a good level of trust between the participants and I, through making adaptations to the data collection process in order to facilitate the participants, and during data analysis when a focus on every detail was maintained in order to produce careful and well thought through accounts. In addition, I attended various training and workshops to ensure an appropriate level of research related skills. All issues requiring clarification were raised with my Supervisory Team throughout this research in order to agree on the optimal solution.

2.7.3 Transparency and Coherence

Transparency refers to a clear description of all steps undertaken during the study (Yardley, 2000, 2011); therefore a trail of all decisions made during the project was kept. Although the analysis followed the steps suggested by Smith et al. (2009), this was enhanced by an in-depth demonstration of how the higher themes were developed (tables as per Appendix 7a and 7b, mind maps as per Appendix 8a and 8b). Exactly the same process of data analysis was followed for each participant.

Coherence refers to the clarity of the arguments stated throughout the research (Yardley, 2000, 2011). The process of writing was precise and in-depth, with systematic drafting and re-drafting of findings until cohesive and comprehensive analysis was completed. A clear demonstration of how each conclusion was made was included. During the data analysis and writing up of the results my decisions were supported with quotations from the original transcripts.

2.7.4 Impact and Importance

Impact and importance relate to research findings and their influence on the profession, which is understood to be a test of real validity (Yardley, 2000, 2011). The findings of this study aspire to provide a new understanding of dysphagia in MND, as it is perceived and experienced by people with MND and their caregivers. This new knowledge (via the process of dissemination of the findings) could potentially contribute to changes in service provision for the people living with MND (both people diagnosed with MND and their caregivers) and could provide directions for future research. Early on (prior to data collection) I disseminated the necessity to investigate dysphagia in MND beyond the medical perspective. Later, when the idiographic

analysis was completed for some participants, the data was presented at national conferences in the form of case studies. As the research evolved, my dissemination expanded and included international conferences relevant to professionals working with MND and dysphagia. Publications in peer reviewed journals are planned.

2.8 Researcher Positionality

I am a Speech and Language Therapist with a particular interest in neurology and dysphagia. I have been living and working in Ireland since 2003 and in 2005 I achieved a postgraduate qualification in dysphagia. Therefore, before the recruitment of this study commenced I had over 10 years of professional experience in the management of swallowing impairments and an advanced knowledge of the Irish health system. I considered this to be important as during the research I was not learning about the Irish context, but rather relating participants' experiences to a context with which I was familiar.

This study emerged from my clinical interest in investigating dysphagia as it is experienced in a home setting, in particular when dysphagia is accompanied by other concomitant disabilities or disorders. In the course of my previous clinical work managing a person with dysphagia typically meant prioritizing medical risks (such as the occurrence of aspiration, choking, dehydration, or malnutrition) over the quality of life principles of the person with dysphagia, especially when working within limited resources. My experience proved that working under significant time pressure often does not allow for a more holistic approach to patients presenting with dysphagia nor for the provision of adequate support to the caregivers. I aimed to carry out a research study, which could provide new insights into the personal perspectives and

experiences of people living with dysphagia and I aspired to translate the findings into clinical practice.

2.8.1 Potential Issues that Concerned Me

The participants in this study were aware of my clinical background as this was stated in the invitation form. This could have potentially impacted on the data collection process. For instance, during the pilot study I was asked to offer clinical support. I later wondered if the desire to obtain my clinical input could have influenced the participants' decision to take part in this research. However, I believe that my professional background did not significantly influence the data collection process as the participants did not associate me with any specific organization or institution. During the data analysis it emerged that had I been representing a particular health care facility the participants may not have openly disclosed their opinions, in particular their critique related to the model of service delivery.

I am not Irish and this appeared to be an important consideration for the participants, as they frequently commented on it. At the initial stages of this research I anticipated that coming from Poland may be a barrier. I was worried that perhaps the participants would not engage with me in the same way they would had I been born locally. To my surprise I realised that my origin helped participants to disclose information, as I was perceived as an outsider, someone who lacked local connections.

Despite my professional background in SLT and experience working with adults with various speech impairments, this was my first experience of conducting in-depth interviews with 10 people presenting with dysarthria or anarthria. I was aware that some people with bulbar onset of MND experience dysphagia soon after

diagnosis. However, they may also present with a significant degree of dysarthria meaning that their speech would not be adequately intelligible to allow for their participation in the interview. I suspected that some participants with dysarthria may not have an effective communication system in place. I intended to use my professional knowledge of AAC to help the participants, in conjunction with their SLT, to source a suitable device via a loan system from the IMNDA. I aspired to leave the device with the person with MND following their participation in the project. However, I anticipated potential difficulties related to the practical side of introducing a new communication device, particularly considering my role was not to be a SLT, but a researcher. Although I aimed to liaise with the relevant SLTs I was not able to predict in advance if this collaboration would be fruitful. I was relieved to learn that all non-verbal participants already had AAC systems in place.

There were other issues that I anticipated prior to the commencement of this project. The geographical areas chosen for this study involved many rural locations, where unfortunately people with MND may have limited access to health services. I identified this as a potential barrier to recruitment. In the event of difficulties recruiting participants within the selected geographical area, I intended to expand the study nationally. Although this expansion was not required, the recruitment phase took approximately two months longer than anticipated.

2.8.2 My Presuppositions

At this stage I would like to admit my two biggest presuppositions, which I re-evaluated during this research. Firstly, prior to this study I was convinced that having dysphagia must be significantly upsetting therefore it would form a central concern

for any person affected. I almost expected my participants to produce rich descriptions of their dysphagia related difficulties without requiring any support from me to do so. My presupposition was quickly challenged during the pilot study (section 2.6), when the participants chose to speak about other aspects of living with MND and dysphagia did not appear to be very concerning to them.

Another prejudgment was that I expected that the majority of people with MND and dysphagia would require a caregiver to manage their swallowing impairment. I did not appreciate that many people with MND maintain their independence despite experiencing multiple disabilities and significant dysphagia. I was concerned about identifying suitable people with MND and I expected that finding the caregivers would not be an issue. This research found the opposite to be the case, as the recruitment of caregivers was longer and more challenging. I underestimated not only the independence of people with MND in relation to dysphagia-management, but also the enormous time pressure the caregivers lived under.

2.8.3 The Journey through this Research

I strongly identify with England (1994), who stated that "research is a *process*, not just a product" (p.82). It is a process, which initiates long before the official commencement of the study, and lasts beyond the final write-up. I perceived my engagement in this research as a complex journey, that despite my in-depth preparation I met many challenges and surprises, such as when my assumptions were proved to be incorrect. As a professional I learnt that it is only through careful observation and analysis that the complexity of dysphagia in MND can be understood. This understanding required a significant amount of time, in order to closely approach the

experience of each individual and then create shared characteristics of each group, while still maintaining the focus on individuals, who were unique. This understanding of dysphagia in MND could not have been achieved in the course of clinical work.

In a wider context, this research influenced me as a person. I felt privileged to be in the homes of my participants, honored to be allowed some insight into their lives and experiences. I considered the data collected from the participants to be very special. I strived to ensure that I reflected everything that seemed important to them, especially as many passed away shortly following their participation. Although this research originated from my own clinical interest and I aimed to fulfil the gap in our professional knowledge, my biggest concern was to ensure that the voices of the participants would be adequately disseminated.

At the early stages, when I had decided to conduct research but was still unsure about the topic, I was advised to choose something I love. In a way I did not listen. I chose something I absolutely hate: a disease which makes people unable to "scratch an infuriating itch, or wipe a sorrowful tear from [their] eye" (Orpen, 2017). MND is horrendous, unfair, ugly, and for many an unexpected disease, which to date is unstoppable. How people living with MND cope and maintain a positive attitude was an amazing discovery. That they devoted their precious time to meeting me was an honour. How this experience enriched me as a person is beyond words.

During this research each time when I wrote: "a person with MND" and labeled a person with a medical diagnosis I felt uncomfortable. This had never concerned me in my clinical practice. However, each participant with MND I met was in fact A PERSON with mnd. This disease did not define people and their existence.

The term “caregiver” is also an understatement. These PEOPLE were spouses or children of SOMEONE with mnd, which meant they were not born as caregivers. This duty appeared suddenly, influencing their emotional, social, and professional lives. When the caregivers reported that their life was on hold I felt their hope to return to their previous life. However, I did not believe that this would ever be possible. Being a caregiver, sharing the experience of this frightening and merciless disease, transforms human identity and re-evaluates priorities. In my opinion, life after MND for the caregivers would never the same as it was prior to this disease.

Finally, I will be forever grateful and privileged for this experience. I consider having the opportunity to talk to people facing terminal illness and attempting to understand their stories as something very unique and special. This truly was an enriched journey from both a personal and professional perspective.

CHAPTER 3: PARTICIPANTS

Introduction

This chapter aims to introduce the participants of this study by presenting their demographic and contextual characteristics (short stories) to provide more insight into their lives and personal circumstances. Each participant was assigned a pseudonym. The first section of this chapter presents information pertaining to people with MND and dysphagia and the second section relates to the caregivers of people with MND and dysphagia.

3.1 The People with MND

A sample of five men and five women were included in the study. Table 3.1 presents the demographic background of the participants (in alphabetical order) at the time of entry to the study. Given the progressive nature of MND some changes in these demographics were expected in the course of this research. This was predominantly evident in relation to participants' communication abilities. Five participants (Joseph, Sally, Sheila, Ted, and Terence) were completely non-verbal from the outset and communicated through various alternative means (which corresponded to levels 1 and 2 of the ALS Severity Scale: Speech; Yorkston et al., 1993). Two participants (Carmel and Mike) presented with very limited speech with greatly reduced intelligibility (levels 3 and 4) and also required alternative communication methods in order to participate in the interviews. Three participants (Bob, Eileen and Fiona) were verbal on entry to this study (level 6), however, two (Bob and Fiona) became non-verbal by the second interview and needed to use writing

to complete their participation in this research. In summary, as the study progressed nine out of ten participants with MND relied on alternative systems to communicate.

The Eurostat definitions have been used to classify participants' living areas into rural, urban, and suburban (Eurostat, 2017). Over half of the people with MND in this study lived in rural areas of Ireland. The mean age was 67.2 (ranging from 55-80 years). The majority of participants were married. The mean time since diagnosis of MND was 20 months (ranging from 2 months - 6yrs), and the majority (9/10) presented with bulbar onset of MND.

Table 3.1 Demographic Characteristics of People with MND (PwMND)

PwMND (pseudonym)	Age	Employment status	Living area	Marital status	Time since diagnosis of MND	MND onset	ALS Health Score	ALS Severity Scale: Speech	Cognitive status
Bob	64	Retired teacher	Rural	Married	2+ months	Bulbar	2 – moderate	6	Normal
Carmel	58	Employed chef	Urban	Single	6 months	Bulbar	2 – moderate	4	Normal
Eileen	73	Retired nurse	Rural	Married	8 months	Bulbar	2 – moderate	6	Normal
Fiona	69	Retired teacher	Suburban	Married	6 months	Bulbar	2 – moderate	6	Normal
Joseph	65	Retired teacher	Suburban	Married	6 years	Spinal	4 – terminal	1	Normal
Mike	55	Self-employed farmer	Rural	Married	1.5 years	Bulbar	2 – moderate	3	Normal
Sheila	65	Retired housewife	Rural	Married	3 years	Bulbar	4 – terminal	2	Normal
Sally	80	Retired housewife	Rural	Married	3 months	Bulbar	2 – moderate	2	Normal
Ted	75	Retired teacher	Suburban	Widowed	6 months	Bulbar	3 – severe	1	Mildly impaired
Terence	68	Retired farmer	Rural	Married	1 year, 9 months	Bulbar	3 – severe	2	Normal

3.1.1 Introducing the People with MND: Short Stories

Bob

Bob was seen for one face-to-face interview in a palliative care centre followed by interviews via email. Bob lived at home with his wife and had recently retired. They did not have any children. Bob reported that MND predominantly affected his speech and respiration. Although Bob was still able to walk independently, he admitted having to be extra careful as his feet were becoming unsteady. Bob's diagnosis was delayed and it was officially confirmed only two months prior to his involvement in this study, thereby giving him the shortest experience of living with MND of all the participants.

During the first interview Bob appeared to have difficulty describing his experiences and he tended to provide limited answers. The intelligibility of his speech was affected by decreased respiratory support and dysarthria, and it was observed to further deteriorate as the interview progressed. Therefore Bob was offered the option of continuing his participation via email, which he accepted.

During his participation in this study, Bob did not appear to have adjusted to his diagnosis, he seemed confused when talking about MND and appeared not to understand the nature of the disease in any great detail. Bob reported that he did not have any knowledge of MND and felt overwhelmed by what was happening to his body and to his life in general. MND had put an end to Bob's post-retirement plans including his wish to travel and move abroad.

Bob presented with mild dysphagia and he was not concerned about his ability to swallow. Instead, his main concern was related to his deteriorating respiratory functions. Bob avoided eating-out as he experienced increased respiratory difficulties in public places, especially if they were crowded and small.

Bob passed away two months after his last interview.

Carmel

Carmel was interviewed twice face-to-face in her home where she lived with other tenants. She was single and did not have any children. She reported that she enjoyed spending time outside and going for walks with her friends. Carmel presented with significant dysarthria leading to poor speech intelligibility. She therefore required an augmentative communication system and used handwriting while she waited for a computerised AAC device. Carmel's limbs were unaffected by the disease and she was fully independent and mobile.

Carmel was worried about her future with MND especially as she did not have a family of her own. She knew she would not be able to stay in her house when her condition deteriorated. At the beginning of the first interview Carmel became very emotional when thinking about her future, but she declined the offer to terminate the meeting. Later, she reported that she still maintained hope that she would be able to return to work. She had many friends in her work environment and continued to meet them regularly. However, MND was reported to isolate Carmel from the other tenants in the house, predominantly because of anarthria.

Carmel's swallowing was still relatively good; however, she opted for a gastrostomy shortly after her diagnosis. She did not regret this decision as she reported

that supplementing her oral diet with the gastrostomy feeds reduced the need to cook meals and allowed her to enjoy time outside for longer. Carmel believed that non - oral feeding would preserve her independence for longer by ensuring she maintained the correct level of nutrients.

Eileen

Eileen was seen for two face-to-face interviews in her home where she lived with her husband (her children are now adults). One of her grandsons visited her frequently and she reported having an especially strong bond with him. Eileen was very animated during interviews and often used her whole body to demonstrate her experiences, for example, she acted out her recent choking episode by reconstructing where and how it happened. Perhaps Eileen was attempting to compensate for her dysarthric speech and unsure as to whether her experience was understood, but also it is possible that those events were associated with significant emotions, which sometimes cannot be described in words.

Eileen was fully mobile and not only able to look after herself, but also her husband, their house, and their garden. Eileen appeared to be determined to stay positive. She reported developing her own strategies to help her overcome various MND related problems and she admitted that some she applied almost subconsciously.

Eileen had been experiencing some choking episodes, which made her very nervous. She described "feeling uneasy all the time". She made many adaptations in her lifestyle in an attempt to minimise the likelihood of choking. She stated that MND took away her confidence. This, however, was not observed externally, as Eileen came across as very content, organised, and confident.

Fiona

Fiona was seen for two face-to-face interviews in her home where she lived with her husband. Her children were adults and she was involved in looking after her grandchildren, which was a source of great joy for her. Fiona appeared to be very proud and strong and focused on her abilities rather than her difficulties. She aimed to enjoy life, travel, and participate in hobbies, although she slowly had to limit some of her activities. Fiona's limbs were unaffected by MND, however, her dysarthria and dysphagia were progressing. During the first interview Fiona was able to speak relatively well but by the second interview her ability to talk had deteriorated significantly and she decided to participate through handwriting to ensure her intelligibility. Fiona appeared to be very aware of the changes happening in her body due to MND and she was able to precisely describe her new symptoms. She was calm and focused on finding solutions to help her manage the disease. Her approach to life seemed very pragmatic and realistic.

Fiona reported some difficulties in managing solid food, especially hard and slippery textures. She knew about the associated risk of choking and applied specific techniques to increase her safety during meals. She decided to eliminate dangerous textures from her diet to maximise her safety. This did not affect her eating-out routines.

Joseph

Joseph participated in two face-to face interviews in his home supplemented by interviews via email. Joseph lived at home with his wife and an adult daughter. He had spinal onset MND (diagnosed six years prior) leading to minimal usage of his

limbs. He used a wheelchair and required full care. He was non-verbal due to anarthria and communicated by typing on a tablet using his index finger. Despite his physical limitations Joseph maintained his role in the family as it had been pre-MND. He was not only engaged in many cognitive tasks (for example banking), but also attempted to participate in more physical duties (making dinner). This demanded adaptations and support from his carers such as leaving food and utensils accessible.

Joseph was always well prepared for the interviews and pre-recorded some of his messages (such as greetings) on his communication device. He appeared to be very knowledgeable regarding MND. His writing skills were exceptionally good and he was able to provide rich descriptions of his experiences. In addition, Joseph appeared to enjoy sharing his stories.

Joseph presented with dysphagia, however, he was able to eat a modified diet quite well. He enjoyed eating and was focussed on the quality of food. He perceived multiple benefits associated with nutritious and healthy meals, such as, for example, being in better form after a good meal. He ate out regularly with his family despite reporting his embarrassment when being fed in public.

Mike

Mike was the youngest participant in this study. He was seen for two face-to-face interviews in his home which were followed up by emails. Mike lived at home with his wife and three children. Mike presented with anarthria and communicated via typing on his tablet. He was still mobile, but unable to walk long distances. His energy levels were decreasing and he needed to rest more frequently. Although he was still working on the farm, he required additional support, such as employing someone to

assist him with his work duties. Mike appeared to be a very sociable person and had a great sense of humour. He seemed to accept his diagnosis of MND as he perceived that he did not have any other choice. He was determined to maintain his previous routines in order to feel useful not only for his family, but also for his community.

Mike presented with significant dysphagia and he was eating a modified diet with difficulty. He also experienced frequent coughing or choking episodes during meals. Despite being aware of his swallowing problems Mike aimed to continue eating orally for as long as possible. He enjoyed the social aspect of meals very much. During the first interview, however, Mike reported that a decision regarding non - oral feeding was imminent and he was unsure if he should accept or reject it. He knew that his dysphagia was progressing and that his respiratory functions were also deteriorating, which he reported resulted in him feeling pressurised into making his final decision. The thought of living with a gastrostomy tube appeared to be extremely difficult for Mike for a number of reasons. Firstly, Mike perceived non - oral feeding as a means of prolonging his life. However, he was unsure whether he wanted to live longer at the advanced stages of MND. Mike reported that he had seen first-hand the terminal stages of MND as his cousin had died from this disease in the recent past. Although Mike wished to be with his family for as long as possible, he indicated that prolonging his life would increase their burden.

Sally

Sally was seen for two face-to-face interviews in her home, where she lived with her husband. Although Sally was relatively independent, she perceived that MND had caused a radical change to her lifestyle. She reported that the lack of speech was

the main limiting factor for her. It negatively impacted on her social interactions and made her feel less confident. It stopped her from going to her retirement club. As the eldest participant Sally had limited experience of technology and chose handwriting over the use of a speaking device. However, communicating by writing was quite challenging as Sally had dyslexia.

During the first interview Sally appeared somewhat distant from the world around her and it was hard to read her facial expressions and judge her emotional state. In addition, Sally appeared to be embarrassed due to her difficulties in managing her own secretions causing her to dribble and cough. Although Sally participated in two face-to-face interviews only the first was included in the analysis due to there being a gap of over 5 months between them.

Sally was non-verbal and presented with severe dysphagia. She was eating and drinking exclusively via a gastrostomy tube. Although Sally admitted that she missed food she knew that an oral diet would never be part of her life again. Sally's physical abilities were preserved and she was still fully mobile and independent. She was observed to engage in many household duties including cooking and baking. Sally was a house wife and viewed looking after her family as her priority. She was also talented at art and many of her paintings were displayed in her house.

Sally passed away eight months after her last interview.

Sheila

Sheila was seen for two face-to-face interviews in her home where she lived with her husband (her children were adults). She used a wheelchair, had limited functioning in her upper limbs, was non-verbal, and relied on non - oral feeding.

Sheila's life largely involved being in the house, predominantly in a living room and in front of the TV. Her grandchildren visited her frequently. Sheila's daughter was her primary caregiver and she referred to her as "my rock".

The interviews with Sheila were carried out in her living room. Sheila was seated in her wheelchair and connected to her gastrostomy tube. During the first interview Sheila initially appeared uncertain as she had not spoken to a stranger in a long time. She requested that her daughter be present during the interview for emotional support. At times, Sheila looked to her daughter for help when she had difficultly recalling past events. However, Sheila became noticeably more confident as the interview progressed and at one point in the interview she almost did not appear to notice when her daughter left the room to take a phone call. Unfortunately, the second interview with Sheila had to be terminated earlier than planned as Sheila became drowsy as a side effect of her medication.

Sheila reported that she missed her voice a great deal. She communicated well through writing but frequently complemented this with facial expressions and gestures as these seemed more natural to her. She attempted to use her body to enhance her communication and appeared to find the writing quite limiting. Sheila tried to express her emotions while writing through adjusting the size of the letters (important words were larger in size). Sheila reported that being non-verbal frequently led to being perceived as having an intellectual disability. She admitted that being respected by others (including health professionals) was very important to her.

Sheila presented with severe dysphagia and was unable to eat anything orally for about one year before this study.

Sheila passed away three months after the last interview.

Ted

Ted was seen for one face-to-face interview in his home followed up with a written interview. Ted's house appeared busy as he lived with his grown-up children and young grandchildren. His wife passed away over a year before this study. Ted strove to maintain his independence and was reported to frequently refuse offers of help from his children. Ted was still mobile but required support (such as a walking frame) due to decreased balance. He was non-verbal due to anarthria and communicated by writing. Ted's MND seemed to progress at a fast rate providing him and his family with little time to adjust. Despite the rapid physical deterioration Ted aimed to maintain his lifestyle as it had been prior to his diagnosis. He continued to engage in physically challenging activities on his own, such as driving long distances or hiking.

Ted presented with significant dysphagia. This resulted in his meals being prolonged and led to coughing and choking while eating. He attempted to manage these by limiting his diet to familiar foods. During the first interview Ted brought out the food he ate every day. His diet consisted of a few ready-made meals, mainly by a specific brand. Although it was very repetitive, it meant that Ted could ensure that his meals were the correct texture. However, shortly after the first interview Ted's swallowing deteriorated further and he decided to undergo a gastrostomy. This decision came as a surprise to his family, who reported not being aware that Ted had made the appointment for the procedure. Ted perceived non - oral feeding as a way of prolonging his independence by the provision of adequate nutrition, which would contribute to improved energy levels. He believed that it would assist him with

maintaining his current lifestyle for a longer period of time. This appeared to be his main motivation for undergoing the gastrostomy.

Ted passed away two months after his last interview.

Terence

Terence was interviewed face-to-face twice in his house, where he lived with his wife and a grown up son. Terence was non-verbal and communicated by typing on a tablet using his thumb. Terence reported that he was very aware of the continuous progression of MND. His fine motor skills were deteriorating making it hard for him to conduct any fine motor tasks, such as dressing or eating. Although he was still mobile, he reported losing his balance and strength in his legs. Despite his physical limitations Terence attempted not only to be independent, but also to be helpful to his family and community. He was very creative at finding solutions to manage his difficulties and developed some tools himself in order to improve his independence, for example, his own grips for his eating utensils to enable self-feeding. Terence also invented a bib to protect his clothes while he was eating, which he was able to put on and take off independently using his wrists only.

Terence appeared to enjoy our interviews and he reported being a very sociable person. He had strong faith in God and praying was very important to him. The spiritual side of life helped him cope with MND and provided a sense of calmness.

Terence had a gastrostomy tube in situ but refused to use it and reported that he greatly regretted having had it inserted. He appeared to be very aware of which food textures would be safe and suitable for him. He also commenced thickened liquids during his participation in this study.

Terence passed away five months after his last interview.

3.2 The Caregivers

The aim of this section is to introduce the reader to the caregivers who participated in this study. The sample of caregivers included two men and eight women all of whom were married. The mean age was 57.5 years (ranging from 37 to 83). All participants presented with a normal cognitive status. The mean duration of caregiving was estimated at above 24 months (ranging from 2 months to over 7 years). Six of the caregivers were spouses of a person with MND and four were a son or daughter of a person with MND.

Further demographic characteristics are presented in the table 2.2 below, followed by the contextual characteristics of each participant to provide more insight into their lives. Participant are presented in alphabetical order and each of them was assigned a pseudonym.

Table 3.2 Demographic Characteristics of the Caregivers

Caregivers	Age	Living area	Employment status	Number of children	Relation to the PwMND	Duration of caring
Angela	42	Rural	Part-time	2	Daughter	> 7 years
Helen	51	Suburban	Full-time	0	Daughter	5 months
Jack	70	Suburban	Retired	3	Husband	8 months
Jackie	53	Rural	Full-time	3	Wife	1.5 years
Kay	46	Rural	Part-time	3	Daughter	3 years
Mary	61	Urban	Full-time	1	Wife	> 6 years
Martha	65	Rural	Retired	0	Wife	> 2 months
Sophia	67	Suburban	Retired	5	Wife	8 months
Tom	83	Rural	Retired	5	Husband	3 months
Una	37	Suburban	Leave of absence	3	Daughter	6 months

3.2.1 Introducing the Caregivers: Short Stories

Angela

Angela participated in one face-to-face interview in her mother's house supported by emails and phone calls. She was a secondary carer to her mother as she lived in a different city. She aimed, however, to visit her mother as much as possible; in order to also provide support to the main caregiver (mother's partner). Angela's mother presented with a significant physical disability and her speech was severely dysarthric. Although she had dysphagia, she was still able to swallow softer consistencies. Angela was involved in all caregiving duties, including food preparation, when she stayed with her mother.

During the interview Angela was observed to be the most positive and enthusiastic of the caregivers. This was likely to be related to her having had the longest adjustment time, her provision of periodical care, and her younger age. For Angela, the hardest part was her mother's refusal to leave the house, attend for medical appointments and accept specialised equipment.

After the face-to-face interview was completed Angela informed the researcher that she had moved into her mother's house and became a full-time caregiver.

Helen

Helen was seen for one face-to-face interview carried out at her work place. She was involved in a shared model of care for her father (jointly with other siblings). Her father presented with a mild physical impairment but this was deteriorating rapidly. He was becoming more forgetful and his speech was becoming more dysarthric. His swallowing ability was decreasing too. Helen and her siblings attempted to make plans for the future in anticipation of their father's increased disability. This was particularly stressful, mainly due to differences in personal opinions.

Helen was a very active person who reported her love for sports and strong dislike of hospitals. She expressed a significant lifestyle change when her caregiving role began. All of a sudden she found herself confined to the house and unable to travel as she had before, which affected her preferred way of living, sense of autonomy, and previously established routines. Helen attempted to protect herself by maintaining some participation in her hobbies, but this required constant negotiation with her siblings. She was the only one without children and therefore was perceived as having more spare time and availability to take on caregiving duties.

For Helen, the interview was a rare opportunity to speak openly about her father and the issues related to his care. She reported that she found her participation in this study to be very therapeutic. Following the interview she admitted: "Actually I found it a great help / therapy for me to talk to non-family about dad. So… THANK YOU. You can quote that in your research" (personal communication, 02.06.2016).

Jack

Jack was seen for two face-to-face interviews in his home where he lived with his wife. Jack's wife was still physically independent at the time of this research, but had lost the ability to communicate verbally. Jack perceived his wife's dysphagia to be very mild and he did not appear to be very concerned about it. Instead, he spoke about his own health at length.

During the first interview Jack reported that he had been diagnosed with a life-threatening illness at the same time as his wife had been diagnosed with MND. He was required to undergo complex surgery in order to save his life. Jack's recovery period was complicated and he was still in hospital when his wife was given the diagnosis of MND. At the time of data collection Jack's condition had stabilised and he appeared to be in good form. However, he reported that he needed to be mindful of his own health and make some lifestyle adaptations due to his decreased health status. Jack approached his wife's disease from his own perspective which was shaped by his own experience of a serious illness. For him the MND, although not curable and terminal, was gradual and progressed slowly providing more time to adjust. In contrast, his own illness had been sudden and developed rapidly thus not allowing him time to adapt.

Jackie

Jackie was interviewed twice in her home where she lived with her husband and three children. At the time of her participation in this study her husband was still able to walk, but this was deteriorating. He was non-verbal and presented with significant dysphagia. Jackie was involved in preparing food for her husband. She

appeared to be very concerned about the consistency of food and its nutritional value. Jackie wanted her husband to opt for non - oral feeding as she hoped that this could prolong his life. However, she strongly believed that this should be his own decision and she attempted not to pressurise her husband into consenting for a gastrostomy tube insertion.

Jackie was emotional during the interviews as she reported being fearful of the upcoming future. She attempted to stay positive but this was difficult, especially when her family were still very young. Jackie knew that the future of the whole family would unavoidably change and she attempted to make some plans to ensure a sense of stability for her children. However, thoughts related to the future seemed to cause a significant emotional reaction in Jackie. Jackie knew that she was not able to stop the progression of MND and this helplessness appeared to be devastating for her. She aimed to maintain a "sense of normality" for her children, however, the rapid and visible progression of MND made her attempts very difficult.

Kay

Kay was seen for one face-to-face interview in her mother's house, followed by indirect contacts. She was the primary caregiver for her mother although she did not live in the same house. Kay's mother presented with a severe physical disability, anarthria, and dysphagia. Kay had witnessed all stages of her mother's dysphagia from her eating a mildly modified diet to being fed exclusively via a gastrostomy tube.

During the interview Kay reported that she had been managing her caring duties mostly alone as she received inconsistent help from external services. Kay highlighted that becoming a caregiver had not been her choice; she felt that she had

been expected to do it based on her personal circumstances i.e. not having a full-time job.

Kay perceived there to be a strong link between her own and her mother's emotional status. Therefore she was determined to be positive and avoided focusing on the difficulties related to MND, especially in front of her mother. She admitted that supressing her own feelings was the hardest part of caregiving for her.

Mary

Mary was seen for two face-to-face interviews in her home where she lived with her husband and their grown up daughter. Her husband presented with a severe physical disability and required assistance with all activities of daily living. He was also unable to talk due to anarthria. Although he presented with dysphagia he was managing a modified diet relatively well. Mary was responsible for preparing an appropriate diet for her husband and she also fed him at times. She described her husband as "very demanding". She reported that he refused respite, which meant she was unable to take a break from her caregiving duties. Mary's last holiday was around 6 years ago.

Mary was interviewed twice in her living room. Her husband was in the kitchen and Mary whispered at times to ensure he would not hear her utterances. Mary appeared to be quite nervous during the first interview. She seemed tired and worried as she saw "no end to it" [her caring role]. Her sleep was disturbed by caring duties. She was glad to be working full time, although she reported that she did not have any time to herself. Her life was very busy, one duty followed another. Mary reported that work had become an escape from her life with MND and allowed her to maintain her

preferred standard of living. Despite expressing her physical and mental exhaustion, Mary appreciated having a good job and not experiencing any financial strain.

Martha

Martha was interviewed twice in a palliative care centre where her husband accessed multidisciplinary services. Martha's husband was diagnosed with MND a couple of months prior to this study; however, he was reported to deteriorate rapidly allowing her little time to adjust. Although still physically independent, he was losing his speech and ability to breathe. His stamina was poor, which affected their mutual routines, such as taking their dogs for a walk or going for short city breaks. His dysphagia appeared to be mild; however, Martha was very aware of the risks associated with having a swallowing impairment. She provided discrete supervision and modified her husband's food textures covertly, as she attempted not to make him aware of his dysphagia. Her husband's ill-health had destroyed all their future plans as the couple had wished to travel and had considered moving abroad, which was no longer possible. Martha had no children and after her husband became sick she reported feeling very lonely. She spent most of her days alone as her husband needed to rest frequently.

Martha retired shortly before her husband was diagnosed with MND. They lived in a rural part of Ireland and had to travel over 90 km to access a hospital, which was a source of additional stress for Martha. She often reported that she did not feel adequately supported in managing her husband's ill-health at home.

Sophia

Sophia was seen for one face-to-face interview in her home where she lived with her husband. Sophia's husband had been through another serious illness shortly prior to MND, which complicated his diagnosis. At the time of this research he was fully mobile, but became completely non-verbal. His swallowing ability was deteriorating rapidly and he needed to supplement his oral diet with non - oral feeding. Sophia continued cooking for her husband and dining out with him despite reporting feeling embarrassed at times.

Sophia was calm and confident during the interview and seemed well prepared and able to express her reflections and opinions very clearly. She reported that she had required about six months to adjust to her new life with MND and to be able to talk about MND. Sophia's priority seemed to be finding a balance between having to deal with the difficulties related to MND and maintaining a good quality of life.

Tom

Tom was seen for two face-to-face interviews in his home where he lived with his wife. They had three grown up children. One of his children lived close by and visited him daily. Tom's wife was fully mobile, but she was non-verbal and presented with severe dysphagia (she was unable to eat orally). Tom admitted that his wife's illness came as a complete shock to him. She was younger than him and Tom always thought that she would be looking after him in his old age.

Tom was involved in cleaning his wife's gastrostomy tube and administering the feeds and medications. Although Tom strived to provide the best possible care for

his wife he found it overwhelming, with his own health issues (such as deteriorating vision) no doubt contributing to the strain. As the couple lived in a rural part of Ireland they had to travel over 80 km to avail of specialised services at a hospital.

Although Tom was generally calm and relaxed during the interviews, he became quite angry when describing some of his experiences of the health service. He reported lengthy delays in addition to miscommunication between professionals. Tom felt that his wife's diagnosis was made very late, as she had been experiencing visible symptoms of MND, such as losing her ability to speak and swallow, for a long time prior to the diagnosis. This delay, in his opinion, deprived her of benefiting from early treatment with medication.

Una

Una was interviewed once in her home. She had moved in with her husband and children to live with her dad, who was diagnosed with MND, shortly before taking part in the interview. Una's life was very busy as she had young children. Moving in with her father was more convenient and enabled her to spend more time with him. Una's father was non-verbal and presented with severe dysphagia requiring a non - oral diet.

Una had to attend to her children during the interview. She appeared very energetic and able to multi-task well. The quality of her father's life was her main priority and Una attempted to support her father's wishes as much as possible. Sometimes this created conflict with the professionals involved in his care, when her own / her father's perspective differed from the medical approach.

CHAPTER 4 RESULTS: THE PEOPLE WITH MND

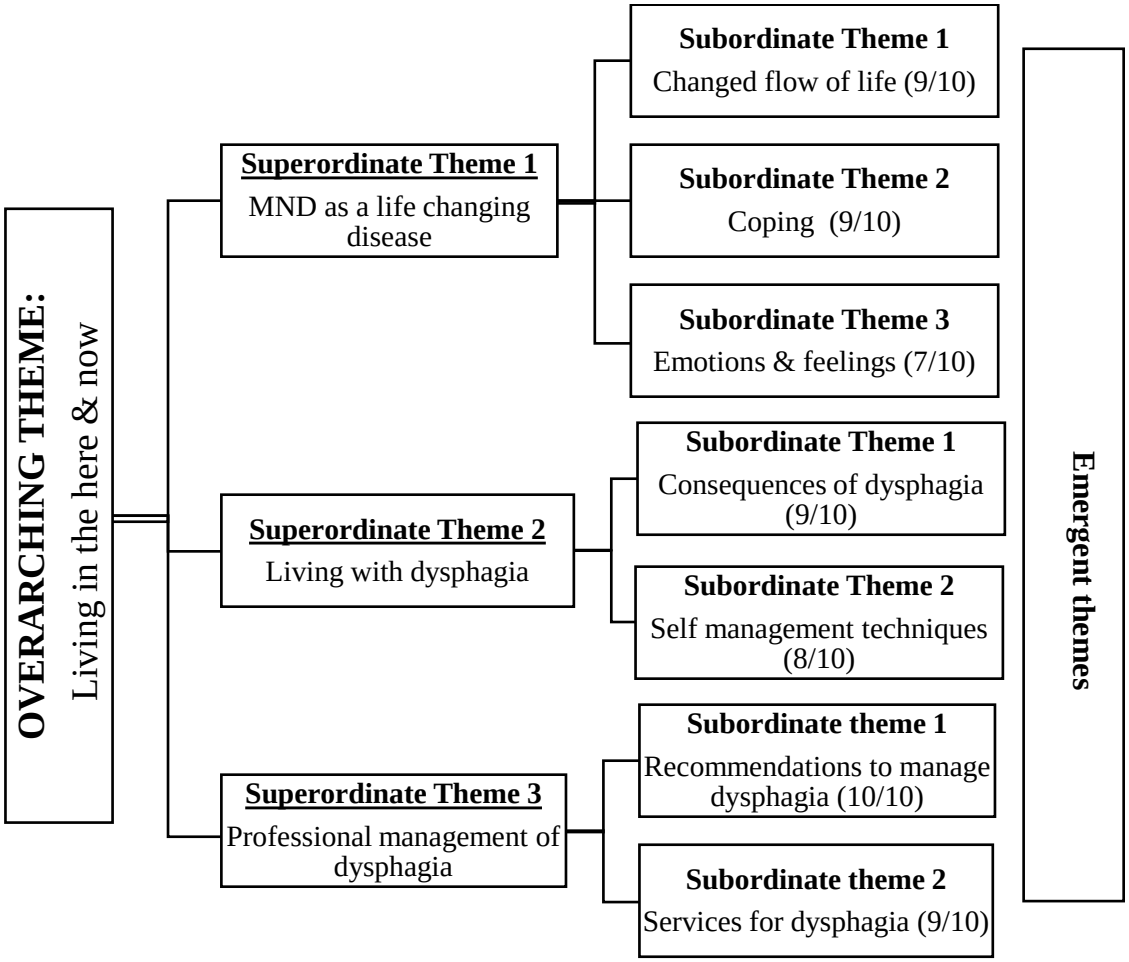
Introduction

This study aimed to investigate the experiences of dysphagia in MND and how people living with MND (both people diagnosed with MND and their caregivers) make sense of their swallowing impairments. This chapter focuses on the group of participants with MND. The results of the Interpretative Phenomenological Analysis are outlined in this chapter including (1) a brief overview of the hierarchy of themes and the frequency of subordinate themes in the sample (Figure 4.1), (2) a more in-depth presentation of the hierarchy of themes including the emergent themes and their prevalence (Table 4.1), (3) a presentation of themes supported by selected quotes from the sample (Table 4.2) followed by (4) the textual overview of themes. This chapter follows the guidelines developed by Smith et al. (2009) regarding the writing up of studies with larger samples where a group-level analysis of the main themes is supported with the idiographic details of representative participants. The researcher attempted to balance the convergence and divergence within the group when structuring this chapter. This was achieved by presenting the unique / idiographic emergent themes in addition to recurrent themes in order to preserve the specific context and meaning expressed by an individual.

A visual overview of the results in relation to the hierarchy of themes is presented in figure 4.1 below. One overarching theme was identified during cross-case analysis: Living in the here and now. In addition, three superordinate themes emerged: (1) MND as a life changing disease, (2) Living with dysphagia, and (3) Professional management of dysphagia. The number of participants who represented each subordinate theme is presented in the bracket following the name of the theme (Figure

4.1). The prevalence of themes was calculated when establishing the higher order of themes, as according to Smith et al. (2009) it is important to measure and define the recurrence of themes within the group of participants to enhance the validity of the results. In this study, subordinate themes were included in constructing superordinate themes only if they were present in over half of the sample. In accordance with the guidelines of IPA (Smith et al. 2009) and acknowledging the researcher's background, the data was interpreted from a speech & language therapist's perspective.

Figure 4.1 Hierarchy of Themes: PwMND



Presentation of Findings

This section presents the main themes which emerged during the cross-case analysis of the participants with MND. The hierarchy of themes is presented in detail in table 4.1 in order to enable the reader to observe how the higher order themes were established. The overarching theme is located at the top of the table. The three superordinate themes are located in the first column, the seven subordinate themes in the second column, and finally the representative emergent themes are presented in the third column (with the frequency of emergent themes presented in brackets).

In the subsequent table (Table 4.2) the participants' quotations are presented in order to provide examples of how the emergent themes relate to the original transcripts and how they were used to create superordinate themes. The most representative, significant, and rich quotations were chosen to support each theme. In addition, an effort was made to include the voice of every participant. The number of supporting quotations for each emergent theme, therefore, is not the same. Finally, as the majority of people with MND in this study (9/10) communicated non-verbally and often had physical difficulties accessing their communication devices, the quotations obtained from this group of participants were generally shorter in comparison to the group of the caregivers.

Following table 4.2, the overarching theme is introduced. The three superordinate themes are then presented. The first superordinate theme was related to living with MND in general and reflected issues reported as important by the participants. Although this theme did not include any data directly related to dysphagia, it provided a deeper understanding of participants' experiences of living with a swallowing impairment as part of MND. Data analysis revealed that dysphagia

in MND should not be viewed in isolation as participants' approach to dysphagia was strongly rooted in their every-day experiences.

Table 4.1 Hierarchy of Themes: PwMND

OVERARCHING THEME: Living in the here and now		
SUPERORDINATE THEMES	**SUBORDINATE THEMES**	**EMERGENT THEMES**
MND as a Life Changing Disease	**Changed Flow of Life**	Awareness of deterioration (10/10) Lost skills & lost independence (9/10) Changed rhythm of life (6/10) Fragility of life (4/10) Ruined plans (3/10)
	Coping	Maintaining previous routines (10/10) Finding solutions to manage (8/10) Family support (10/10) Avoidance (5/10) Making sense of MND (4/10) Maintaining a positive attitude (4/10)
	Emotions and Feelings	Loss & grief (10/10) Nervousness & uncertainty (8/10) Frustration (5/10) Happiness (2/10) Hatred (1/10) Diagnosis as a relief (1/10)
Living with Dysphagia	**Consequences of Dysphagia**	Eating as risk (9/10) Changed role of food & diminished pleasure from eating and drinking (9/10) Changes in social aspect of meals (8/10) Repetitive & limited diet (6/10) Inability to eat preferred food (5/10) Emotional impact (5/10) Changed participation in celebrations (1/10)
	Self-management Techniques	Noticing physical symptoms of dysphagia (10/10) Paying special attention during meals & modifying the eating environment (10/10) Changed preparation of food (9/10) Excluding risky food (9/10) Eating, drinking, & swallowing routines (7/10)
Professional Management of Dysphagia	**Recommendations to Manage Dysphagia**	Non - oral feeding (10/10) Modifications of fluid consistency & textures of food (10/10) Adherence & adjustment (10/10)
	Services for Dysphagia	General satisfaction (10/10) Associated emotions (7/10) Suggestions for change (4/10)

Table 4.2 Presentation of Themes Supported by Chosen Quotations from Transcripts: PwMND

Superordinate Theme 1:

MND as a Life Changing Disease

Subordinate Theme 1: Changed Flow of Life
Emergent Themes

Awareness of deterioration

Bob: The main motivation [to lose weight] was that my mother died aged 88 and I wanted to live as long as she did (that is not possible) and lost eight stone over 20 months. (lines 804-805)
Carmel: (…) they told me that there was no cure. (…) and that all my legs and my hands might go. (lines 154-156)
Joseph: The message from my brain isn't being transmitted fast enough to my muscles. I guess motor neurone cells are dying. (lines 48-49)

Lost skills and lost independence

Mike: I miss talking. (…) I miss phone calls. (…) Not being able to do my business. (…) On the phone. (…) Or at the shop, bank, chemist, counters. (lines 1135-1145)
Sally: Well I cannot even do shopping without Liam [her husband]. (…) I have to have Liam all the time (lines 59-60) I would not be able to do all myself. The not being able to talk. Or go out myself. (lines 115-116)
Joseph: My wife gets me out of bed at 6.30am I eat breakfast. I shave and wash teeth. (…) I stay in night-coat until carer arrives about eight. She [the carer] helps me shower and changes me into day clothes. (lines 23-26). (…) I can't prepare food for myself if it's outside my routine. I look forward to carer calling. (line 215)

3. **Changed rhythm of life**

Terence: Everything is slow now. (…) make porridge, clean up, check clothes, trips to toilet, check phone and email, visit my uncle who lives down yard, do some exercises and bookwork. (lines 5-6)

Fragility of life

Eileen: And I think you have to go through it to understand it. (…) You couldn't explain it. You just couldn't explain it to people how fragile everything is in your life. I think that is probably what gives you the feeling of insecurity. (…) And lack of confidence whatever you like, you know. (lines 937-944)

Ruined plans

Bob: It [MND] ruined all our plans. (…) We're both retired and we were going to travel a lot. (lines 330-333)

Subordinate Theme 2: Coping
Emergent Themes

Maintaining previous routines

Mike: I do some light work now [on the farm]. Driving. (lines 56-61).

Bob: The last time we went to a restaurant was last Wednesday. We stayed over in the [names] Hotel to see how I could cope with it. It worked out very well and the bar/restaurant was spacious and well aired and we had a full dinner in there. Yesterday (Tuesday) we went for coffee and cake in [named a café] and that was fine, we go there once a week. (lines 730-734)

Finding solutions to manage

Terence: The swallowing is not my biggest problem with bread. Preparing it with butter, cheese or marmalade, putting it into my mouth and keeping it inside is a bigger problem. I will find some way around it. (lines 664-666)

Joseph: I have news update prepared before carers call. As a result I'm more relaxed when carers call if I have something typed in advance. (lines 526-527)

Family support

Eileen: I'm, I consider myself very lucky to have so much [support from family]. (line 46)

Sheila: My daughter… She is my rock. (line 251)

Avoidance

Eileen: (…) I don't give myself the time to analyse myself. (lines 87-88)

Sheila: I don't like it [MND clinic]. (…) The team are lovely. (Interviewer: There is something you don't like and I would like to know what). Looking at other people. Different stages. (lines 183-188)

Making sense of MND

Sally: We have had a good life and I was going to offer any pain or discomfort to God in thanks giving for all. (line 54-55)

Eileen: Em, but it is like this, we'll all die sooner or later and something will kill us and I suppose it's God's will. (946-947)

Maintaining a positive attitude

Joseph: I consider myself lucky that I haven't lost my eyesight and that my brain is still functioning normally. (At least I think so. Ha!). (lines 324-325)

Subordinate Theme 3: Emotions and Feelings

Emergent Themes

Loss and grief

Mike: I miss talking (…) I miss phone calls. (lines 1137-1139)

Nervousness and uncertainty

Eileen: I suppose, I got nervous and I still have that nervousness. (…) And I lost my confidence. I couldn't bring myself to go up and read my [readings in the church] and if I had them I'd be going, I'd be totally…(…) Stressed. (…) then I found, em, I was a bit nervous going out on my own. (…) I think it was a generalised thing. (…) that I got nervous of everything. (lines 657-680)

Fiona: I don't know how my illness will progress or what I will need "down the line." (line 1169)

Frustration

Joseph: I'm sighing because I'm getting frustrated with myself. (line 47)

Happiness

Terence: Lucky I am never sad or down now thank God but there are many things that keep me happy and very happy. (lines 246-248)

Hatred

Joseph: I hate all the medical baggage that goes with mnd. (line 45)

Diagnosis as a relief

Sally: It [diagnosis of MND] was such a relief to know at last what was wrong with me. (line 30)

Superordinate Theme 2:

Living with Dysphagia

Subordinate Theme 1: Consequences of Dysphagia

Emergent Themes

Eating as risk

Sheila: For safety I stopped eating. (line 209)

Changed role of food and diminished pleasure from eating and drinking

Mike: I think it [food] is important to maintain weight. (line 854)

Carmel: It's [food] really important for the muscles. (…) Before I didn't care. (lines 595-606)

Ted: I drank two cups of tea. (…) I didn't enjoy it. (lines 57-59)

Changes in social aspect of meals

Eileen: Believe it or not I'm happier eating here in the home now. I wouldn't be comfortable [eating in restaurants]. (…) If you have strangers with you you're more likely to splutter. (lines: 214-216)

Terence: [talking about eating out] Messy. Easier to eat at home. (lines 143-146)

Repetitive and limited diet

Joseph: Ann [wife] prepares casserole in a bag (Knorr season and shake) the night before. Chicken pieces, tomatoes, peppers. This is served with peas and mashed potatoes. (…) On alternate days I eat Donegal Catch fish, cooked in a sauce. This is served with vegetables and mashed potato. Followed by yogurt and cup of tea. (lines 107-112)

Inability to eat preferred food

Eileen: I like brown bread but have recently changed to soda bread [brown bread too crumbly] which I toast lightly and is perfect. (line 1030)

Emotional impact

Eileen: Em, it was as if you had the most ferocious feeling of panic that you were going to choke. (lines 498-499)

Changed participation in celebrations

Sally: I was 80 last week. All the children came and we were all together. Lidia [daughter] had a nice meal just the … but I could not eat. Or have a glass of champagne (1.125-127).

Subordinate Theme 2: Self-management Techniques

Emergent Themes

Noticing physical symptoms of dysphagia

Sheila: (Interviewer: (…) When you say it [swallow] was difficult, what was happening?)
Choking. Getting stuck in throat. (…) The tongue couldn't move. The food stayed in there. (lines 34-37)

Paying special attention during meals and modifying the eating environment

Mike: I have to be… (writing) Careful. (…) About bite size. (lines 159-163)
Joseph: I still have swallowing difficulties at times. I'm continuously on my guard. (line 569)
Eileen: I won't go out [to eat] anymore because I don't want to, I'm afraid in case something would happen like that [choking]. (lines 125-126)

Changed preparation of food

Terence: I use minced a lot. Boil potatoes and veg well so they are soft. (lines 381-382)

Excluding risky food

Carmel: I find it hard to eat tomatoes. (…) Because they're too hard. (…) I can have potatoes (writes: potatoes, salad). (lines 394-400)

Eating, drinking, and swallowing routines

Ted: I eat same things. (…) I have it [ready-made meal] every day. Same meals. (lines 78-80)

Superordinate Theme 3:

Professional Management of Dysphagia

Subordinate Theme 1: Recommendations to Manage Dysphagia

Emergent Themes

Non - oral feeding

Terence: Before that [PEG tube insertion] I would climb over gates. Now I am very aware of danger of falling. (lines 214-215)

Eileen: I mean all I know [about tube feeding] is you get your full diet, you know and I mean if it's necessary its necessary and they said I could still take a sip if I wanted to and they told me that if I had too much, em, secretions they can give me something to dry the mouth. (lines 296-300)

Sally: (Interviewer: What was your impression [when you saw the feeding tube for the first time]?) (wrote) Relieved. (lines 50-53)

Modifications of fluids consistency and textures of food

Mike: (Interviewer: Oh you have a thickener. How is that going?) *Mike throws his eyes to heaven. Laughs*. (Writes) Not good. (…) Terrible… (…) I'm… managing okay. (lines 415-438)

Adherence and adjustment

Interviewer: How did you feel about the changes in the diet?

Sheila: I didn't like it at all. No. Takes time. (…) You have to process it in your head. And believe that the danger was there. (lines 1.201-208)

Joseph: I follow the advice I have been given. I eat moist mashed food and drink slowly. (line 466)

<u>Subordinate Theme 2: Professional Services for Dysphagia</u>

Emergent Themes

General satisfaction

Bob: Everybody has been very great. (line 424)
Ted: (Interviewer: What do you think about the services you received for your swallowing?) OK – for a swallowing. (line 117)
Carmel: I'm very happy [with the service received for dysphagia]. (lines 656)

Associated emotions

Terence: I was pleased that she [SLT] thought I was not bad. (lines 331-332)
Eileen: It can be a stressful hour [MND Clinic]. When you come home you're exhausted. (…) I find it stressful. (lines 339- 341). It is, very [stressful], and especially when the whole group [MND Clinic] are in front of you, you dread it before you go. (line 508)

Suggestions for change

Joseph: Community Speech Therapist texts me every few months. She decides whether or not to visit me based on my self-analysis. (…) It's not for me to decide how often my swallow should be tested. (lines 453-461)

Over-arching Theme: Living in the Here and Now

This section introduces the overarching theme for the participants with MND as identified through the process of IPA: Living in the here and now. This was how the people with MND in this study approached and coped with their disease – by taking it day by day, by being focused on the present, by continuing participation in their hobbies, work, and routines. The overarching theme should be acknowledged throughout this chapter as it forms a basis for understanding participants' lives, including their experiences and perceptions of dysphagia.

Living with MND seems to resemble a journey: from the shock of the diagnosis, through the gradual acknowledgment of their own deterioration, to the commencement of a new life with MND. This new life with MND appears to be strongly focused on being in the here and now. Although the diagnosis was perceived as cruel and irreversible by almost all participants (except Sally), the majority reported trying to adjust to it (as they reported having "no choice") in an attempt to "make life as normal as can be". Joseph reported: "I have mnd so long I've worked out strategies for coping and making life as normal as can be". (…) "I have no choice but to accept mnd" (line 158). The only participant who appeared unable to adjust to his diagnosis of MND was Bob. This could be possibly explained by the fact that he had had the shortest time post-diagnosis of all participants (just over 2 months).

The desire to live in the here and now appeared to develop over time. At some point after the diagnosis all participants acknowledged their own deterioration and began to gradually adjust to their new lives with MND. The majority reported aiming to maintain their own independence and develop own strategies to manage their

physical disabilities caused by MND, as if trying to maintain a sense of control over their lives. As the course of disease progression in MND is unpredictable, most participants did not appear to be focused on the future. In order to cope, they also appeared to distance themselves from their lives pre-MND when they were in a good health. Instead, their main focus was on the here and now, the new reality, the current day, the current moment. The "now" became most important, the past gradually faded, and any thoughts regarding the future were reported as frequently blocked, often in self-protection. As their bodies physically deteriorated, participants' cognitive functions remained intact. Achieving a sense of being in the now required a cognitive effort and in the current study the power of the mind appeared to dominate over the physical deterioration.

The overarching theme is frequently reflected upon in the presentation of the findings throughout this chapter as it provides an important context for understanding the participants' experiences.

4.1 Superordinate Theme 1: MND as a Life Changing Disease

This section presents the first superordinate theme and the corresponding subordinate themes which emerged from the data. The superordinate theme 1: MND as a life changing disease consists of three subordinate themes: (1) Changed flow of life, (2) Coping, and (3) Emotions and feelings. Although superordinate theme 1 is not directly related to dysphagia, it is presented first in order to orientate the reader to various aspects of living with MND, which appeared important to the participants. It includes the data which was spontaneously shared by the participants, unlike the topic of dysphagia which often needed to be prompted by the researcher. Prior to reporting

on dysphagia - related experiences in subordinate theme 2 and 3 it seems crucial to acknowledge how MND affected the lives of the participants' in general. This knowledge should form a basis for understanding how the people in this study approached and managed their swallowing impairments.

4.1.1 Subordinate Theme 1: Changed Flow of Life

It emerged that the diagnosis of MND marked the start of a new chapter in life for the participants in this study: living with MND. Almost all participants (apart from Fiona) reported significant life changes due to MND. In total, six emergent themes were identified and these are presented in separate sections below: (1) Awareness of deterioration, (2) Changed rhythm of life, (3) Lost skills and lost independence, (4) Inability to live as before, (5) Fragility of life, and (6) Ruined plans.

Emergent Theme 1: Awareness of Upcoming Deterioration

All participants indicated their awareness that MND was progressive and not treatable. Their prognosis was typically communicated at the time of diagnosis thus making the moment of diagnosis even more traumatic. Carmel became significantly upset during the interview when she reported: "They [doctors] told me that there was no cure. (…) and that all my legs and my hands might go" (lines 145-147). However, none of the participants reported being given any specific time lines related to the expected progression of MND and the disease was frequently described as unpredictable. While participants attempted to cope by immersing themselves in the

here and now, they also appeared to maintain some hope, in relation to having sufficient time to experience important events, a slow progression of MND, managing to live with the disease every day, and finally hope that a cure may be found in time. Each participant viewed their disease through their own lens, which was shaped by their previous experiences, beliefs, circumstances, cultural influences, and personality. Their perceptions appeared to differ from those of the medical professionals, for example, although Terence reported being diagnosed with a bulbar onset of MND by his neurologist, he stated that the disease for him had started in his hands. Terence perceived his hand function to be more important than his ability to speak and swallow. He underwent a gastrostomy tube insertion hoping to manage his dysphagia and began using an electronic device to communicate. However, losing hand function meant an inability to work and be independent with no possibility of compensating for these losses.

All participants were aware of their unavoidable deterioration and often used words which indicated their expectation of change, such as adding the phrase "at the moment" when describing their abilities or using the word "yet" indicating their expectation of further deterioration ("I don't have any problems yet"; Fiona, line 339). However, no one knew when and how their disease was going to progress. This awareness of their upcoming deterioration greatly impacted on all participants, who often commented on approaching their lives differently post-diagnosis. All participants reported that as the physical impairments began to develop they altered the previous flow of their lives.

Emergent Theme 2: Lost Skills and Independence

The changed flow of life resulted in many losses. All but one participant (Fiona) reflected on their deteriorating abilities, such as driving or walking. However, the loss of speech appeared to be one of the most devastating consequences as it disrupted the participants' sense of their own identity and prevented many social interactions. Almost all participants in this study (apart from Eileen) lost their ability to communicate verbally which was associated with many consequences, such as the inability to continue to work or participate in social events subsequently leading to an increased sense of isolation. Joseph highlighted his distress associated with having to communicate non-verbally: "I listen a lot to radio. I read news online. It gives me good leeway to communicate with carers. Makes me less nervous meeting people" (lines 33-34). He also added "I find it a little stressful interacting with different people" (lines 479-480). When asked about his interactions with other people Bob stated: "I try to avoid them" (line 622). Although all non-verbal participants attempted to use an alternative method of communication, the majority indicated that this altered way of communicating negatively impacted on their social interactions as it was not perceived to be as natural as speech.

Mike described his current life as "a massive change" and grieved over his lost skills. He pointed out that the change affected not only him, but also others in his environment.

> It is a massive change to everyone you know. (…) And for myself also. (…) Everybody has to work around me, meals, etc. (…) I am not as mobile as before. I miss talking. (…) As they do. I miss phone calls. (…) Not being able

to do my business. (…) On the phone. (…) Or at the shop, bank, chemist, counters. (Mike, lines 579-588)

In saying "I miss talking. As they do" Mike acknowledged the impact of his disability on his family, which he found distressing. He stated "Everyone has to work around me" which was not a usual situation for Mike, who was the youngest participant. Suddenly his needs were prioritised by the family, his role in the family had changed. The physical deterioration influenced his independence and prevented him from conducting some of his important routines. When Mike began to name things he was unable to do anymore his rate of writing became faster. One word triggered another. This was the only time during the data collection that Mike seemed to complain about his situation. The analysis of Mike's interviews revealed that he perceived himself as a growing burden, which was hard for him to accept. All of the sudden he was not able to protect his family in the same way as he had been prior to MND. Instead, he required their ongoing help as his level of independence diminished.

When Sally spoke about living with MND she reported: "Well I cannot even do shopping without [husband]. We used to go for meals out and have friends in but never … I cannot do that. I have to have [husband] all the time and he gets very tired because of me" (Sally, lines 59-60). This quotation illustrates the extent of the change in Sally's life: lost independence, changed participations, changed social life, and the awareness of being a burden to a caregiver. For half of the people with MND in this study routines previously carried out almost automatically (such as shopping) became challenging and gradually impossible due to MND.

Emergent Theme 3: Changed Rhythm of Life

A changed rhythm of life was reported by over half of the participants. This was primarily related to the progression of physical disabilities which extended the time required by people with MND to conduct their everyday activities, such as dressing or eating. "Everything is slow now", reported Terence. Terence's "now" referred to his new reality with MND, where his physical abilities were deteriorating. The slowness affected Terence's ability to "make porridge, clean up, check clothes, trips to toilet, check phone and email, visit my uncle who lives down yard, do some exercises and bookwork" (lines 5-6). The changed rhythm of life meant that participants became unable to maintain their previous lifestyles. Joseph, for example, indicated his incapacity now to be physically active: "Prior to mnd I used to exercise daily. Swim or walk (preventative medicine)" (line 80).

Emergent Theme 4: Fragility of Life

Some participants described their lives as uncertain and fragile, which was associated with not knowing when new symptoms of MND would develop. "You just couldn't explain it to people how fragile everything is in your life" said Eileen (lines 437-439). Moreover, she later indicated that the fragility of life made her feel at constant risk: "I go to mass on a Saturday night and [my husband] comes with me and you might even feel nervous during the mass. (…) In case something would happen, you know" (lines 646-648).

Emergent Theme 5: Ruined Plans

Three participants stated that MND had ruined their future plans, such as travelling or moving abroad. Bob spoke about ruined plans most frequently. He appeared particularly angry and upset when speaking about it. His diagnosis of MND had only been confirmed 2 months prior to this study and he did not seem to have adjusted to living with this disease as much as Sheila and Mike had, who were respectively 3 and 1.5 years post-diagnosis.

4.1.2 Subordinate Theme 2: Coping

The participants with MND attempted to cope with the disease on their own and none reported looking for psychological help by contacting professionals or even other family members. It became apparent that the people with MND in this study aimed to avoid being a burden to their caregivers, family members, friends, and also professionals. The physical deterioration was beyond their control and there was a shared desire to maintain participation in various routines and hobbies, even if this required extensive adaptations. Developing their own strategies for coping with MND gave the participants the feeling of living with MND rather than dying from it. Their strong motivation to adapt and cope with MND appeared to reflect their desire to live. The topic of life and death was intermittently initiated by the majority of participants, for example, Joseph reported "I want to live but I'm not afraid of dying" (line 62). He indicated that he was not afraid of dying from MND, but also that he was not afraid of MND in general. He appeared not to have any respect for MND and he was proudly continuing to fully live his life despite his severe disability. He was so careful about spelling on his device that his consistent de-capitalising of "MND" appeared to be a

conscious decision, as if to demonstrate that MND didn't deserve to be capitalised. He didn't want the letters to stand out in his text; it was not a name he respected. "I have mnd so long I've worked out strategies for coping and making life as normal as can be" (Joseph, line 156).

In total, six emergent themes were identified under sub-ordinate theme 2 (Coping) and these are presented in separate sections below: (1) Maintaining previous routines, (2) Finding solutions to manage, (3) Family support, (4) Avoidance , (5) Making sense of MND, and (6) Maintaining a positive attitude.

Emergent Theme 1: Maintaining Previous Routines

All participants spoke of their desire to continue their participation in previous routines and hobbies despite adaptations being required. Bob, for example, reported: "I used to do the market in the summer and (*uninteligible*) but I can't do that now. Then we moved it [the market] in the house" (lines 377-379). Joseph, who was one of the most physically disabled participants, "still run the house from [his] I-Pad" (line 20). "I sit here and answer emails do banking etc." (line 28). Joseph felt useful to his family despite his severe physical disability "I guess they [family] need me to balance the books etc" (line 60).

> After three I think about making dinner. My wife usually prepares most of it night before. I lock my wheelchair behind me when I'm standing near sink. Sylvia [daughter] sometimes helps me make dinner (lines 35-37). (…) I help my daughter to prepare dinner in the evening so that it'll be ready when my wife gets home (218-219). (…) I try my best to tidy kitchen etc. from my wheelchair. (lines 475-476)

Joseph's engagement in dinner preparation was an important part of his daily schedule. This appeared to be the only activity carried out jointly with his daughter ("she helps me" / "I help her"). His involvment in making dinner was also Joseph's way of supporting his wife (the main caregiver) in that the food was ready for her on her return from work.

It emerged that the participants wished to maintain their engagement in various activities not only to satisfy themselves, but to feel useful and helpful to other people. Therefore reciprocal interactions with others were not uncommon, for example, Terence managed to complete a few community projects despite his multiple disabilities caused by MND. These accomplishments were a positive experience for him and he was pleased that his work had been appreciated by others.

> Last year I brought to a close two things I was working for a long time. In May we published the local [name] Club [name] History. I was editor with a great committee. Later I worked with some locals (…) to record all the headstones in our local [name] graveyard (…). Getting this done was a great relief and hearing compliments from time to time makes me happy too (lines 284-291). (…)
>
> *Interviewer: So you've done the book, the head stones, what's next?* Putting my own accounts and things in order. (lines 536-538)

Terence seemed to be motivated to continue his contributions to the community although he was aware of his own unavoidable deterioration. He aimed to "put his own accounts in order", not to leave any unfinished tasks behind. Recording the old head stones could indicate Terence's respect for human life in general. This could be

viewed as his motivation to stay positive, strong, and proactive, despite having to face a terminal disease. He appeared grateful for his own gift of life.

Emergent Theme 2: Finding Solution to Manage

The majority of participants were focused on finding solutions to overcome the limitations caused by MND, which appeared to optimise their sense of independence, dignity, and participation. For some, looking for solutions to manage a particular issue caused by MND appeared to be quite a creative process requiring good problem solving skills. It emerged that in order to live in the here and now the participants generally focused on finding solutions to manage their problems rather than just focusing on the problems themselves. "The swallowing is not my biggest problem with bread. Preparing it with butter, cheese or marmalade, putting it into my mouth and keeping it inside is a bigger problem. I will find some way around it" (lines 664-666). In the quotation above Terence spoke about his eating difficulties, but he did not complain about it. Instead, he immediately stated his ability to find a solution.

Emergent Theme 3: Family Support

All participants reported that family support was an important factor which influenced their ability to cope with MND. Joseph, for example, stated that: "My daughter and my wife give me a reason to live" (line 59). Sheila referred to her daughter as being her "rock" (line 251). Terence indicated the importance of his family and caring about "peace" at home: "Also at home as long as there is peace in the home and family I am happy" (line 250). He also suggested that there was a relationship between his own mood and well-being, and that of his family.

Emergent Theme 4: Avoidance

This continuous process of adapting to life with MND required the participants to stay focused on the here and now. It emerged that most participants distanced themselves from the past (often demonstrated by their difficulties recalling / talking about pre-MND life) and avoided thinking about the future. This became particularly evident when participants discussed their attendance at MND clinics, which they often reported as being a stressful and disliked experience as it involved meeting people at different stages of the disease, thus interrupting their efforts to live in the here and now. Sheila said: "I don't like it [MND clinic]. (…) The team are lovely". *(Interviewer: There is something you don't like and I would like to know what*). "Looking at other people. Different stages" (lines 183-187). Half of the participants reported that they avoided self-reflection regarding their lives with MND. Eileen, for example, stated: "I don't give myself the time to analyse myself" (line 85) indicating that she was not giving herself any time to reflect on her situation. Instead, she aimed to be constantly occupied.

Participants' insight into their own abilities and disabilities appeared to be complex in the context of MND. On one hand, participants had to be aware of their own difficulties in order to make the necessary adaptations. For example, Terence reported: "I wouldn't be able to do any of the heavy work on the farm, milking etc. My son is farming full time now" (lines 84-85). On the other hand, participants avoided self-analysis in relation to their medical status, i.e. the stage of MND they were currently at. Joseph admitted that he "would not trust his own analysis" and that his perceptions of his own health may vary from that of the professional (lines 454-455). Mike reported that attending medical appointments was "hard" as it involved

being told about the progression of his MND. This was not something Mike would be able to predict himself (lines 518-528). Mike's primary focus was not on self-evaluation, in fact, he avoided it as much as possible. Instead, he strived to maintain his previous routines, aimed to be useful to his family and attempted to fulfil his duties as before. It appeared that the people with MND were selective during self-analysis and avoided reflecting on their problems from a medical perspective. However, in order to adapt and maintain their participation they had to maintain a level of insight into their own abilities. Therefore it appeared as if this group approached MND pragmatically, focusing on the aspects which were important to them and those which they could potentially influence.

Emergent Theme 5: Making Sense of MND

Some participants spoke about how they made sense of having to live with MND. In most cases faith in God and an acceptance of the disease being "God's will" or a destination were reported. Being grateful for the life experiences they had prior to diagnosis was frequently expressed. Sally, for example, decided to offer her suffering to God as thanksgiving for her life pre-MND: "We have had a good life and I was going to offer any pain or discomfort to God in thanks giving for all" (lines 54-55). Eileen referred to religion as her "backup" (line 1016). Terence admitted that religion helped him to stay positive and cope with his problems. When asked if religion was important to him, Terence stated: "Very. I don't feel down" (1.165-166). He further explained the spiritual benefits of his faith and its importance in maintaining a positive attitude:

> What keeps me happy all the time particularly at night is my faith in Jesus, his father, Holy Mary and all the saints, especially St. Therese of the Roses. I still benefit from my wife and brother and his wife with me on our visit to Lourdes in April (lines 263-267).

Taking part in various research projects was seen as another way of making sense of having to live with MND. Over half of the sample reported taking part in research projects outside of this study.

Emergent Theme 6: Maintaining a Positive Attitude

Some participants reported that maintaining a positive attitude helped them to cope with MND. "Positive is huge" wrote Mike whilst talking about his ways of coping with the disease (line 87). Terence strongly belived that there was no point in worrying about anything which he was unable to change, such as his diagnosis of MND:

> About 20 years ago my daughter got a loan of a book from a friend of hers. I never forget some things in it. One point was never worry about anything you can't do anything about. I always remind myself of it and it works. It [the book] also said a lot about positive thinking, looking on the bright side of life. (lines 297-301)

Although the majority of participants spoke about enjoying their lives despite MND, Bob and Sheila indicated otherwise. Bob appeared to still be in shock post-diagnosis and had not had the time to develop his own coping strategies nor adapt to his changed life. Sheila, on the other hand, had lived with MND for over three years and was becoming exhausted due to her progressive disabilities. She indicated little

enjoyment with her life as she was confined to one room and unable to be independent anymore.

4.1.3 Subordinate Theme 3: Emotions and Feelings

The diagnosis of MND and having to live with this disease evoked many emotions and feelings in the participants. In total, seven emergent themes were identified: (1) Loss and grief, (2) Nervousness and uncertainty, (3) Frustration, (4) Happiness, (5) Hatred, and (6) Diagnosis as a relief .

Emergent Theme 1: Loss and Grief

Loss, and grief over lost skills was reported by all participants. Mike, for example, stated: "I miss talking (…) I miss phone calls" (lines 1137-1139). Sheila became upset when describing her favourite restaurants and hotels. However, grieving emerged as a highly individual experience depending on many factors, such as individual circumstances and coping style. Although all participants with MND named a few skills or activities which had become impossible to carry out due to MND, some participants appeared to have adjusted more to their loss then others.

Emergent Theme 2: Nervousness and Uncertainty

The feeling of uncertainty was particularly strong for the majority of people with MND, and it appeared to be related to the trajectory of the disease and not knowing when the symptoms of MND would progress. Fiona stated: "I don't know how my illness will progress or what I will need down the line" (line 1169). Eileen

indicated that the feeling of nervousness had negatively affected her confidence and prevented her from continuing some of her previous activities.

> I am still nervous. (…) In case something would happen. (…) That you collapse or something... (*voice breaking).* (…) I suppose, knowing that you have Motor Neuron and it could easily happen. (lines 234-241). (…) I got nervous and I still have that nervousness. (…) And I lost my confidence. I couldn't bring myself to go up and read my [readings in the church] and if I had them (…) I'd be totally…(…) Stressed.(…) I was a bit nervous going out on my own. (…) I think it was a generalised thing. (…) that I got nervous of everything. (lines 657-680)

Emergent Theme 3: Frustration

Half of the people with MND in this study reported feeling frustrated with their inability to be as active and involved as they had been prior to their illness. Joseph, for example, stated: "I'm getting frustrated with myself" (line 47). This was reported when Joseph was typing his answers to the researcher. After a while he began to dribble and could not reach for the paper towel himself. Despite having lived with MND for over 6 years Joseph still felt frustrated with regard to his level of dependence, especially when he perceived his mind as being very sharp and unaffected by the disease, in contrast to his body. "I'm sighing because I'm getting frustrated with myself. The message from my brain isn't being transmitted fast enough to my muscles. I guess motor neurone cells are dying" (lines 47-49). It appeared that Joseph's frustration was associated with the huge contrast between his mental and physical abilities.

Emergent Theme 4: Happiness

Despite experiencing many negative emotions and feelings, the participants attempted to maintain a positive attitude and enjoy life in order to help them cope with MND. The general atmosphere during the interviews was positive with many moments of laughter and evidence of the participant's sense of humour , for example, Joseph reported "I consider myself lucky that I haven't lost my eyesight and that my brain is functioning normally. (At least I think so. Ha!)" (lines 325-326). Terence and Fiona were, however, the only participants who reported feeling happy with their current lives in spite of MND. It emerged that both participants had a particularly strong sense of control in relation to managing the symptoms of their disease.

Emergent Theme 5: Hatred

Joseph was the only person who used the word "hate" when speaking about "the medical baggage that goes with mnd" (line 45). He had lived with this disease for over 6 years and his house was full of medical equipment. Joseph reported requiring assistive devices to carry out every activity. He used to "swim or walk" (line 80) in the past, but has now become imprisoned in his own body and his physical independence has been stolen by the disease. Joseph hated the fact that MND had also transformed his house. His bedroom resembled a hospital and the equipment, although necessary to provide support, served as a constant reminder of the impact of MND on the lives of the whole family.

Emergent Theme 6: Diagnosis as a Relief

One participant (Sally) reported that the diagnosis came as relief for her: "It was such a relief to know at last what was wrong with me" (line 30). This was related to Sally's particular circumstances as she had become completely unable to talk or eat about 3 weeks prior to receiving the diagnosis. She could not understand what was happening to her body and desperately attempted to get professional help, however she experienced lengthy delays. Sally was not sure where she should go to seek help. "I was thinking we have the money to go anywhere in the world to get help" (line 32). Before becoming completely unable to swallow and talk she spent months attending various medical appointments and even underwent extensive rehabilitation. Subsequently, her daughter became suspicious that Sally had MND after researching it on the internet, and contacted one of the MND clinics. Sally, therefore, perceived her diagnosis as a relief, an explanation for her symptoms, and a sense of belonging to a group of people living with the same condition.

4.2 Superordinate Theme 2: Living with Dysphagia

This section commences the presentation of findings directly related to dysphagia. Superordinate theme 2: Living with dysphagia is presented by reporting on the two corresponding subordinate themes (Consequences of dysphagia and Self-management techniques) and their relevant emergent themes. Similar to superordinate theme 1, the two subordinate themes were included in creating the superordinate theme if they were prevalent in over half of the sample. It should be acknowledged, however, that when establishing the frequency of themes all participants were considered, even Sally and Sheila who were not eating orally anymore and whose experiences and perceptions were therefore distinctive from the rest of the group. Sally and Sheila provided some retrospective data regarding their physical symptoms of dysphagia, however, they did not report any self-management techniques developed to manage their swallowing impairments. Both ladies, similar to other participants, were immersed in living in the here and now (overarching theme), and therefore maintained their primary focus on non - oral feeding.

Prior to reporting the findings of superordinate theme 2, a table summarising the eating and swallowing status of the participants is presented (Table 4.3) to provide the reader with dysphagia - related background information, in order to provide a context for the results and assist in gaining a deeper understanding of the experiences of this group.

Table 4.3 Dysphagia Related Status of the PwMND during the Data Collection Period

PwMND	Type of diet taken	FOIS*	Gastrostomy tube in situ	Ability to self-feed (orally or non - orally)	Requiring a caregiver at meals	Using specialised utensils for dysphagia
Bob	ORAL	5	NO	YES	NO	NO
Carmel	ORAL & NON - ORAL	3	YES	YES	NO	NO
Eileen	ORAL	5	NO	YES	NO	NO
Fiona	ORAL	5	NO	YES	NO	NO
Joseph	ORAL	5	NO	YES	YES	NO
Mike	ORAL	5	NO	YES	NO	NO
Sally	NON - ORAL	1	YES	NO	YES	N/A
Sheila	NON - ORAL	1	YES	NO	YES	N/A
Ted	ORAL & NON - ORAL	5→4	NO → YES	YES	NO	NO
Terence	ORAL	5	YES	YES	NO	YES

FOIS: Functional Oral Intake Scale (Crary, Mann, & Groher, 2005; Appendix 2)

4.2.1 Subordinate Theme 1: Consequences of Dysphagia

Almost all participants (9/10) reported that experiencing dysphagia led to various consequences, such as changes to diet, perception of food, and social aspects of having a meal. The emotional impact was also reported. In total, seven emergent themes were identified and they are presented in separate sections below: (1) Eating as risk, (2) Changed role of food and diminished pleasure from eating and drinking, (3) Changes in social aspect of meals, (4) Repetitive and limited diet, (5) Inability to eat preferred food , (6) Emotional impact of dysphagia, (7) Changed participation in celebrations.

Emergent Theme 1: Eating as Risk

All participants with MND in this study were aware of being at risk of choking, therefore the majority perceived food as a real risk to their life. Joseph reported being "continuously on my guard. I drink slowly and ensure that solid foods are moist and mashed" (lines 569-570). Sheila reported her previous love for food and reluctance to modify food consistencies as per professional recommendations. After the diagnosis of MND she attempted to eat for as long as she was able to swallow, even after she underwent a gastrostomy and developed severe dysphagia. For some time she managed to eat small amounts orally and supplement this via the gastrostomy tube. This continued until one day when Sheila "believed" (line 208) that food posed a real risk to her life and she decided to cease any oral diet. Having the gastrostomy tube inserted in advance provided Sheila with an alternative – it allowed her to meet the nutritional requirements in a safe and controlled way, but without having to eat orally. The termination of an oral diet was Sheila's own decision, influenced by her own

observation of food being a danger to her life. When she believed that eating became too risky, she stopped it herself. She did not report following any professional recommendations in relation to the cessation of her oral intake. “For safety I stopped eating” (Sheila, line 208).

Ted’s situation was similar. He adhered to a strict and repetitive diet in an attempt to continue his oral intake. When this became too difficult he opted for a gastrostomy, which he perceived as being “okay” (line 108). He also appeared to understand that the continuation of an oral diet would have been too risky. Considering their own eating as a risk seemed to greatly influence participants’ experiences of meals and their approach to food.

Emergent Theme 2: Changed Role of Food and Diminished Pleasure from Eating and Drinking

For the majority of participants the role of food often appeared to transform shortly after their diagnosis of MND when they were informed about the importance of nutrition, and that they should attempt to gain weight. Each participant indicated their own belief that losing weight could be associated with decreased survival. Carmel admitted “not caring about food” prior to MND, but now she believed it “was really important for the muscles” (line 595). Participants began to consider food from a nutritional perspective rather than its taste or appearance. Healthy and nutritious meals made Joseph feel “invigorated” and he ensured that his diet consisted of good quality food. In addition, Joseph reported getting “agitated” if his schedule in relation to meals was not followed. He later referred to food as “fuel” (line 166), which he required to function.

> I can become very agitated if I miss out on main meals. I must have a healthy meal, breakfast, lunchtime and evening dinner. I also need a caffeine boost every two or three hours. I feel invigorated after a healthy meal. (lines 147-150).

The meaning of food seemed to further evolve as the participants developed dysphagia and their eating - related pleasure diminished. Most participants indicated that they experienced little or no enjoyment whilst eating and drinking, which was observed when they shared a meal or a snack with the researcher. Ted described his swallowing as "a bit off putting". (…) "I drank two cups of tea. I didn't enjoy it" (lines 54-57).

In addition, poor appetite and feeling full after only eating small amounts were commonly reported. Mike stated that his food intake became much smaller as he "get full quicker and maybe tiredness is a factor" (line 872). However, despite dysphagia and limited eating - related pleasure, participants strived to ensure that their diet consisted of the appropriate nutrients and calories as they reported having an interest in prolonging their lives. The role of food in MND could be compared to the role of medication in any disease: it is an important contributor to one's health and has to be taken regularly to achieve benefits. The taste and appearance may no longer be worth considering.

Emergent Theme 3: Changes in the Social Aspect of Meals

Changes in the social aspect of eating were reported by the majority of participants. A small number of participants with MND in this study (2/10) avoided eating out as they found the environment too distracting or the food textures could not

be completely predicted which posed a risk of choking. However, most participants decided to continue eating outside of the home environment thus overcoming the symptoms of their dysphagia. Although Terence, for example, described his eating as "messy", he didn't seem to be embarrassed by it or avoid eating with others even though the food "tends to fall out" (line 450). He prioritised the social aspect of eating meals with his family and friends. At the same time Terence was aware of his necessity to implement certain management strategies in order to ensure his safety during meals. Terence was aware that eating alone allows for "concentrating on best way", however, he enjoyed sharing meals with others as he "like[d] company too".

Interviewer: Do you enjoy that [eating with family] or do you prefer to eat on your own?

Terence: Easier to eat (…) on your own, but like company too.

Interviewer: And why do you say it's easier when you are on your own?

Terence: No talking, concentrating on best way. (lines 430-433)

The changes in the social aspects of meals appeared to be associated with participants' attempts to control the symptoms of their own dysphagia, which required an adequate level of concentration. Eating in a noisy environment or maintaining a conversation during meals was therefore reported as being very difficult by most participants.

Emergent Theme 4: Repetitive and Limited Diet

The development of strict routines to manage dysphagia resulted in a limited and repetitive diet, as reported by over half of the sample. This manifested in buying and cooking exactly the same food in order to control the textures. To investigate the

impact of a limited and repetitive diet it seems significant to consider participants' relationship with food prior to their illness to establish how important and enjoyable food was to them then. In this study, all participants declared their previous love of food and pleasure related to eating. This appeared to change during the disease and as dysphagia progressed. For example, when Ted was observed having his dinner he appeared completely immersed in monitoring his chewing and swallowing in order to optimise his safety. He avoided eye contact with others in the kitchen and he chose to eat in the corner, as if hiding himself from others or attempting to limit distractions. There was no sign of pleasure or enjoyment on his face during the meal. Towards the end of the meal Ted was observed taking his medication. There was no change in his facial expression when swallowing tablets either. It appeared that Ted approached his food and medication in a similar manner.

Despite dysphagia causing a limited and repetitive diet for over half of the group, no one complained about their limited food repertoire. The new diet appeared to gradually become part of their lives and become accepted as a consequence of MND and dysphagia. In addition, the changed perception of food (in that it was now seen as important in maintaining the weight of the muscles) is likely to have affected participants' approach. Their diets, although limited and repetitive, were still providing important nutrients necessary for prolonging survival.

Emergent Theme 5: Inability to Eat Preferred Food

Half of the participants in the sample reported excluding their favourite food from their diet to optimise their own safety during meals. Refraining from eating food previously enjoyed seemed to happen almost mechanically and with no reports of any major regrets. This was another change caused by MND; participants perceived no

other choice but to stop eating food which had become too dangerous. The focus was on finding pragmatic solutions to cope with the ongoing progression of the disease. Fiona, for example, substituted the type of bread she liked, as it was "too crumbly", in order to reduce eating - related effort. "I like brown bread but have recently changed to soda bread which I toast lightly and is perfect". *Interviewer: Why was brown bread difficult?* (…) "Crust was too crumbly – it took too long to eat" (lines 355-365).

It emerged that a decision to cease eating a specific food typically followed unsuccessful trials in relation to modifying its texture. As dysphagia progressed the majority of participants (8/10) reported their attempts to adjust the texture of food before excluding it from their diets.

Emergent Theme 6: Emotional Impact of Dysphagia

Half of the participants in the sample reported experiencing negative emotions related to their own swallowing impairment. The cross-case analysis revealed that the main emotion associated with dysphagia was panic and that this panic was predominantly related to choking episodes. When Eileen, for example, described her choking episode she reported "the most ferocious feeling of panic" (line 498) as she was fighting for her life by attempting to catch her breath "because your first reaction is air" (line 526).

> *Interviewer: (…) so you're sitting there eating dinner and what's happening?*
>
> Eileen: And I'm eating away and I'm fine and the next thing I eat a bit of this [broccoli] and it won't swallow. (…).
>
> *Interviewer: So what do you do then?*

Eileen: I went out because your first reaction is air. (…) And I ran out and I was (makes choking sound), you know.

Interviewer: You were trying to cough it up, okay, and was it coming up?

Eileen: It wasn't for a minute or … a few seconds, you know. (…) When it was the first time it happened then needless to say when I wasn't expecting it, you came in and you were, you were in shock and I suppose nervous. (lines 519-542)

In the above quotation Eileen initially reported being unable to cough up the food which was blocking her airway "for a minute" and then she rationalised "or a few seconds". Her first impression ("a minute") may be related to her own feelings and perceptions, for her the choking seemed to last that long. Moreover, the panic caused by choking decreased Eileen's trust in food in general. After her second choking episode she decided to only consume self-cooked food and eliminate all meals prepared by another person from her diet. She began perceiving food as very risky and aimed to carefully control the textures of food to minimise the likelihood of choking.

Emergent Theme 7: Changed Participation in Celebrations

Changed participation in celebrations was reported by Sally, who experienced severe dysphagia and was unable to eat orally. Sally recalled her 80th birthday party and her inability to eat any of the food there: "I was 80 last week. All the children came and we were all together. Lidia [daughter] had a nice meal just the … but I could not eat. Or have a glass of champagne" (lines 125-127). Sally was still very involved in cooking for her husband and she was observed preparing five Christmas puddings when the researcher met her in

early November. She wished to maintain her previous routines, but despite her engagement in the preparations for Christmas celebrations it "all changed" for her. *Interviewer: "Can I ask you about Christmas? What are your plans?"* Sally: "Lidia will come over in the morning. I will have some dinner that evening. We always had Christmas over there [in her daughter's house] so it has all changed" (lines 123-124) [shrugged her shoulders and pointed to her mouth indicating her inability to eat]. Sally referred to having "some dinner" for Christmas which when considered within the context of her whole interview indicated that her attitude towards food had changed. Sally's phrase "some dinner" (referring to Christmas dinner) conveyed no emotion. It emerged that as Sally was unable to even taste any food, she had begun to emotionally detach from it knowing that eating and drinking will not ever be part of her life.

Sally was the only participant who referred to the impact of dysphagia on one's ability to participate in traditional / cultural cerebrations. This issue may be potentially relevant to other people with dysphagia, as the celebrations of special occasions, such as birthdays or Christmas, other than those involving food and drink, appear to be limited in Irish culture today.

4.2.2 Subordinate Theme 2: Self-management Techniques

This section focuses on presenting the techniques used, which participants reported as self-developed, in order to manage their dysphagia. The subordinate theme: Developing own techniques to manage dysphagia is reported using the five corresponding emergent themes as identified during data analysis: (1) Noticing physical symptoms of dysphagia, (2) Paying special attention during meals and

modifying the eating environment, (3) Changed preparation of food, (4) Excluding risky food, and (5) Eating routines. The development of dysphagia management strategies appeared to relate to the overarching theme: Living in the here and now. As the participants were immersed in the present, they focused on solving the problems experienced in the current moment. If people with MND in this study noticed a problem related to dysphagia, they attempted to quickly solve it themselves in order to preserve their feeling of being in the here and now and maintain their usual level of participation and engagement. Although their bodies were failing, the participants seemed to cognitively solve their difficulties for as long as this was possible. In this study, the participants reported being actively engaged in "experimenting" with various dysphagia related strategies in order to "discover" the most suitable one. It emerged that this was their primary method of managing dysphagia, as opposed to following professional recommendations.

Emergent Theme 1: Noticing Physical Symptoms of Dysphagia

All participants reported experiencing physical changes in their swallowing ability such as chewing problems, inability to manage specific types of food, choking, aspiration, and prolonged meals.

Deterioration in the ability to chew food was reported by the majority of participants. Interestingly, when describing their chewing difficulties half of the group provided information specifically related to the functioning of their jaw. Mike, for example, indicated that he had great difficulty with moving his jaw up and down which he described as "terrible" (line 921). Sally reported not being aware of her open mouth posture. Fiona compared her jaw to a "rat trap" and described significant issues with

her ability to move the jaw: “Imagine unlocking something that is closed. You probably have never seen an old rat trap” (lines 89-90). If the jaw represents a door leading to the mouth, the opening of the jaw marks the beginning of the eating and drinking process as it must open for the food or liquids to enter the oral cavity. Fiona, however, compared her jaw to a trap suggesting that the jaw’s movements may have been unpredictable, uncoordinated, rapid, and dangerous. Once her jaw was closed it became hard to open. Comparing parts of her own body to a trap could indicate Fiona’s limited trust in the functioning of her own body as a result of living with a progressive disease of unpredictable trajectory.

An inability to manage specific types of food was reported by all participants with the exception of Bob. When Fiona, for example, stated that “grapes run around in my mouth” (line 996) she not only indicated that grapes were hard to manage, but also that she no longer had the ability to control this type of food. All participants (apart from Bob) identified a number of foods which had been perceived as too difficult to eat. Bob did not report any difficulties in this regard, which could be related to him having the mildest level of dysphagia within the group. Moreover, Bob’s primary concern was his respiratory issues and he was focused on his own breathing ability and therefore did not perceive his dysphagia to be troublesome.

The majority of participants (8/10) reported experiencing choking and aspiration. Joseph indicated that he was “very conscious” of being at risk of food “going the wrong way” and also that choking can be an unpredictable event. “I'm very conscious that food may go down the wrong way or get stuck in my digestive tract” (line 177). (…) “I guess nobody can foresee when a choking episode will occur” (line 464). Terence also reported experiencing difficulties with the pharyngeal state of

swallowing, which sometimes resulted in him having "to leave the table to settle down":

> What I mean is the feel that the drink has gone down wrong way. I start coughing. The minor one is that I don't have to leave the table to settle down. I try to take a tiny sip to free. This works usually. I rarely have to leave the table. The biggest problem would be food but that hasn't happened for a long time. (lines 715-730)

Interestingly, even people who were not eating orally anymore (Sheila and Sally) still recalled and reported their choking episodes. This may be an indication of the long lasting trauma associated with experiencing choking whilst eating food.

Perceived slowness during meals may be the result of, for example, poor functioning of the oro-motor muscles leading to increased effort to masticate food or the diminished ability to control these muscles. In addition, participants were attempting to be careful during meals in order to minimise the risk of aspiration and choking. Carmel, for example, reported: "I am slower eater. (…) All my family was finish and I was still eating". *Interviewer: Why do you think it was slow? What was the problem?* "I was afraid in case I was choking." (lines 288-310)

Participants' noticing the physical symptoms of dysphagia appeared to be followed by attempts to self-develop strategies to cope with their swallowing impairment. Cross-case data analysis revealed the four most common techniques reported as being self-created by the participants in order to manage dysphagia: (1) modifying consistency of food, (2) substituting risky foods with softer textures, (3) monitoring the pace of eating and drinking, and (4) being careful and focused while eating.

Emergent Theme 2: Paying Special Attention during Meals and Modifying Eating Environment

All participants reported having to pay increased attention whilst eating, drinking, and swallowing. Mike, for example, reported having to be "careful. (…) About bite size". (lines 161-163). Fiona stated that being careful during meals improved her ability to swallow and ultimately her safety: "Sometimes I cough when I am not paying attention to drinking. (…) If I am careful it is okay" (lines 907-909). Terence reported that he "can manage [his dysphagia] slow and careful" (lines 78-79). Being "continuously on my guard" whilst eating, as reported by Joseph (line 569) appeared to eliminate the spontaneity of eating as a constant mental effort was required. This was observed to have a negative impact on eating - related pleasure.

Having to be paying increased attention during meals impacted on the participants' eating environment. Some people with MND in this study avoided eating outside of their home to maintain their safety during meals. A more frequent modification to the eating environment, however, was limiting distractions during meals. Carmel, for example, was aware that maintaining her full concentration during meals was important and reported switching off her phone during meals. In addition, she was observed to reduce the volume of the TV while eating.

Interviewer: How is that happening [coughing during meals]?

Carmel: If I eat too fast. (…) Or if I am texting...

Interviewer: Are you saying if you're distracted from eating it can cause you to cough?

Carmel: Yeah. (Writing). I turn off my phone when eating (lines 460-475).

Minimising one risk can however contribute to maximising another. Carmel had no family and was on her own during the days when the other tenants were at work. She reported that the phone was an important tool for her to get help if required (by texting a friend who lived nearby). At the time of her participation in this research Carmel did not have an alarm system in place and shouting or phoning for help would not be possible due to her dysarthria. Switching off the phone during meals therefore reduced her chances of getting help in the event of experiencing any difficulty. However, Carmel similar to other participants, was focused on the here and now (overarching theme) and avoided thinking about any potential consequences of switching off her phone.

Emergent Theme 3: Changed Preparation of Food

As the participants began to observe physical difficulties with their swallowing, all except Bob reported that they began modifying food textures in order to make them easier to manage. This involved, for example, cooking food for longer to make it very soft, cutting and chopping food into small pieces, mashing it with a fork, and using electronic devices for food preparation, such as a mincer or blender. Eileen, for example, started to prepare her toast in a different way in order to minimise her risk of choking: "If I have toast I put lots of butter on it when it's hot. To soften it" (line 145). Terence also reported implementing various techniques to control the texture of food:

> I remove the peel when making the marmalade. For lunch I have mince beef, chicken, fish or other chopped up small. I have mash potatoes and some vegetables. Sometimes I make a stew with mince, potatoes and veg. That would be best (lines 8-11).

Changes in the preparation of food appeared to be quite subtle initially and it emerged that once a problem with a particular type of food was identified the participants gradually began to modify the texture of this problematic food. As dysphagia progressed, however, texture modifications were not always sufficient to ensure safety during swallowing and the particular type of food may have to be excluded from the diet.

Emergent Theme 4: Excluding Risky Food

As the participants learnt that some foods were no longer suitable for them (too hard to swallow) and that there were no more strategies available which could enable safe swallowing, the process of elimination of risky foods began. This was reported by all participants with the exception of Bob. Typically, harder textures were substituted with softer consistencies as these were easier to manage. The participants aimed to minimise their risk of choking and ease the effort related to chewing. Terence, for example, spoke about the changes in his diet which had occurred over the six months preceding the interview:

> *Interviewer: Terence, if we were to go back in the past few months, let's say 6 months… Has anything in the way you eat changed for you?*
>
> Terence: Changed diet.
>
> *Interviewer: Can you tell me a bit more about the diet? What has changed?*
>
> Terence: Porridge instead of muesli. Soft dinners, minced meat.
>
> *Interviewer: Instead of what? What would you have before that?*
>
> Terence: Bacon. Beef. (lines 374-380)

As the process of excluding unsuitable foods from the diet was gradual, the participants appeared able to adjust to this change. It seemed to almost be a natural consequence of living with progressive dysphagia. Participants who continued to have an oral diet did not report any regrets in relation to their inability to manage specific types of food. However, people who were eating non - orally frequently expressed their regrets with regard to no longer being able to eat their favourite food.

Emergent Theme 5: Eating, Drinking, and Swallowing Routines

The majority of people with MND in this study reported the self-development of specific routines to help them manage their swallowing impairments and also to save on their food preparation time. This included, for example, buying only specific brands of food as its consistency was always predictable. Some participants developed a rigid schedule of meals and ate exactly the same foods every day. Ted, for example, managed his dysphagia by eating "the same meals everyday" (lines 78-83) as the texture was always predictable, therefore it was easier to self-monitor his swallow in an attempt to reduce the choking risk. Every day he ate an egg for breakfast, a ready-made soup for lunch, and a ready-made cottage pie or lasagne for dinner (all ready-made meals were by a particular brand). This also minimised the demand of cooking and made his shopping experience more straightforward.

Some self-developed eating, drinking, and swallowing routines resembled common professional recommendations, whilst others were quite individualised and unique, for example, in relation to swallowing tablets. Each participant, who experienced problems with swallowing tablets, developed an individual strategy to

overcome this difficulty. Eileen "put the tablet into her mouth first, then she took some food and swallowed it all together with liquid" (lines 483-485). Fiona had to place a tablet "at the rear of her tongue and swallow it with water" (lines 292-300). Terence swallowed his tablets "with porridge or yoghurt" (line 657). Ted used "only food" to swallow tablets (line 87), whereas Carmel "mixed" tablets with her nutritional "supplements" (lines 520-527).

Participants rarely quoted any professional recommendations received to manage dysphagia. Instead, they seemed to be fully focused on developing self-management techniques almost as if they were attempting to hide their problems from others, perhaps in order to minimise the burden on them. This approach seemed to continue until the progression of MND was too severe to allow for the continuation of self - management and professional help became the only alternative.

It also emerged that participants were generally surprised by the number of strategies they have developed to manage dysphagia, for example, Eileen said: "All these things I do myself" (line 575) realising how much she was actually doing to compensate for her dysphagia. These moments during the interviews, when participants' became aware of the ways in which they were coping with their dysphagia, appeared to be positive for each of them. Being able to self-manage dysphagia meant that the people with MND in this study perceived having some control over their disease.

4.3 Superordinate Theme 3: Professional Management of Dysphagia

This section focuses on the presentation of superordinate theme 3 and corresponding subordinate themes identified in the data. Similar to the presentation of

the findings of superordinate theme 2, subordinate themes are further divided into sub-sections corresponding to their emergent themes. Superordinate theme 3 is: Professional management of dysphagia and it presents a summary of participants' experiences regarding dysphagia - related professional services. These experiences were divided into two subordinate themes: 1) Recommendations to manage dysphagia and 2) Service provision.

4.3.1 Subordinate Theme 1: Recommendations to Manage Dysphagia

Subordinate theme 4.2.2 presented strategies reported as self-developed by the participants in order to manage the symptoms of their dysphagia. In contrast, this subordinate theme focuses on dysphagia - related strategies, which were reported as professionally recommended. There were three emergent themes identified: (1) Non - oral feeding, (2) Modification of fluid consistency or textures of food, (3) and Adherence and adjustment. These emergent themes are presented below.

Emergent Theme 1: Non - oral Feeding

Participant's experiences related to non - oral feeding appeared particularly complex to unravel. In order to acknowledge particular aspects of the participants' experiences in relation to non - oral feeding, this emergent theme is further divided into three main topics (sub-themes): (1) Understanding of non - oral feeding, (2) Decision-making process in relation to non - oral feeding, and (3) Living with non - oral feeing at home.

Topic 1: Understanding of non - oral feeding. Non - oral feeding was generally understood as a source of additional nutrition and hydration which would be required in the event of severe dysphagia. All participants believed that non - oral feeding could prolong survival. However, despite the fact that the entire group presented with a similar perception of the role and benefits of non - oral feeding, the decision-making process regarding whether or not to accept it appeared to be quite individualised.

In addition, it appeared that some participants perceived that they did not have a sufficient or adequate level of knowledge prior to undergoing gastrostomy. Carmel, for example, reported: "I thought it [gastrostomy tube] was up there (pointing to her throat). (…) I thought how am I going to go home?" (lines 410-413). Sheila was convinced that insertion of a gastrostomy tube automatically prevented the continuation of any oral intake, therefore she reported delaying the procedure for as long as she could. Terence reported not being aware of any potential complications related to the gastrostomy and was shocked when he had to stay in hospital for a few days and experienced pain and discomfort following the procedure.

Topic 2: Decision-making process in relation to non - oral feeding. Some participants (Carmel, Sally, and Ted) opted for non - oral feeding relatively quickly and underwent gastrostomy within a few weeks of the initial recommendation being made. Others (Mike, Sheila, and Terence) required more time and reported that their decision-making process was very complex. Sally, for example, received the professional recommendation to commence non - oral feeding when she had already lost the ability to swallow orally and had been unable to consume anything for a couple of weeks. She perceived there to be no alternative and reported that non - oral feeding had saved her life. In contrast, Sheila was still able to swallow when non - oral feeding

was recommended to her. She was reluctant to consent to it as she was still managing an oral diet. Although Sheila was advised to undergo the gastrostomy insertion before it became a necessity (while she was still able to eat orally and while her respiratory status was still good), she required some time and encouragement from her family to finally consent to non - oral feeding. The decision-making process for Sally and Sheila differed as their swallowing status varied considerably. However, the above differences could also be explained from the perspective of the overarching theme: Living in the here and now. Sally required an alternative eating source in her here and now whereas Sheila was still managing her oral diet, therefore having to plan for dysphagia management in advance was more distressing and difficult for her. Similar to Sheila, most participants reported being advised to consider non - oral feeding early in the course of dysphagia and while their respiratory status was relatively good. This complicated the decision for the people with MND in this study, who were immersed in their here and now. Although 9/10 participants had received a recommendation of non - oral feeding from a professional, only half had a gastrostomy tube in situ during this research. Bob, Eileen, and Joseph did not perceive any need for non - oral feeding as, in their opinion, they were managing well without it. Mike was in the process of deciding about non - oral feeding during the data collection phase.

It emerged that the decision-making process in relation to non - oral feeding was also influenced by the presence of a caregiver. The participants who looked after themselves (Carmel and Ted) opted for non - oral feeding quicker in comparison to participants who lived with their family caregiver (although Ted lived with his children and grandchildren, he reported that they had not been involved in his care as he wished to be independent). Carmel and Ted perceived non - oral feeding as a solution which would prolong their independence. Both reported that they experienced

increased energy levels due to non - oral feeding and found this route of hydration and nutrition to be convenient.

Mike's decision-making process with regard to non - oral feeding was particularly difficult and lengthy in comparison to that of the other participants. Mike's experience is presented in greater detail in order to highlight the complexity of his situation. Although non - oral feeding was recommended by a team of professionals who were providing Mike with regular care, he also contacted a person with MND who had a gastrostomy tube in situ in order to gain more insight into living with non - oral feeding at home. Mike reported that the person with MND he contacted was very pleased with his decision to undergo a gastrostomy. It emerged, that despite a number of professional consultations regarding non - oral feeding, Mike perceived that he had "very little" knowledge about it (line 342). Although Mike appeared to have a good understanding of the medical rationale for undergoing a gastrostomy, he did not consider what he had "been told about it [non - oral feeding]" (line 342) sufficient for him to make a decision. He was also concerned about the surgery as he reported that "everything involving operation is difficult" (line 378), especially considering he was aware of his deteriorating medical status and the possibility of complications. Mike's decision-making process was also influenced by his focus on the here and now (overarching theme). At the time of data collection he described his swallow as being "okay" (line 174). He understood non - oral feeding as being an alternative source of nutrition and hydration and he was aware of the importance of appropriate nutrition in MND. However, Mike was taking nutritional supplements (and said that he "loved them", line 774), and reported no issues with hydration, therefore he did not seem convinced with regard to needing to commence non - oral feeding in the near future. Additionally, thoughts about the gastrostomy tube triggered negative emotions related

to Mike's past experiences. Mike had cared for his father who had, in the course of his illness, needed various types of tubes to be attached to his body and having to deal with these tubes was reported as having been very difficult for Mike. Another factor contributing to Mike's difficulty with making a final decision about accepting non - oral feeding was that someone in his family had died of MND in the recent past. That person had refused non - oral feeding for perceived quality of life reasons. During interviews, Mike appeared not to wish to talk about that person in any great detail as he seemed to be deeply affected by this experience.

Mike's experience regarding the consideration of non - oral feeding appeared distinctive within the group. He was the youngest participant and the only one who had had a family member with MND and dysphagia. He also seemed to have the most advanced understanding of the swallowing mechanism and dysphagia amongst the participants. Although Mike was aware of the general benefits of non - oral feeding from a medical perspective, he was still unconvinced regarding his own desire to accept non - oral feeding. Perhaps he perceived undergoing a gastrostomy as letting the MND take even more control over his body and take away another part of his life. Mike's main goal was to maintain his independence and continue his participation in family routines despite MND, including eating together. Even though Mike understood the nature of his progressive disease, he attempted to delay admitting defeat for as long as possible. Considering Mike's previous experiences with his father and relative with MND he may have perceived non - oral feeding as bringing him closer to death. Mike reported his wish to engage in the decision-making process in relation to non - oral feeding alone. He did not wish for his family to participate in this decision.

After nine months of consideration, Mike consented to non - oral feeding. He had to, however, wait for almost two months to receive an appointment to see a specialist in relation to gastrostomy. When Mike finally met the doctor concerned he was advised to re-consider his decision, as in the doctor's opinion gastrostomy was "being pushed" on the people with MND. Mike reported that this appointment was "very hard", "confusing, and disappointing" for him, especially as he had not anticipated any delays or complications with undergoing the gastrostomy (lines 1066). Moreover, non - oral feeding was previously recommended by a team of professionals who had known Mike since his diagnosis of MND and he trusted their professional opinion. Mike was confused to hear a different opinion from a doctor, especially when this person had not been previously involved in his care.

Mike: I was hearing one side up to now.

(…) Interviewer: Okay. Tell me more please.

Mike: Em… In general he [gastroenterologist] felt that the PEG was being pushed… in MND. (lines 485-487)

(…) He felt MND [team] think the PEG (clicks fingers)… are pushing the peg.

Interviewer: What did you think of that?

Mike: Yeah (shrug his shoulders). Very hard. (…) I still am not sure.

Interviewer: Are you not sure whether to get it [gastrostomy] or not?

Mike: Hmm… [indicated yes]. Confused. Disappointed. (lines 501-510)

Following the face-to-face interviews Mike emailed the researcher and reported: "I have reflected on procedure and (…) I wish to go ahead with operation"

(1255-1256). It seemed that the last consultation with the gastroenterologist had not, in fact, ultimately changed Mike's final decision, although it had delayed the procedure and contributed to an increased level of stress for Mike.

Topic 3: Living with non - oral feeding at home. As previously mentioned, five participants had a gastrostomy tube in situ at the time of this research: Carmel, Sally, Sheila, Ted, and Terence. Their experiences of living with non - oral feeding varied. Carmel and Ted (who both reported managing with MND independently) considered non - oral feeding as a convenient alternative to an oral diet, as it reduced the pressure of having to consume adequate nutrients in an attempt to stabilise their weight. Carmel reported: "I find it very convenient. (…) I am out and back and the food is there for me" (lines 534-540).

For Sally and Sheila non - oral feeding contributed to a significant sense of lost independence and a deterioration in quality of life. Sheila's mobility had greatly reduced at the time of her gastrostomy perhaps influencing her perception regarding the impact of non - oral feeing on her life. Sally reported issues with regard to establishing an adequate regime of non - oral feeds. A 'trial and error' approach appeared to have been implemented whereby various types of feeds were offered to see how Sally's digestive system would react. Ultimately, Sally was only able to tolerate a very slow rate of feeding which necessitated that she be connected to the feeding pump for 16 hours per day. Sally required her husband's support to manage the "food thing" [feeding tube] (line 67) which for her meant a huge loss of independence.

> *Interviewer: "If I follow you through your typical day what would I see you doing from the moment you wake up?*

Sally: Liam [husband] takes out the food thing and I have a shower… Liam cleans the RIG and I come out and sit for the rest of the food to finish.

Interviewer: Okay. And I understand the feeding takes a long time now…

Sally: Hmm (yes). 16 hrs (lines 65-70). (…) Well we have to get the feed right I went to the toilet 10 times one day. (…) But we are working on it now." (lines 108-113)

When the researcher arrived for a scheduled appointment with Sally one morning, no one was at home. There had been an emergency as the gastrostomy tube had become loose and Sally and her husband had to seek medical attention immediately. This happened when Sally was hoovering as bending had caused additional pressure on the gastrostomy tube. When Sally returned from the doctor she reported: "I broke the tube" and she appeared to feel guilty (line 5). Sally attempted to balance her desire to maintain her previous routines with having to be mindful of the gastrostomy tube. As Sally was confined to the house due to the prolonged feeding time she was not able to participate in her routines outside the house anymore. Therefore, engaging in the household duties was very important to Sally.

Sheila appeared to adjust to non - oral feeding over time and reported that the gastrostomy tube was keeping her alive, allowing her to be with her family, and watch her grandchildren grow. When a feed was being pumped into Sheila's stomach she almost appeared not to notice it. Even when the alarm indicating the end of the feed sounded, she did not react. Perhaps her mind detached from her failing body.

Terence's experiences of non - oral feeding were quite distinctive as he deeply regretted undergoing the gastrostomy insertion. For him, the gastrostomy tube had brought too many complications, both physical and psychological. As a result, Terence

refused to ever commence non - oral feeding even when his MND and dysphagia progressed significantly and he developed great difficulties with eating and drinking orally.

Terence described his experiences of gastrostomy tube insertion as "a mess" and was unconvinced that his decision was right for him.

> Terence: I thought it [non - oral feeding] made sense then but I am not so sure now (lines108-113).
>
> *Interviewer: So how was it? How did you get the tube inserted?*
>
> Terence: Very bad experience. (…) I was supposed to be in for one night. I was in for six nights. Bad pain, sleeping on chair for three nights (lines 89-94). (…) I have the tube inserted June 25th. I was to be let out that evening but had to stay six nights, three sitting on chair with pain. (…)
>
> *Interviewer: (...) And if you could change anything [in the health services received] what would you ask for? Or would you change anything?*
>
> Terence: The operation for tube was a mess. (lines 476-477)

It emerged that Terence perceived the moment of gastrostomy insertion as his personal diagnosis of MND which marked a monumental and unanticipated change in life for him. When asked about his energy levels Terence reported: "Before the stomach tube operation in early July I would say I was like that … with more energy and able to do more outside. Since then it has changed a lot" (lines 208-210). Terence frequently pointed to his stomach during the interviews and (using facial expressions) indicated his dissatisfaction with gastrostomy. It appeared that the gastrostomy tube served as a constant reminder of his illness as despite not using it as a source of

hydration or nutrition, he had to flush it daily. Having to clean the tube every day could have provoked Terence to dwell on the upcoming future and disturb his attempts to live in the here and now. Terence divided his experiences of MND into pre - and post - gastrostomy tube insertion. "Before that [PEG tube insertion] I would climb over gates. Now I am very aware of danger of falling" (lines 214-215). In this quotation Terence indicated that undergoing the gastrostomy made him aware of the risks associated with MND and diminished his confidence. Prior to gastrostomy tube insertion Terence used to "climb over gates", he believed in his abilities and strength despite his medical diagnosis of MND. Following the gastrostomy tube insertion Terence not only became aware of the risks associated with MND, but also noted a physical deterioration in his body. Moreover, he reported having been inadequately prepared for the gastrostomy procedure as well as for the reality of life with a gastrostomy tube.

Emergent Theme 2: Modification of Fluid Consistency and Textures of Food

All participants reported that they had received recommendations from a SLT in relation to modifying the textures of food or consistency of fluids to manage dysphagia. However, not all participants chose to follow these professional recommendations, as the majority reported decreased enjoyment from eating or drinking when the consistencies had been modified. Ted, for example, stated that the thickener was "fine in hot drinks, but unbearable in cold drinks. Doesn't mix. No taste" (lines 61-62) and that it was "awkward" and "off putting" (lines 63-64). Mike admitted to not using the thickener at home and reported two reasons for his decision. First of all, Mike perceived the change in fluid consistency to be too significant to be tolerated. He described the thickener as "not nice" and that his cup of tea tasted "terrible" after

it had been thickened (line 442). Secondly, he appeared not to be convinced that he required the thickener, as in his opinion he was able to manage well without it.

It emerged that Terence found using the thickener difficult due to his limited fine motor skills. He reported: "I had a problem using the measure in container. I have changed now to using an ordinary spoon. The mixing can be a bit messy but I'll get used to it" (lines 700-702). In addition, Terence highlighted his difficulties with following instructions regarding the recommended usage of the thickener, as what was written on the tin was reported to be different to professional recommendations.

Some participants noticed health benefits associated with taking fluids of thickened consistency. Terence reported: "My daughter Alice said she was told that it [the thickener] should be used all the time. I agree with that because I found more blocking when only using it once or twice a day" (lines 695-697). Ted admitted: "I cough less with the thickener. But I dislike the taste" (line 67). During this research Ted underwent a gastrostomy and commenced non - oral feeding. When reflecting on his experiences Ted reported preferring non - oral feeding over thickened fluids as the former was easier to manage and more pleasant for him.

Emergent Theme 3: Adherence and Adjustment

Participants' adherence to dysphagia - related professional recommendations was not the focus of this study and therefore checking participants' reports against professional records was not attempted. However, the issue of adherence was voluntarily raised by all participants in relation to dysphagia management. It emerged that the people with MND in this study refused to follow professional recommendations if they considered the recommendations to be unjustified,

unnecessary, or too disruptive. In addition, it emerged that even a relatively minor professional recommendation may be perceived as major by a person with MND. When a SLT asked Terence, for example, to limit his consumption of bread he immediately "was thinking of breaking it up and eating with spoon" (line 336) as he "wouldn't like to rule them [the bread] out yet" (line 622). This quotation illustrated Terence's reluctance to refrain from eating bread and also his focus on developing his own solutions to manage dysphagia. Although Terence indicated his understanding of the rationale for limiting the consumption of bread, he strived not to rule out this food. He appeared aware that modifying the consistency of bread may allow him to eat it for a longer period.

The majority of participants reported that in order to follow a professional recommendation to manage dysphagia they required an adequate length of time to consider it. Sheila highlighted that she required time to believe that a particular recommendation was justified and appropriate for her, indicating that professional advice alone was not sufficient for her. Instead, she had to "believe" in the necessity of modifying her current way of eating and drinking in that the symptoms of dysphagia had to appear problematic in Sheila's here and now.

Interviewer: How did you feel about the changes in the diet?

Sheila: I didn't like it at all. No. Takes time.

Interviewer: Would you say it takes time to adjust to changes of food?

Sheila: You have to process it in your head. And believe that the danger was there. (…) For safety I stopped eating. (lines 203-208)

As the process of adjusting to dysphagia - related professional recommendations emerged as being complex, an in - depth analysis of data obtained

from Terence's transcript is presented below as an example of how a person with MND may approach a professional recommendation received to manage dysphagia. Similar to other participants in this study, when Terence was advised by his SLT to commence thickened liquids he did not follow this recommendation immediately. Instead, Terence engaged in a multi - steps process of consideration (Figure 4.2) during which he transformed a professional recommendation into one of his own making. This personalisation of a professional recommendation occurred as Terence "experimented" with it in an attempt to find the most suitable way of implementing it at home. By the end of this process Terence had "re-discovered" the recommendation but he no longer perceived it as having come from a professional and instead viewed it as having been developed by him.

Figure 4.2 Terence's Approach to a Professional Recommendation to Thicken Fluids

1. Willingness
   - "I might try it"
   - "I have tried the thickener but I don't use it with every drink. I use it at least twice a day"
   - "I would hope to get myself to use it more"
2. Experimenting
   - "I have been experimenting with the thickener for the past week"
3. Difficulties
   - "I had problem using the measure in container"
   - "A little bit of work in stirring it and having to put it into a cup before the liquid"
   - "I tend to use it more when eating on my own"
4. Problem solving
   - "I have changed now to using an ordinary spoon"
   - "Much easier to add the thickener to the drink"
5. Family support
   - "My daughter said she was told that it should be used all the time"
6. Noticing changes
   - "It is definitely true that if you are using the thickener most times (you are) more likely to have problem when you don't use it"
7. Seeing benefits
   - "I found more blocking when only using it once or twice a day"
8. Discovering own ways
   - "I find it helps the swallowing but only if you follow the same rules. Keep slow, bend down the chin and separate foods".

4.3.2 Subordinate Theme 2: Service Provision

Although all participants in this study attended dysphagia - related services, achieving rich descriptions of their experiences was difficult as the people with MND in this study were reluctant to talk about the health services in any great detail (despite encouragement from the researcher). They tended instead to provide a general opinion of the services, such as describing them as being "okay" or "fine". It was observed that the participants much preferred to report their personal experiences of managing dysphagia at home, rather than recalling the professional input obtained.

Subordinate Theme 2: Service Provision consisted of three emergent themes: (1) General satisfaction, (2) Associated emotions, and (3) Suggestions for change.

Emergent Theme 1: General Satisfaction

All participants reported their general satisfaction with the services received for dysphagia. Eileen, for example, when asked about her experiences of dysphagia related services reported: "Oh A1! I couldn't say enough about them. It's marvellous. I think it is brilliant the back up. (…) Absolutely brilliant. I couldn't fault a thing. (…) I feel supported a hundred percent. (…) And I see no reason to change anything, you know" (lines 385-396). Bob stated: "Everybody has been very great (line 424)". When Ted was asked: "What do you think about the services you received for your swallowing?" He wrote: "OK – for a swallowing" (line 117) indicating his satisfactions with the services received for dysphagia, but also suggesting his possible dissatisfaction with other services.

However, despite being generally satisfied with the services received for dysphagia, six participants also reported some negative experiences. Some participants expressed their suggestions for change in relation to how dysphagia - related services were organised. This data is presented in the subsections below.

Emergent Theme 2: Associated Emotions

Attending professional appointments was reported as being stressful by the majority of participants. This appeared to be related to the nature of MND, which meant that progression of the disease was imminent. There was a general feeling of anxiety related to finding out how the disease had progressed. In addition, some participants (Bob, Mike, Sheila, and Ted) felt overwhelmed with the amount of information given by the professionals. Mike, for example, reported: "With regards to the visit to the gastroenterologist I was given a lot of information about the subject by Dr [name]. A huge amount to take in on the first appointment" (lines 23-25). Mike also admitted feeling uncomfortable being observed while eating as part of his dysphagia review carried out be a SLT.

The majority of participants reported that they had attended MND clinics. They described the MND Clinics as involving a team of professionals specialising in MND, who provided a service to people with MND from diagnosis throughout the disease. The participants typically saw a few members of the multidisciplinary team on the same visit and had to move between the various rooms until all consultations had been completed. Feeling overwhelmed and uncomfortable when attending MND clinics was reported by half the group. Moreover, these participants reported anticipatory stress and fear, as they were anxious to hear about the progression of the disease.

Eileen, for example, reported "dreading it": "especially when the whole group are in front of you" and she "dread it [MND Clinic] before you go" (line 508).

Another issue related to attending MND clinics was the guaranteed meeting of other people with MND at various stages of the disease. This could be understood from the perspective of the over - arching theme: Living in the here and now, as seeing people at different stages of the disease would be disturbing for someone who is aiming to live in the present. When asked about the MND clinics Sheila reported: "I don't like it". When probed by the interviewer *("Can you tell me more?)* she answered: "The team are lovely". When further encouraged to provide more information *("There is something you don't like and I would like to know what)"*, she admitted: "Looking at other people. Different stages" (lines 183-188).

One participant (Terence) reported feeling "nice" after a consultation with a SLT, as he was given "good news" that his swallowing ability was "not bad" (Terence, line 332). Terence reported hearing some positive feedback to be significant for him, especially that he was aware of the progressive nature of MND. It emerged that the majority of participants perceived the attitude of the professionals to be important to them. The majority of people with MND in this study reported their desire to feel respected and listened to by the professionals involved in their care. This appeared to be a particularly sensitive issue for non - verbal participants, who reported feeling ignored by the professionals when they talked to their caregivers rather than to them.

Having a positive rapport with a professional was reported as being very valued by the majority of participants. Moreover, participants appeared to engage in the service more if they liked the professional involved in their care, and they avoided consultations with professionals perceived as incompetent or disrespectful. The

people with MND in this study wished to have positive experiences when attending dysphagia services and valued the professionals who were kind and respectful. When asked how the service related to swallowing was Sheila replied: "It was good. (…) Depends on the person." *Interviewer: What do you think made Monica [SLT] special?* "She was at ease and made me comfortable. She was listening to me. (…) She gave advice through different stages. Didn't force" (lines 217-255). Sheila was non - verbal and used a wheelchair. Her dysphagia had progressed, making swallowing impossible. She had been engaged in dysphagia services for over three years. She knew there was no treatment to stop her dysphagia from progressing and she had gradually accepted non - oral feeding. Sheila spoke about Monica [SLT] a few times and she always smiled when recalling her appointments with Monica.

Emergent Theme 3: Suggestions for Change

Some participants (Fiona, Sally, and Sheila) reported being seen by professionals who, in their opinion, did not possess adequate knowledge or experience in relation to MND. This appeared to cause additional strain, particularly considering that participants perceived attending medical appointments as being generally stressful. Fiona, for example, became anxious when recalling meeting a professional, who was unfamiliar with her type of MND: "When I went to speech and language first she never heard of it [bulbar onset of MND]" (426-427). Fiona viewed attending appointments with inexperienced professionals to not only be a waste of her time, but also to be disrespectful to her.

Joseph appeared unhappy about having to self - refer for dysphagia reviews as he did not trust his "self - analysis". Instead, he would have preferred if the frequency

of reviews had been dictated by professional recommendations. Joseph reported: "Community Speech Therapist texts me every few months. She decides whether or not to visit me based on my self-analysis (lines 453-454). (…) "It's not for me to decide how often my swallow should be tested" (line 461).

Fiona's suggestion related to her dissatisfaction with the communication between professionals involved in her care. Fiona reported that she viewed adequate communication between professionals as an opportunity for them to be appropriately prepared for consultations.

> *Interviewer: Would you have any suggestion for change [in regard to professional services received for dysphagia]?*
>
> Fiona: I think when the [names organisation] or [names a hospital] are contacting say physios and speech therapist for our appointment, if they let them know what the patient is suffering from. (…) Because it was news to them. (…) They may have said I'm suffering from Motor Neuron but they should have said what problem or what form because when I went to speech and language first she never heard of it [bulbar onset of MND]. (…) And then the physio looked at me and said the limbs aren't a problem. What else? (lines 417-431)

4.4 Chapter Summary

This chapter presented the results of the analysis of the participants with MND. One overarching theme was revealed through the process of IPA: Living in the here and now. Participants' attempts to focus on the present were evident in their general

approach to MND, including dysphagia. MND transformed the flow of life for all participants and resulted in monumental change. However, the people with MND in this study aimed to adjust to their progressive disabilities in order to continue their participation in hobbies and "make life as normal as can be" (Joseph, line 156).

The experiences of dysphagia in MND were complex to unravel. Although participants frequently reported physical problems related to dysphagia, most denied having a swallowing impairment during the initial stages of data collection. Participants appeared to employ a problem solving approach to their illness, including dysphagia. Once they noted a particular difficulty with eating or drinking they immediately attempted to find a solution to manage it. Once the solution had been found the participants distanced themselves from the problem and focused on their here and now. It emerged that the people with MND in this study attempted to self - manage their dysphagia rather than seek professional help.

Dysphagia appeared to have multiple consequences for the people with MND in this study, from affecting their diet, requiring changes in the eating environment, to triggering a number of negative emotions. The fear of choking appeared to be particularly strong for participants. The more severe the dysphagia the more it appeared to affect participants' lives.

It became evident that as the participants noticed new symptoms of dysphagia and attempted to self - manage them, their relationship with food changed and their appetites diminished. Eating was reported as risky and physically and mentally exhausting by the majority of participants. In addition, a shared awareness of nutrition being an important prognostic factor in MND influenced participants' approach to food, as they often prioritised the nutritional value of food over its taste. Gradually, eating related pleasure decreased and mealtimes were not perceived as enjoyable.

There appeared to be a correlation between participants' attitude towards food, their perception of their own swallowing ability, and their willingness to accept non - oral feeding. Once the role of food became more medical and once participants began to experience dysphagia in their "here and now" it became somewhat easier to consent to non - oral supplementation.

Although the participants expressed their general satisfaction with dysphagia related services, they also indicated areas for improvement. Rapport with the professionals appeared to be very important and influenced participants' level of engagement with a professional service. People with MND in this study valued and engaged more with professionals who listened to them, respected their choices, and guided them through the different stages of MND with kindness and positivity.

CHAPTER 5 RESULTS: THE CAREGIVERS OF PEOPLE WITH MND (PwMND)

Introduction

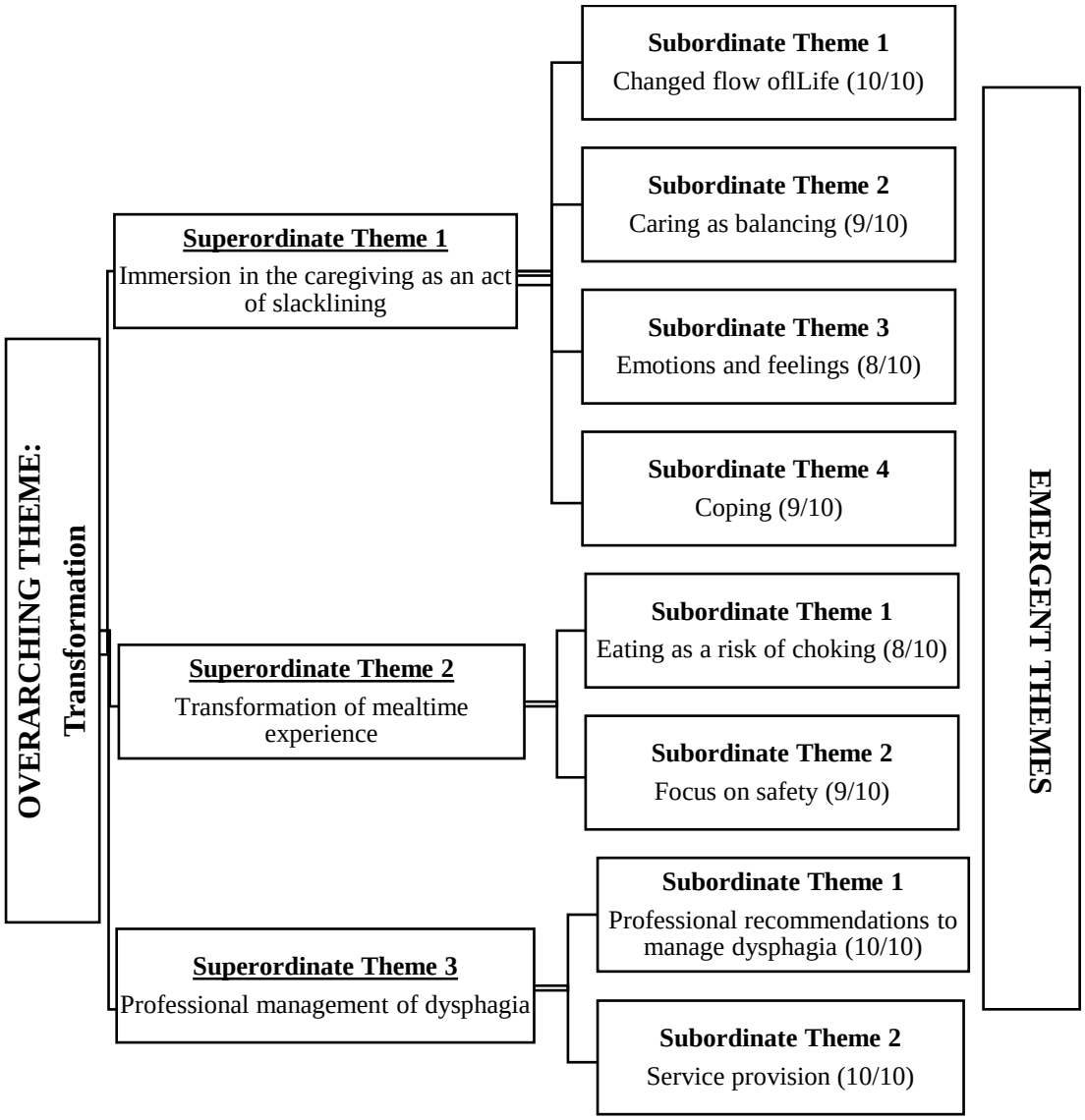
This study aimed to investigate the experiences of dysphagia in MND from the perspective of both those diagnosed with MND and those caring for people with MND. This chapter presents the findings related to the caregivers. The results of the Interpretative Phenomenological Analysis [IPA] are outlined in this chapter, including (1) a brief overview of the hierarchy of themes and the frequency of subordinate themes in the sample (Figure 5.1), (2) a more in - depth presentation of the hierarchy of themes including the emergent themes and their prevalence (Table 5.1), (3) a presentation of the themes supported by selected quotes from the sample, and (4) the textual overview of themes. This chapter, similar to chapter 4 and in accordance with the principles of IPA (Smith et al., 2009), includes both the convergence and divergence within the group by presenting the unique / idiographic emergent themes in addition to the recurrent themes.

A visual overview of the results in relation to the hierarchy of themes is presented in figure 5.1 below. One overarching theme was identified during cross - case analysis: Transformation. In addition, three superordinate themes emerged: (1) Immersion in the caregiving as an act of slacklining, (2) Transformation of mealtime experience, and (3) Professional input for dysphagia. The frequency of each subordinate theme is presented in brackets. The prevalence of themes was calculated to enhance the validity of the results (Smith et al., 2009). In this study, subordinate themes were included in constructing superordinate themes only if they were present in over half of the sample (as for the group of people with MND). However, as

subordinate themes operate at a broader level in comparison to emergent themes, variations may still remain within each subordinate theme, as participants may represent the subordinate theme from the perspective of alternative emergent themes.

In accordance with the guidelines of IPA (Smith et al. 2009) and acknowledging the researcher's background, the data was interpreted from a speech & language therapist's perspective.

Figure 5.1 Hierarchy of Themes: The Caregivers



Presentation of Findings

This section focuses on the presentation of the main themes which emerged during the cross - case analysis of the group of caregivers. The hierarchy of themes (which emerged during the process of IPA) is presented in table form (Table 5.1.) in order to enable the reader to observe how the higher order themes were established and to observe the frequency of emergent themes. The overarching theme is located at the top of the table. The three superordinate themes are located in the first column, the eight subordinate themes in the second column, and the representative emergent themes in the third column (with the frequency of each emergent theme presented in brackets).

In the subsequent table (Table 5.2), the participants' quotations are presented to provide examples regarding how the emergent themes relate to the original transcripts and how they were used to create superordinate themes. The most representative, significant, and rich quotations were chosen to support each theme. The number of supporting quotations for each emergent theme is therefore not the same. In addition, an effort was made to include the voice of every participant.

Following table 5.2, the overarching theme is introduced. The three superordinate themes are then presented. Similar to the findings of the participants with MND the first superordinate theme was related to living with MND in general and reflected issues reported as important by the participants. While superordinate theme one did not include any data directly related to dysphagia, it was considered to provide a basis for a more in - depth understanding of dysphagia in MND. Data analysis revealed that the caregivers' experiences of dysphagia in MND were embedded in their every-day experiences.

Table 5.1 Hierarchy of Themes: The Caregivers

OVERARCHING THEME: Transformation		
SUPERORDINATE THEMES	**SUBORDINATE THEMES**	**EMERGENT THEMES**
Immersion in the Caregiving as an Act of Slacklining[15]	**1.1 Changed Flow of Life**	Unpredictability (10/10) Changed plans (10/10) Living by routines (8/10) Having no break (6/10) Disturbed sleep (4/10) Neglecting self (3/10)
	1.2 Caring as Balancing	Having to cope with life on top of MND (7/10) Attempting to protect own needs (5/10) Adjusting level of care to promote independence of PwMND (5/10) QOL versus professional recommendations (4/10) Intermittent desire to know the prognosis (1/10)
	1.3 Emotions and Feelings	Shock of the diagnosis (10/10) Frustration (10/10) Loneliness (3/10) Lack of appreciation (1/10)
	Coping	Avoidance (7/10) Positive thinking (5/10) Focus on the here and now (5/10) Reciprocal relationship with the person with MND (6/10)
Transformation of Mealtime Experience	**2.1 Eating as a Risk of Choking**	Visibility of dysphagia (10/10) Food as danger (9/10) Emotional impact of dysphagia (9/10)
	Focus on Safety	Diet modifications (9/10) Supervision (7/10) Changed approach to food (6/10)
Professional Management of Dysphagia	**3.1 Professional Recommendations to Manage Dysphagia**	Consistency modifications (10/10) Non - oral feeding (10/10) Adjustment & Adherence (5/10) Contradictory recommendations (2/10)
	3.2 Service Provision	Accessibility (7/10) MND clinic (7/10) Involvement of the caregiver (6/10) Professional approach (5/10) Domiciliary service (2/10) Training (1/10)

[15] *Slacklining - walking or balancing along a suspended length of flat webbing that is tensioned between two anchors and bounces almost like a long and narrow trampoline (Utton, 2015)*

Table 5.2 Presentation of Themes Supported by Chosen Quotations from Transcripts: The Caregivers

Superordinate Theme 1:

Immersion in the Caregiving Duties as an Act of Slacklining

Subordinate Theme 1: Changed Flow of Life

Emergent themes

Unpredictability

Tom: But what we don't know is what the prognosis is. Like we don't know what's going to happen? Is it going to just stay in this area? Is it going to move and if it's going to move where is it going to move and you know, seemingly it's not an ordinary case of MND. So we're sort of in limbo as far as that's concerned. (lines 89-94)

Kay: (…) you don't know what you're coming down facing again in the morning. (lines 384-385)

Changed plans

Martha: We were going to find somewhere else [to live] so that's part of the trouble, we were visiting different places (…). You know not straightaway. We have two dogs. We were going to wait till, you know, maybe one of the dogs died or something (laughs). (…) so we used to then, Mark [husband] used to plan the year and we'd have lots of trips. (…) You know to break up the year and you know. So, I think, that's the thing that disappointed him most. He's had the year planned and never had to cancel it. (Upset) (lines 500-516)

Living by routines

Angela: You just become a bit robotic with it all. It's just all routine, isn't it? (…) Its real routine, isn't it? It's a rigid routine as well. It's like its exact timing. (lines 979-982)

Having no break

Mary: So, it's [caring] kind of full-time like you're on the go all the time. There's no let off like. (line 898)

Kay: (…) my head is constantly is she [her mother] sleeping? (…) what kind of a night is she having? (…) My head is constantly. (…) I don't think I ever, even if I was gone away, you know, the rare times that I could go away for a night I don't think I switch off because I'm thinking, I hope the lads are after giving her that drug or I hope they're after doing this, you know, I'm constantly wound up. (lines 371-378)

Sophia: (…) if I leave the house, I play golf and I haven't been playing that much golf but I went back now last week (…) I find that even on the golf course I'm just thinking, I hope now he's okay. (lines 526-529)

Martha: I suppose I'm waking up earlier than I ever used to (…) I'm in and out to the kitchen and I'm looking in on him the whole time (lines 610-615). So all those little jobs around the house and then I'm in and out to him all the time because I pass the bedroom. I leave the door open just to see if he's awake. (lines 633-634)

Disturbed sleep

Mary: The biggest thing I find is, em, sleep deprivation, em, (sighs). I have to be very strict so I'd be trying to get him [her husband] to bed between half nine and ten. Like its terrible you can't just say I want to go to bed. (lines 656-658).

Jackie: I might wake earlier some nights but like obviously Chris [husband] is on the air machine and I know it's supposed to be silent but like some nights it wouldn't be silent. (lines 192-193)

Neglecting Self

Interviewer: So, em, if I was to ask what do you do for yourself?

Kay: Truthfully, nothing (laughs). I don't think there's nothing, I don't know. There's nothing. (lines 420-422)

Subordinate Theme 2: Caring as Balancing

Emergent Themes

Having to cope with life on top of MND

Angela: And I have three children of my own that go to school. (…) And so I come up and down to mum as regular as I can when they're off school. (lines 21-24)

Jackie: (…) we just tried to stay positive and we still do but it's harder because Chris [her husband] is deteriorating. Well I suppose I find I can kind of cope away with my husband and life but then when something else happens (…) it knocks you big time. If you understand? (lines 143-146)

Attempting to protect own needs

Mary: (…) I try and walk in [to work] because I always liked exercise and its hard now to get it so I'm up at half five in the morning. (lines 20-21) (…) Try and read the papers if I can because I always liked looking, you know, reading the papers. (lines 97-98)

Helen: I'm cutting back on the travel that I do for work because of it [caregiving duties] (…) We [siblings] have issues, you know, I've been told no more long holidays and I'm going excuse I work hard. I think I'm entitled to my holidays. (lines 760-764) (Interviewer: What keeps you going?) Getting away (laughs). Exercise. (…) Exercise like I run and I cycle and I swim. (…) Like if it means I have to get up and cycle early or I do something in the evening then I'll do it like yeah, yeah. (lines 914-922)

Adjusting level of care to promote independence of PwMND

Sophia: He does it [tube feeding] himself and I leave him do it. I did it in the very beginning with him. I used help him but now he's able to do it and I say look I'll leave him do it as long as he's able. (…) Because I feel when he's isn't I will be there to do it, you know. (lines 868-873)

QOL versus professional recommendations

Una: (…) no offence to John [Physiotherapist] but the day he came I went oh now here, enough. Because it was the same hammering of the powder to dad and he had a full day in [names a hospital] and I just felt he could do without it for one day that bloody powder. (…) It is massively about quality of life. (lines 56-64)

Intermittent desire to know the prognosis

Kay: (…) what's down the line mammy doesn't want to know any of that. So, I suppose, I don't want to know either even though sometimes I do and sometimes I don't. It all depends on the particular day. I'd say God why didn't I find out, is this normal or this is, you know. (lines 805-810)

Subordinate Theme 3: Emotions and Feelings

Emergent Themes

Shock of the diagnosis

Kay: I was just speechless, do you know like didn't think and I wasn't expecting that [MND] and when he [doctor] said to me he said he kind of said: you know what I'm going to say and I was kind of looking at him saying no I don't (…) and he said Motor Neuron and I was there oh my God. Do you know and I suppose I knew from hearing from television at that time. When mammy was diagnosed it was kind of in the television like. A newsreader had died from it and you were just kind of saying oh Motor Neuron but you don't know (…) you don't know what was involved in it. (…) Yeah, so that was a bit of a… That day is one day I'll never forget. (lines 508-518)

Frustration

Tom: And it can be a bit frustrating too because there's so many different, there must be about six different types of medication to be given at six different times in the day. (lines 236-238)

Jackie: You're hitting a brick wall the whole time like but, em, it doesn't make any difference. It doesn't make any difference because nobody can suggest the slightest thing that's of help. That's why we gave up going to [names a service] because nobody can do anything. (lines 1005-1008)

Loneliness

Martha: Its more lonely than anything (…) because he sleeps a lot of the time so, you know, very often when I've got everything done and he's asleep, you know, I'm (upset) there on my own sort of thing, you know. (lines 1236-1240)

Lack of appreciation

Mary: Yeah he's [her husband] no empathy at all from my side of the problem. That he's the victim in all this. (lines 1433-1434)

Subordinate Theme 4: Coping

Emergent Themes

Avoidance

Mary: So like I just go along blindly and say right keep going and go into work, do your work and enjoy the social side of it inside as well, you know (lines 461-462). That's what I say and I'm lucky I'm working like because I'm working with a big gang and em, there's great camaraderie and that, do you know what I mean? So if I was here all day I'm sure I'd ate him. I wouldn't cope at all. (lines 1061-1063). And I never talk about it [MND], you know, I never talk. (line 1586)

Martha: I'm trying to distract myself with cleaning the house and stupid things, you know, that I wouldn't normally bother so much about, you know, just to keep busy. (…) And then I'm not thinking about things. (lines 239-242)

Positive thinking

Sophia: Positive, I think is a huge, a huge thing. Yeah, it keeps you…and laugh as much as possible yeah because life is short really (…) I said to Paul [husband] in the beginning you'll see a lot of your friends (…) will be gone before you I bet you. (…) he'd say ah sure I probably won't have long to live (…) I'd say Paul you don't know. I could be gone before you. (…) you have to be positive because you just never know. (lines 1035-1045)

Una: So life is good (…) as good as can be, isn't it? (lines 59-60)

Jackie: (…) be positive and to get up every day and make the most of every day and to carry on as normal. (…) I would say carry on as normal once you get over the initial shock and make the most of every day and don't let anything pass by that you could do. (…) and like we've tried to keep life normal. (lines 628-655)

Focus on the here and now

Kay: I just kind of this is what we're at now. This is how she is, do you know, okay what do we do, you know. That's the way it is like. I don't know whether it's a good thing or it's a bad thing but that's just the way we can cope. (lines 588-590)

Tom: I've a feeling that's [medication] working because she [wife] doesn't seem to be getting worse since (…) her speech is definitely gone but I don't think she's getting worse. I don't think she is, you know. Hopefully she's not. She's getting stronger which is good. (lines 927-931)

Reciprocal relationship with the person with MND

Kay: I can cope better, I suppose, if her [mother's] mood is, if she's not too bad but when she's down I'm, you'd be down yourself too. (lines 392-393)

Superordinate Theme 2:

Transformation of Mealtime Experience

Subordinate Theme 1: Eating as a Risk of Choking

Emergent Themes

Visibility of dysphagia

Kay: Because she [her mother] could be sitting on there and she'd be coughing and next minute you'd have to tap her on the back and I'd be going Jesus is it stuck? Yeah the food was kind of lodging back along. (lines 248-250)

Jackie: Well they [children] can see that [signs of dysphagia]. (…) They know, they can see that because Chris [husband] eats slowly number one, and number two he, em, can kind of gag on his food at times and he can get dreadful coughs trying to bring up stuff and they can see that his dinner is soft and that his potatoes are mashed up with butter and they can see. They know like that he finds it hard to swallow and they know he finds it hard to swallow and hard to talk. (lines 812-817)

Food as danger

Mary: It [choking] wouldn't happen all that often but you do have to be careful of what you give him and like there would be certain things he couldn't eat at all. (lines 783-784)

Tom: (…) and then [doctor] more or less said, you know, you have to try and cut the food out because it's too dangerous. (lines 913-914)

Helen: (…) he's [her father] putting a load of stuff on the fork. I'm there going please don't choke on that, (lines 920-921)

Sophia: Now we went to a restaurant there one time (…) and something got caught very … something very, very small got caught (…) It was a tiny little piece of lettuce, a tiny, tiny, yeah. It probably shouldn't have been on the plate. (…) It was just accidentally the little thing got in yeah. That has happened to us about three times. (lines 538-556)

Jackie: He [her husband] got a fright one day with bread and he went off bread straightaway then. (…) it kind of caught yeah. (…) he must have got a fright. (lines 308-313)

Emotional impact of dysphagia

Kay: Because she [her mother] could be sitting on there and she'd be coughing and next minute you'd have to tap her on the back and I'd be going Jesus is it stuck? (…) You were uneasy all the time when she was eating because do you know you'd be kind of watching was it going down or was it staying and getting stuck. (lines 248-253)

Martha: (…) it [the swallowing] is at the back of my mind because it's all here, the swallowing is on my mind. Do you know I don't want him choke. It is on my mind so if I think about it, if I'm doing him a sandwich. (lines 357-359)

Subordinate Theme 2: Focus on Safety

Emergent themes

Diet modifications

Mary: I wouldn't be a marvellous cook (…) so we decided we'd stick to fish one night and chicken the next night (lines 188-191). (…) So we live on chicken and fish (line 270) (…) Like we kind of, we're very bland like. We stick to the same things all the time. (lines 301-302)

Angela: Everything, literally everything gets chopped up. (line 145)

Jackie: Yeah dinner then would come then. I would basically, em, we have mince now today. Well basically then I would mash the, I did mash Chris' [her husband's] potato with butter and make it buttery and creamy and the vegetables would be mashed up with plenty butter (laughs) added as well and the meat would be obviously soft. Em, it would all be very soft. (lines 239-244)

Martha: Like I said to you last time I am conscious when I'm cutting the fruit up in the morning, I'm making sure there are no big bits, you know. (lines 171-172)

Supervision

Helen: We come back to the house and I do dinner. It's just a matter of heating up the shepherd's pie and he [her father] lashes into it. I have to go, "Dad slow down." (lines 456-458)

Sophia: I just say [in restaurants] he's a difficult swallow. He had to have a soft diet and they say okay and they're probably looking at me like, can he eat? Now I tell them what he can eat and mostly they have would have vegetables that would be like beans or you know, there would be chunky vegetables on and I'd say don't bring any vegetables. Just bring mashed potato and salmon. That will be fine. I'd rather just have the mashed potato and the salmon rather than have anything else on the plate. (lines 978-983)

Helen: (…) Dad gets up, I know, because I live a field above him. So when I'm sitting down our house is split level so my kitchen and everything is upstairs so I can actually see down to his driveway. (…) I can see him strolling down to the shop. (…) if I'm having breakfast and I'm finished at eleven and I haven't seen him go for a walk I'll go down to see if everything is okay. (lines 427-444)

Changed Approach to Food

Jackie: Well I buy cream and double cream on a very regular basis. (…) for calories. (…) I might make the soup but I'd put a lot of cream in Chris' (…) to keep the calories because like I was told to maintain his weight. On day one the first thing, one of the Motor Neuron nurses she says just keep the weight on. (lines 216-222)

Superordinate Theme 3:

Professional Management of Dysphagia

Subordinate Theme 1: Professional Recommendations to Manage Dysphagia

Emergent themes

Consistency modifications

Helen: In the [hospital] they were looking at changing his diet and the whole lot. So that's why he's eating… (…) basically that there was going to be, (…) no more T-bone steaks (laughs). (…) And that type of food. Keep it nice and soft yeah. (lines 245-270)

Non - oral feeding

Tom: Oh yeah I'm very pleased [with artificial feeding] and Orla is gaining strength. (…) She's getting heavier and she's gaining strength so I'm very pleased with that. (lines 936-939)

Martha: She [SLT] was going on about, you know, the fact he said you can't leave it. She was quite forceful in that she was sort of saying you can't leave it. You have to make this decision because your breathing is bad and you won't go through an operation having the tube fitted if you're, you know, so don't leave it to the last minute. You need to make a decision. (lines 503-507)

Adjustment and Adherence

Kay: (Interviewer: What was going through your head when you heard about the [feeding] tube?) Psychologically that she [her mother] has to have this, and you know the headset at that time it was just in case but sure only in your head you're kind of saying oh it's just in case but when that day actually came that you knew it was, this is it now, that you have to use that tube for as long as we can, that was the hard part trying to get that around. (…) That adjustment, do you know, because when she first got it I thought oh yeah, yeah it's down the line. You'd be thinking oh its way down the line we're going to have to use it but as soon as she got it, it was only a couple of months like that we were using it like, do you know. (lines 204-241)

Sophia: Yes they [SLTs] did mention thickener and I bought it and bought a little container and all to mix it and no, he wouldn't touch it. He just wouldn't it. He just couldn't go there. No. (…) And I can see it would be very difficult. (…) No and if he doesn't want to do it that's it, that's it. Still have the carton there. I held onto it in case that he might may be say at some stage that he might like maybe even a drink of orange or anything you know, but no he doesn't really. (lines 414-423)

Contradictory recommendations

Tom: She [SLT] gave her [wife] a chart with exercises to do for her mouth (…) And they told us not to do that in [names another SLT service]. (lines 235-238)

Subordinate Theme 2: Service Provision

Emergent Themes

Accessibility

Kay: (…) like she [SLT] was there if you ever have any problem just ring me up with anything. (line 637)

MND Clinic

Mary: You've, it's kind of a multidisciplinary team. (…) Well you go in. We'd often get an early appointment and there mightn't be many there (…) And they're good enough. One of the team might call him [her husband] in or the team might call him in and talk to him and all this, have you any issues and all the rest of it and then or he might see the doctor first whichever. So they kind of work around one another (…) So you see all the services in the one day so that's good. (lines 465-478)

Kay: I don't particularly like it [MND clinic]. Because (…) you are looking around at like everybody that comes in. You're there oh my God they sound like mam or (…) are they worse than mam and you're walking so you're not, (…) that's what you're like while you're waiting to go in to see the lads, like that's what you're basically doing do you know. Zooming, just eyeing up the room and just seeing do you know that's. Like sometimes I nearly prefer to say if you had an appointment and bypass all that and just straight into the team and that's it. (lines 562-570)

Jack: Well we're going back there [names service] again alright in February like, towards the end of February but, eh, I don't know. You know, you'd hope that (…) that whatever has to be done would be done quicker in that and not the same amount of hanging around, you know. (lines 305-308)

Involvement of the Caregiver

Sophia: I think if a partner could get to talk they [professionals] might see a different side. (Interviewer: So would you say that maybe going for an appointment on your own would help?) I would, I would, yeah, yeah. Now and you don't want to make it worse but you'd like to give the truth. (…) The truth is all you need really, you know. (…) you won't get to the next stage fast enough I feel if you don't give the proper information, you know, Paul [her husband] may not have had the peg maybe, maybe may not have the food if he [doctor] was going by Paul. They'd think he was fine, you know. (lines 1125-1151)

Professional approach

Helen: No in the [names hospital] and they were looking at changing his diet and the whole lot. So that's why he's eating…But there was a [therapist] (..) and was awful. It was very much like an assembly line therapy (…) And it like talking to my father like a child and my father just blanked it. (…) My father was walking slowly in front. Olga [her with the therapist behind him and the therapist basically handed over the exercises because he gave it to my father and my father said, "We don't need that". Right! So the therapist picked it up and gave it to my sister on the way out and my father spotted it and took it off her. I said, "We don't need that." (Laughs) (lines 245-262)

Professional approach (continued)

Una: Patrick [therapist], who just was there gave out to dad the last time because dad took a drink of water, he [Patrick] wasn't in the door 2 minutes and he said: Una you just stood there and I said that's what I do. I don't ever. (…) he [the father] is a grown up man (…), but then when Patrick said I didn't understand it. In cold water it [the thickener] doesn't dissolve so it is manky. You couldn't blame anybody (…). I mean actually, no offence to Patrick but the day he came I went oh now here, enough. Because it was the same hammering of the powder to dad and he had a full day in [names hospital] and I just felt he could do without it for one day that bloody powder. Sorry. (lines 43-59)

Domiciliary service

Angela: Yeah, well I mean for someone like mum no one is willing to come out, you know, no one wants to come here to see her. They want her to go like to go there. (…) You know, that even in the palliative care unit there isn't anyone like that.(…) Like home, no one will come out to see her and the way she is she doesn't want to go in which is difficult so she just doesn't get seen by anyone really. (lines 722-728)

Training

Jackie: Well to be honest most of it [dysphagia training for the caregivers] I felt I hadn't learnt much new there because I had been, I spend an hour; I had been with Chris [husband] the previous week or two [in speech therapy]. I had been above there on an individual basis and I kind of… (lines 403-407)

The Overarching Theme: Transformation

This section introduces the overarching theme for the caregivers as identified through the process of IPA: Transformation. This transformation occurred as a result of the changed flow of life (Subordinate theme 5.1.1), which was experienced as the participants immersed themselves in their caregiving duties (Superordinate theme 1). This change of life was related to various aspects of living with MND including dysphagia (Superordinate theme 2).

The process of transformation appeared to commence once the diagnosis of MND was confirmed and the participants became aware of the nature of this disease. "Terminal", "no cure", "progressive" – the prognosis was typically communicated at the time of diagnosis, although some participants (3/10) reported suspecting MND before it was officially confirmed. The initial period of being a caregiver was rife with strong emotions (Subordinate theme 1.3) such as shock, as individuals were thrust into this role suddenly and often unexpectedly. With time, the caregivers strove to adjust to the reality of living with MND. However, this adjustment led to a significant lifestyle transformation. This transformation was noted to involve various areas of life, for example, work, sleep, and participation in social activities.

The role of caregiving involved many additional and often new duties, such as having to administer non - oral feeds through a gastrostomy tube. Duties such as this appeared to significantly alter participants' previous roles within their families. Children and spouses of people with MND became caregivers with the roles and relationships gradually transforming as the disease progressed.

The process of transformation appeared to be ongoing as a result of the progressive nature of MND. Although there were some moments of stability, the

participants seemed aware that these moments would be brief as MND would undoubtedly continue to transform their lives. It was noted that some caregivers maintained the hope that they would resume their pre - MND life at some point in the future. Perhaps holding on to this hope helped them to cope with living with MND, however, the possibility that MND may lead to a permanent and non - reversible transformation of life cannot be ruled out.

Acknowledging the overarching theme may lead to an improved understanding of the caregivers' lives in general. Therefore throughout this chapter references to the over - arching theme are made. The overarching theme is further reflected upon when describing specific aspects of participants' life, such as their caregiving duties, coping strategies, and dysphagia - related experiences.

5.1 Superordinate Theme 1: Immersion in the Caregiving as an Act of Slacklining

This section focuses on the presentation of Superordinate theme 1 and the corresponding subordinate themes found in the data. This superordinate theme strongly emerged from the data and it was included in the cross - case analysis despite the fact that it did not appear to be directly related to the topic of dysphagia. However, it was hypothesised that having a more in -depth awareness of the participants' caregiving experiences in general could enhance the understanding of their experiences and perceptions of dysphagia.

Superordinate theme 1 likened participants' immersion in the caregiving duties to an act of slacklining. Slacklining refers to walking or balancing along a suspended length of flat webbing that is tensioned between two anchors (Utton, 2015).

Slacklining differs from slack rope walking and tightrope walking as slacklines are tensioned significantly less than tightropes. In slacklining the line is dynamic and it stretches and bounces almost like a long and narrow trampoline. This appears to resemble participants' caregiving duties which were intensive, dynamic, unpredictable, and full of "ups and downs". In addition, the participants had to maintain their full concentration and focus in order to balance their general duties (such as looking after own children) with providing care to their terminally ill relative. When Mary reflected on her life she highlighted that caregiving demanded constant mental effort: "You've to be on the ball. If you forget a thing then like, this awful, you know" (lines 804-805).

Superordinate theme 1 consisted of four subordinate themes: (1) Changed flow of life, (2) Caring as balancing (3) Emotions and feelings, and (4) Coping. These themes are discussed in the subsequent sections to provide the reader with a more advanced understanding of the caregiving experiences of the participants.

5.1.1 Subordinate Theme 1: Changed Flow of Life

MND transformed participants' lives significantly. This change became particularly evident when the caregivers of people with MND in this study began to compare their current lifestyle with their lifestyle prior to MND. Participants perceived that their "own life" (life pre - MND) was "put on hold" and they were hoping to be able to return to it at some point in the future. Mary, for example, spoke about "her life being on standby" (line 131) which she contrasted with her life pre - MND ("we were just moving on and on", line 1066). She also reported "We are surviving" and "we get by", a few times throughout the interview. Her husband had been surviving

with MND for over six years (far beyond his original prognosis) and she was surviving her life, which had come "to a cruel dead stop".

> Do you know I find it extremely difficult like because things always went our way and we were just moving on and on, (…) but it just came to a cruel, dead stop. (…) we're stuck in a rut now but what can we do? (Mary, lines 1065-1067).

In order to provide specific details on how MND changed the flow of life for the participants, the emergent themes are presented below. There were six emergent themes distinguished during the data analysis process: (1) Unpredictability, (2) Changed plans, (3) Living by routines, (4) Having no breaks, (5) Disturbed sleep, and (6) Neglecting self.

Emergent Theme 1: Unpredictability

All caregivers in this study reported that their lives had become uncertain and unpredictable. This uncertainty appeared to be related to their inability to predict the trajectory of MND. Tom, for example, reported "we don't know what's going to happen" (line 89). Although he understood MND to be a terminal disease, he expressed his uncertainty in relation to the progression of the symptoms of this disease. This uncertainty affected the flow of Tom's life, as it made him feel "in limbo".

> But what we don't know is what the prognosis is. Like we don't know what's going to happen? Is it going to just stay in this area? Is it going to move and if it going to move where is it going to move and you know, seemingly it's not an ordinary case of MND. (…) So we're sort of in limbo as far as that's concerned. (lines 89-94)

Kay also indicated that her caring duties were unpredictable: "You don't know what you're coming down facing again in the morning (lines 384-385). She further reported that her mother's mental state and ability to cope with MND fluctuated, which also caused fluctuations in her own mood. She additionally reported that when her mother was in good form she attempted to assist Kay with her caregiving duties, which reduced her own burden of care.

Emergent Theme 2: Changed Plans

MND came into participant's lives unexpectedly and significantly affected their plans for the future. All caregivers reported having to be at home with the person with MND the majority of the time, which destroyed their plans to travel and affected many other activities typically carried out outside the home, such as meeting friends. Martha stated that having to cancel travel plans due to MND was most disappointing for her husband: [he] used to plan the year and we'd have lots of trips. (…) to break up the year (…) I think, that's the thing that disappointed him most. He's had the year planned and never had to cancel it. (Upset) (lines 500-516). Mary reported that prior to MND her family went on holiday twice a year, however, "we don't go anywhere now" (lines 248-249).

Emergent Theme 3: Living by Routines

The flow of caregivers' lives appeared to become very repetitive as it was dictated by rigid routines developed by the caregivers in order to manage the needs of the person with MND. When asked about their typical day all participants reported the exact timing and sequence of their activities. Angela reported that she was

becoming "a bit robotic with it all. It's just all routine, isn't it? (…) Its real routine, isn't it? It's a rigid routine as well. It's like its exact timing". (lines 979-982). Routine appeared to be important to the participants and it was frequently reported as the best way of managing life with MND.

Emergent Theme 4: Having no Break

Caring was reported to be a continuous duty especially considering that the participants provided both physical and psychological support to the people with MND. Kay stated that her head was constantly occupied with thinking about her mother, therefore she perceived herself to be unable to "switch off" from her caring role:

> My head is constantly is she [her mother] sleeping? (…) what kind of a night is she having? (…) My head is constantly. (…) even if I was gone away, (…) I don't think I switch off because I'm thinking, I hope the lads are after giving her that drug or I hope they're after doing this (…) I'm constantly wound up. (lines 371-378)

Similarly, Sophia reported thinking about her husband even when she was alone and engaged in her hobby: "if I leave the house, (…) I find that even on the golf course I'm just thinking, I hope now he's okay". (lines 526-529)

Both Mary and Tom compared caregiving to a full time job. Mary reported: "So, it's [caring] kind of full - time like you're on the go all the time. There's no let off like" (line 898). Tom stated: "I used to sort of potter around and I'd get up in the mornings and say what will I do today? I don't, I can't do that anymore because I know what I have to do, you know and it's a full - time job". (lines 128-130). It should

be noted that Mary's husband was severely affected by the disease from a physical point of view and therefore required a high level of assistance to carry out activities of daily living, such as dressing or toileting. In comparison, Tom's wife was independent for the majority of household related activities, such as cleaning or cooking. She required Tom's help with non - oral feeding and when attending medical appointments. Interestingly, despite these significant differences in relation to the physical impact of MND on their spouses, both caregivers perceived their caregiving duties to be intensive and ongoing (a full time job). This finding may suggest that caregiver's perception of their burden of care may be individualised and not directly related to the physical abilities of the person with MND.

The caregivers appeared to be unwilling to leave the person with MND on their own and they strove to provide constant supervision. Martha reported:

> I suppose I'm waking up earlier than I ever used to (…) I'm in and out to the kitchen and I'm looking in on him the whole time (lines 610-615). So all those little jobs around the house and then I'm in and out to him all the time because I pass the bedroom. I leave the door open just to see if he's awake. (lines 633-634)

Emergent Theme 5: Disturbed Sleep

Some participants reported not being able to sleep well due to their caregiving. For Jackie, the noise coming from her husband's non - invasive ventilator was disturbing: "I might wake earlier some nights but like obviously Chris [husband] is on the air machine and I know it's supposed to be silent but like some nights it wouldn't be silent" (lines 192-193). Mary reported her sleep deprivation far more than other

participants, perhaps because she had the longest caregiving experience and also maintained a full time job. Lack of sleep was "the biggest thing" for her: "The biggest thing I find is sleep deprivation (sighs). (…) Like it's terrible you can't just say I want to go to bed". (lines 656-658).

Emergent Theme 6: Neglecting Self

Maintaining their full focus on their caregiving duties appeared to take priority over the caregivers' own needs. When Kay was asked about what things she would do for herself she reported: "Truthfully? Nothing (laughs). I don't think there's nothing, I don't know. There's nothing." (lines 416-417). Kay's life now revolved around the needs of her mother and these needs often took priority over the needs of her children and husband. She admitted that she found it difficult to maintain the same level of engagement with her family as she had pre - MND due to time constraints and exhaustion.

Mary reported having no opportunity to do anything she would like to do for herself as she had to prioritise her husband's needs. Her life imprisoned her as every minute of Mary's time was carefully planned around her caregiving duties. She compared her life to "a cruel, dead stop" (lines 1066-1067), indicating how hard and merciless it had become for her. In the quotation below Mary reported how she had lost her freedom and ability to make her own choices due to having to be constantly mindful of her husband's needs.

> It is a very, very hard life because you can't do what you want. (…) it would be great to get up on the morning and say oh God I'll go down now and have

my breakfast and I'll head out but you can't because you have to consider him. (Mary, lines 1299-1304)

5.1.2 Subordinate Theme 2: Caring as Balancing

Being a caregiver of a person with MND required having to balance various aspects of life and having to face many difficult decisions. This is further explained through the presentation of the five emergent themes: (1) Having to cope with life on top of MND, (2) Attempting to protect own needs, (3) Adjusting level of care to promote the independence of the person with MND, (4) Quality of life versus professional recommendations, and (5) Intermittent desire to know the prognosis.

Emergent Theme 1: Having to Cope with Life on Top of MND

Becoming a caregiver for a person with MND did not appear to reduce the need to fulfil other duties, such as looking after children, cleaning the house, shopping, or having to work. The majority of caregivers reported that they had to balance their general duties with their caregiving duties. Although participants perceived their own lives as "being on hold", their life around them continue to flow, therefore maintaining their full focus on the person with MND was challenged by other demands. Even if a sense of balance in life was maintained for some time, new challenges, often unrelated to MND, could present themselves at any time. These challenges were reported by the majority of the group as particularly difficult to manage when MND had already transformed their lives. Jackie reported: (…) "We just tried to stay positive and we still do but it's harder because Chris [her husband] is deteriorating. Well I suppose I find I can kind of cope away with my husband and life but then when something else

happens (…) it knocks you big time. If you understand?" (lines 143-146). It seemed that providing care to a person with MND was exhausting and overwhelming and meant that any additional stressful life events which occurred led to participants feeling unable to cope. Caregiving was perceived as an ongoing process of adjustment; however, this process did not appear to be linear or smooth. There were frequent moments of panic, fear, and despair, sometimes related to the progression of MND, and at other times associated with other life issues. However, even when participants felt "knocked big time" (Jackie, lines 145-146), they reported that they had no choice but to cope with each new challenge in order to continue providing care for the person with MND.

Emergent Theme 2: Attempting to Protect Own Needs

Half of the caregivers reported that they made attempts to protect their own needs, such as participating in their hobbies. Participants considered these attempts as a way of protecting their quality of life and ultimately improving their ability to cope with MND. Mary, for example, chose to walk to work over taking the bus in order to maintain some level of physical activity, especially considering that walking home after work was no longer a possibility as she had to rush home to feed her husband. However, walking to work in the morning meant that Mary had to get up much earlier than she previously did, which considering her previous reports of sleep deprivation seems important. Perhaps Mary found walking to work in the morning peaceful, a chance to be alone with her thoughts, and an opportunity to walk towards something pleasant, as she frequently reported that work had a positive impact on her life with MND.

Helen reported that she felt "entitled" to get a break, as she worked full time job and also cared for her father in the evenings and at the weekends: "I work hard. I think I'm entitled to my holidays. Well we can't be looking after dad all the time and I'm doing weekends" (lines 762-725). This appeared to be quite an emotional issue for Helen, as she admitted feeling expected to provide more caregiving than her siblings, as she was the only one without children. However, Helen appeared motivated in her attempts to establish a balance in her life. She reported meeting her siblings in order to negotiate and agree the division of caregiving duties so that she could maintain her participation in other activities. Helen also highlighted that a shared model of care provision required good communication, planning, and negotiation skills. Although conflict between siblings was reported as being unavoidable, the needs of the person with MND always formed the central concern. Helen reported:

> We did this [planning for care] in terms of the weekends and we had a meeting. We all sat down and had a meeting with our schedules and went okay I can do that weekend. I can't do that weekend. I'm away that weekend. I'm away and then if there's a major issue then we go right how can we work this out? We've worked it out between us. (lines 875-878)

Emergent Theme 3: Adjusting Level of Care to Promote Independence of the Person with MND

Half of the caregivers reported their attempts to optimise the level of independence and engagement of the person with MND. Sophia, for example, decided not to interfere with the administration of her husband's non - oral feeds for as long as he was able to do it independently. Jackie reported encouraging her husband to go to work, even when he did not have enough stamina to carry out any work related duties.

Una referred to her father as "a grown up man" and when she attempted to assist him during meals: "it came to a point that I was holding the cup and he was pouring down" (lines 46-47). She reported feeling more comfortable allowing her father to be as independent as possible, while at the same time monitoring him closely to ensure his safety.

Emergent Theme 4: Quality of Life (QOL) Versus Professional Recommendations

Some caregivers reported that, in their opinion, particular recommendations made by professionals had a negative impact on the QOL of the person with MND. Una, for example, began to speak faster and louder when reporting that she had repeatedly received the recommendation to thicken her father's liquids. She reported that thickened liquids had a negative impact on her father's QOL and therefore she had to balance the professional recommendations with QOL values, which she considered most important for her father.

> Adam [therapist] who just was there gave out to dad the last time because dad took a drink of water; he [Adam] wasn't in the door two minutes and he said: Una, you just stood there and I said that's what I do. I don't ever (…) In cold water it [the thickener] doesn't dissolve, so it is manky. You couldn't blame anybody. (lines 43-50)

Emergent Theme 5: Intermittent Desire to Know the Prognosis

Although the caregivers reported their uncertainty in relation to the trajectory of MND, they appeared to be unsure whether they would prefer to know the detailed

prognosis of the person with MND or not, especially considering that the majority coped with MND through avoidance. Kay illustrated her conflicting thoughts in relation to knowing her mother's prognosis. Kay admitted that her wish to know "what is down the line" varied depending on the day: "I suppose, I don't want to know either [what's down the line] even though sometimes I do and sometimes I don't. It all depends on the particular day. I'd say God why didn't I find out, is this normal or this is, you know". (lines 808-811). It appeared that sometimes Kay coped better by staying focused on the here and now, but at other times she regretted not having asked questions related to her mother's prognosis. For Kay, regular access to professionals was very important as it allowed her to balance her needs and access information when she was feeling able to handle it.

5.1.3 Subordinate Theme 3: Emotions and Feelings

Caregiving appeared to resemble a journey through a range of emotions and feelings. In total, four themes emerged during data analysis: (1) Shock of the diagnosis, (2) Frustration, (3) Loneliness, and (4) Lack of appreciation.

Emergent Theme 1: Shock of the Diagnosis

The moment of diagnosis of MND was reported by all participants in this study as being the biggest shock. Kay, for example, described the day when her mother was diagnosed with MND as: "That day is one day I'll never forget (…) It was horrendous …(…) she [mother] was just roaring crying and I'd say for the next three days afterwards it was horrendous" (lines 518-524). Moreover, even the caregivers who had suspected MND prior to it being medically diagnosed reported their shock in

relation to receiving confirmation of the diagnosis. Although Sophia felt "geared up" for the diagnosis of MND she still described it as a "big shock".

> I just assumed straightaway that it was Motor Neuron. I just got it into my head. My other daughter said I think you're really exaggerating now, but I said I've a funny feeling that it's something like that. (…) I got a big shock when we got the news but it wasn't as devastating a shock because I was kind of geared up for it. (lines 87-91)

Jackie reported the impact the diagnosis had on her and how she developed physical symptoms related to the stress of hearing her husband's prognosis. She appeared hurt by a question asked by the doctor in relation to their "stolen future".

> Well, all I remember is her [doctor's] first words to us… We went into the office and we sat down (…). We had a chat about (…) and this and that (…) and the weather and then she spoke. Her first words were, "Well, how do you feel that your future has been stolen?" and I'll just never forget those words. And no answer. (Short pause). So, I suppose at the time (crying) I was extremely upset, obviously (…) sometimes you'd hear people say a kind of a phrase: I got a pain in my stomach when I heard it or something to that affect, but I actually never knew what that meant until then because I could physically feel a pain in my stomach. (lines 122-137)

The moment of diagnosis, although emotionally very significant, did not appear to mark the beginning of the journey for the majority of caregivers. Instead, they recalled the time prior to diagnosis, when they noticed deterioration in the health of their loved ones without knowing the cause. All caregivers perceived that the official diagnosis of MND had been delayed and two main reasons emerged in relation

to this. Firstly, participants reported that the professionals had difficulty recognising the disease (such as misinterpreting the symptoms of MND for a cerebrovascular accident or cancer). Secondly, some people with MND appeared reluctant to look for medical help in order to explain their symptoms, which also led to the diagnostic delay. Both situations appeared to result in frustration for the caregivers.

Emergent Theme 2: Frustration

All participants reported their feelings of frustration, predominantly in relation to their perceived inability to stop MND or influence its course despite their best efforts. Helen, Mary and Kay's frustration, however, also became evident when they reported that the role of caregiver had been an expected duty rather than a choice. Mary stated having "no choice in the matter" (line 105). Kay reported that she felt "expected" to look after her mother as she was the only person in the family working part - time. Additionally, being female also appeared to be a factor, as according to Kay caregiving is "expected of the girls" (line 461). Helen also felt pressurised to take on more caregiving duties as unlike her siblings she did not have any children. All three participants reported their frustration when comparing their current life style with that of other family members.

Emergent Theme 3: Loneliness

Loneliness was often experienced as the participants had few opportunities to engage in social activities due their full focus on the person with MND. Tom, for example, reported losing contact with friends due to his wife's MND. In his opinion, her inability to speak negatively impacted on their visitors as it made them feel

embarrassed or awkward. It emerged that MND took away Tom's friends and instead brought many new strangers (professionals) into his life. Over time only the postman continued to visit Tom's house regularly.

> (…) She [Tom's wife] was always bubbly, very bubbly (…) so it's just a complete change. (…) And you find a lot of people now (…) they be embarrassed or something that [she] can't talk. We don't see an awful lot (lines 704-708). Well it would be very awkward for them [friends], you know. Very difficult for them and I think they're more scared than anything else (lines 721-722). We had friends, we had lots of friends around that we'd call in and that sort of thing but not anymore really. (lines 275-276)

Some caregivers reported having to judge how much information to share with other people, mainly in an attempt to minimise their worry. Martha, for example, shared her husband's diagnosis with her family, but didn't mention his particularly bad prognosis. This was her way of protecting the family from the knowledge that her husband was unlikely to survive beyond a few months. Martha admitted protecting her relatives from becoming even more upset. However, this decision appeared to gradually isolate her from her family and friends and she reported feeling lonely and unable to talk about her husband's condition to people in her environment.

Emergent Theme 4: Lack of Appreciation

There were few reports regarding how the caregivers' efforts were perceived by the person with MND. Mary was the only participant who reported receiving no empathy and appreciation from her husband. She stated: "He's no empathy at all from my side of the problem. That he's the victim in all this" (lines 1433-1434). Her

husband's attitude appeared to have emotional consequences for Mary, who reported: "I'm beaten down like. Beaten down" (line 738). For Mary, the significant change in her life and the inevitability of coming to the "cruel dead end" was not acknowledged by her husband who appeared focused on his own concerns.

Although the participants experienced a range of feelings and emotions in their role as caregiver (as described throughout this section) they often attempted to supress these in front of the person with MND. This was done in order to protect the person with MND from becoming upset, as illustrated by Kay: "I can't start crying (…) in front of her [mother]. You have to try and bring her up to some level and just kind of keep her. That's what I find the hardest" (lines 413-414). With time, the participants appeared to develop multiple coping skills as they aimed to continue to provide the best possible care for the person with MND.

5.1.4 Subordinate Theme 4: Coping

As the caregivers' lives transformed they developed many coping strategies in order to manage their new life with MND. It emerged that participants generally attempted to cope on their own to avoid upsetting others. Martha stated:

> There's no point in me ringing her [sister], (…) anyway she's so far away (…) you don't like to worry her either.(…) I don't like to worry my mum either because I'll know she'll (upset) worry anyway, so God this is stupid. (lines 1263-1268)

There were four themes which emerged in relation to coping: (1) Avoidance, (2) Positive thinking, (3) Focus on the here and now, and (4) Reciprocal relationship with the person with MND.

Emergent Theme 1: Avoidance

Avoidance emerged as the most frequent coping mechanism expressed by the majority of caregivers. Mary, for example, reported attempting to "go along blindly and say right keep going" (line 461). She also admitted that she "never talk(s) about it [MND]" (line 1586). Martha reported "trying to distract myself with cleaning the house and stupid things, (…) that I wouldn't normally bother so much about, (…) just to keep busy. (…) And then I'm not thinking about things (lines 239-242).

This avoidance could have been associated with many factors. The caregivers were under enormous time pressure and had to fulfil many "concrete" duties, therefore they perceived that they did not have the time to reflect on their own situation. All caregivers, however, understood the progressive nature of MND and that the premature death of their loved ones was imminent. Jackie referred to a "golden time" (line 642), which for her was now, when her husband was still alive. She said: "you've plenty time for crying after" (lines 642-643). It emerged that the caregivers immersed themselves in the needs of the people with MND, whilst neglecting their own needs and avoiding contemplating about their own lives. The caregivers appeared to be aware that reflecting on their situation could evoke emotional distress and increase their frustration, as they were unable to protect the person with MND in the long term.

Emergent Theme 2: Positive Thinking

The participants frequently reported aiming to "get by" (Mary, line 835) their caregiving and the majority (8/10) spoke about the importance of maintaining a positive attitude, for example, Sophia reported:

> I did say to [my husband] from the very start like we're really going to be positive. I said we don't want any doom and gloom now in the house. Everything has to be positive and it's not easy but we have to, yeah and it makes life easier for him too. (lines 276-279)

Maintaining a positive attitude during the "golden time" (Jackie, line 642) when the person with MND was still alive was difficult as the caregivers could not fail to notice the ongoing progression of MND. When asked what advice would she give someone who was just about to become a caregiver of a person with MND Jackie stated:

> And don't be (…) sad over it. (…) you'll cry and cry but generally make the most of it (…). A cousin of mine said to me: I don't want to see you crying. This is your golden time. You enjoy it and she said you've plenty time for crying after (lines 640-643). (…) We just tried to stay positive and we still do but it's harder because Chris [her husband] is deteriorating. (lines 143-144)

Emergent Theme 3: Focus on the Here and Now

Half of participants reported that they were aiming to live in the here and now. Martha, for example, reported that she and her husband had decided to stay focused on the here and now. When seen for a second interview (approximately three months after her husband's diagnosis of MND) she reported:

> We're at a level where we're just taking it a day at a time. If it's a good day we go out (…) making the best of the day and any day that he [husband]'s tired we just go with the flow for that day, do you know? That's what we're doing taking it a day at a time. (lines 570-573)

Sophia also spoke about the importance of remaining in the here and now and not focusing on the future too much. In addition, she stated that although the transformation of life (overarching theme) is unavoidable, a step-by-step approach is best when caregiving for someone with MND.

> I would be very slow to give them [early caregivers] a picture too far down the road. (…) because (…) you're very emotional in the very beginning (…) you just feel you're going to be on tough road and everything is going to change and you just know things are not going to be the same (…) gradually now I know that everything is going to change and I would tell them to be positive. Positive, I think is a huge, a huge thing. (lines 1019-1024)

Maintaining hope was also reported as being important by some caregivers. Although they were aware of the terminal nature of MND, they still hoped for a slow progression rate of the disease ("I just hope he'll stay well", Mary, line 182); that the medications would provide benefits ("I've a feeling that [medication] is working because she [wife] doesn't seem to be getting worse since, Tom, line 931); and that non - oral feeding would prolong the person's life ("I did say to him (…) we want you around as long as possible because we had heard like that it [gastrostomy] will prolong life, you know, at this stage it will prolong life"; Jackie, lines 356-358).

Jack's focus on the here and now meant that he was predominantly concerned with his own illness. Although he understood the progressive and untreatable nature of MND, each time when he reflected on his wife's status he immediately referred to his own health and ability.

> I mean physically like Jesus she'd [his wife] be able to do a lot more than I would be able to do. (…) We were walking (…) about five miles and (…) I

was tired but she wasn't too tired anyhow. So (…) she'd have more energy than I would have for getting around and doing things. (lines 973-979)

Emergent Theme 4: Reciprocal Relationship with the Person with MND

Despite the fact that becoming a caregiver had not been a choice for the participants, they attempted to deliver the best care they could. They appeared to appreciate their relationship with the person with MND and respected the person's past when they were healthy and able to support the family. Those memories appeared to motivate the participants and provide an additional dimension to their caregiving experience. A very special and unique connection emerged between the participants and the person with MND who they cared for. Kay, for example, illustrated how her relied on her for emotional support rather than other family members:

> She [mother] may be a bit upset and dad would be kind of oh my God I can't. She's so used to me now you see. (…) I want Kay she'd say to daddy. She'd write it down and I'd come down (…) I'd be there don't cry, (…) get her a bit, in the right frame of mind again. (..) She doesn't want to be crying in front of my other siblings but she would do it in front of me. (lines 336-346)

Kay's experiences, similar to four of the others caregivers in this study, suggest that there is a reciprocal relationship between the caregiver and the person with MND:

> I can cope better, (…) if her [mother's] mood is, if she's not too bad, but when she'd down (…) you'd be down yourself too. (…) It makes it so difficult (…) when she's down (…) she hasn't the energy to lift her hands, nothing. So if she's a small bit up then you kind of say, she'd say come on, come on, do you

know, you could see it then and that lifts, do you know. I feel it lifts me up then as well like. (lines 392-397)

5.2 Superordinate Theme 2: Transformation of Mealtime Experience

This section focuses on the presentation of superordinate theme 2 and the corresponding subordinate themes which emerged from the data. Each subordinate theme is divided into sub - sections corresponding to the emergent themes. The themes were considered recurrent if the occurred in at least half of the sample. Prior to reporting on the subordinate themes, a table summarising the eating and swallowing status of the people with MND under the care of the participants is presented (Table 5.3). The main aim of this table is to provide a context for understanding the caregivers' experiences related to dysphagia. There were two subordinate themes established during the data analysis process: (1) Eating as a risk of choking, and (2) Focus on safety.

Table 5.3 Dysphagia - related Status of the PwMND under the Care of the Participants

Participant's relative with MND	Oral diet	Non - oral feeding	Modified diet	Able to self-feed	Caregiver required at every meal
Angela's mother	YES	NO	YES	YES	YES
Helen's mother	YES	NO	YES	YES	NO
Jack's wife	YES	NO	YES	YES	NO
Jackie's husband	YES	NO	YES	YES	NO
Kay's mother	NO	YES	N/A	NO	YES
Martha's husband	YES	NO	YES	YES	NO
Mary' husband	YES	NO	YES	INTERMITTENT	YES
Sophia's husband	YES	YES	YES	YES	NO
Tom' wife	NO	YES	N/A	NO	YES
Una's father	YES	YES	YES	YES	NO

5.2.1 Subordinate Theme 1: Eating as a Risk of Choking

As the caregivers experienced progressive dysphagia associated with MND, they began to perceive food as a significant risk, particularly due to their reported fear of choking. This section presents subordinate theme 1: Eating as a risk of choking by reporting on three emergent themes: (1) Visibility of dysphagia, (2) Food as danger, and (3) Emotional impact of dysphagia.

Emergent Theme 1: Visibility of Dysphagia

All caregivers stated that the signs of dysphagia were visible to them. This referred to both the physical symptoms of dysphagia (such as coughing, choking, or prolonged meals), and the transformed attitude of the person with MND towards food (such as low appetite or decreased food-related enjoyment). Helen, for example, described how her father stopped enjoying the food ("he's just lost his appetite") and she observed that he began to "push the food around his plate" (lines 100-102). Jackie reported that even her children noticed their father's dysphagia not only because of his physical symptoms (such as gagging and coughing while eating), but also because the consistency of his food was different to everyone else's:

> Well they [children] can see that [signs of dysphagia]. (…) because Chris [husband] eats slowly number one, and number two he can kind of gag on his food at times and he can get dreadful coughs trying to bring up stuff and they can see that his dinner is soft and that his potatoes are mashed up with butter and they can see. They know like that he finds it hard to swallow and they know he finds hard to swallow and hard to talk. (lines 812-817)

As the disease progressed, all participants began to recognise dysphagia as a problem directly impacting on the safety of the person with MND.

Emergent Theme 2: Food as Danger

All caregivers (with the exception of Jack) reported that dysphagia transformed their own perception of food, in that they began to consider it as a danger. Eating was no longer viewed as a relaxing and pleasurable experience. Instead it was seen as a serious hazard which could cause choking and could potentially lead to the death of the person with MND. Kay reported watching her mother struggle to swallow food: ”Yeah the food was kind of lodging back along (…) you’d be kind of watching was it going down or was it staying and getting stuck.(…) She would start coughing and coughing and coughing” (lines 248-257).

Attempts to monitor the person with MND during meals when eating out were also reported. The caregivers admitted that the experience of eating out had transformed for them (overarching theme), for example, the eating environment was considered as now being important with some participants looking for more spacious and bright venues, and others choosing places with darker and quieter corners. This transformation in relation to eating out was reported regardless of the severity of the person with MND’s dysphagia. For example, Martha reported that her husband’s breathing problems influenced their eating out experience:

> We would (…) go out to lunch really during the day, so and now the big problem is places to go because he doesn’t like confined spaces. (…) Because of the breathing so he likes to be near a window or it’s got to be a big room otherwise he feels like he’s getting claustrophobic. (lines 430-435)

However, the caregivers of people with more significant swallowing impairments (Angela, Helen, Mary and Sophia) additionally reported having to carefully check the menu for suitable food consistencies, which is further reported in section 5.2.2 (Emergent theme 2: Focus on safety).

Emergent Theme 3: Emotional Impact of Dysphagia

The most significant emotional impact of dysphagia emerged as being related to participants' experiences of choking episodes. Choking appeared to evoke a high level of stress and anxiety amongst all caregivers (except Jack), as they felt expected to manage the person with MND during the choking episodes. However, uncertainty and lack of preparation for this role was frequently mentioned, for example, Martha described how fearful and unsure she felt in relation to helping her husband in the event of him choking:

> I suppose that [choking] is a fear in a way now, and the choking episode on my part, what do I do? Call an ambulance? What can they do? I don't know. Will they bring him to [hospital]? I don't know (lines 1183-1185).

Sophia recalled her almost automatic response to her husband's choking episode and reported the need for professional training in the management of choking. She was particularly nervous about the possibility of damaging her husband's gastrostomy tube whilst attempting to assist him in the event of him choking "in case we hurt him or do damage to it [gastrostomy tube]" (line 545).

> Well I actually came behind him and I tried to get it [food, which got stuck in her husband's airway]. I had no experience of it really, but when you're in the situation it just kicks in. (…) You just do it and it [food] came up. (…) I kept

> at him and you're not meant to bend him up. (…). I keep saying I really must go for a course on it (…) because it is a good thing to know. (lines 609-625)

Mary expressed a different perspective on choking although she also perceived food as being dangerous. As she had been caring for her husband for over six years she appeared to accept choking as part of his illness. She reported that she was used to it and did not experience any strong emotions when it occurred. It emerged that Mary had come to the conclusion that there was nothing she could do to help her husband during an episode of choking and she had decided to allow him manage on his own. Mary's adult daughter, however, had a very different perspective despite having witnessed the choking episodes for exactly the same amount of time.

> There could be lumps, he'd choke. He'd kind of choke in it but I'm so used to, you know, when you'd be living with it. I don't panic or and there's not much can do like I feel. (…) She'd [daughter] be roaring. (Laughs). (…) Yeah and I'd be saying look there's nothing I can do about it. (lines 778-782)

Mary's journey as a caregiver was noticeably unique, not only because she had lived with MND for the longest time of all participants, but also because she was the only one who described her husband as "bossy" and not appreciative of her care. Although she was strongly hoping for some change in her life, she was worried that this change may not ever happen. She hoped to maintain good health in order to be able to return to her previous lifestyle following her husband's death:

> Your life is on standby, it's on hold, and I just hope that at the end of the day that I'll have good health myself and get some time on this side of the earth but whether you do or not is another thing, you know, so I don't know." (lines 131-133)

At the subsequent interview Mary again indicated that she would like to "still be alive after him" to "have a good life then". However, she was unsure if this was going to happen. She was concerned that the transformation of her life as a result of her husband's ill - health may have a negative effect on her own health.

> But I just hope at the end of the day that I'll still be alive after him and that I'll have a good life then. Now whether I will or not is another thing because he's living and living and living so I could be gone well ahead of him. (lines 1400-1402)

Mary's ability to remain calm during choking episodes seemed to be due to her almost waiting for destiny to take its course. She was extremely tired and saw no end to her caring duties, as her husband's status appeared to have plateaued after six long years. She perceived his overall health to be better than hers and she reported that "he was living and living and living" (lines 1401-1402) indicating that the end of her caregiving duties may not come in the foreseeable future. She was chronically tired due to continuously disturbed sleep and having to maintain a full time job. Did her husband's choking represent a moment of hope for her? What did she think when he was fighting to catch his breath and her daughter was panicking? For Mary, MND did not seem to be a rapidly progressive disease. It became a chronic condition which changed her life and took away her plans, her time to herself, her sleep, and ultimately some parts of her mental and physical health. In addition, she also didn't feel that her husband appreciated her efforts at all. She perceived him to be "the boss" of the family, despite his severe physical disability and inability to talk. Although her husband was "always demanding" Mary had been able to maintain her independence prior to his ill - health. Now, her husband's expectations regarding her caregiving duties were influenced by

his demanding nature and this was greatly affecting Mary's life. She perceived herself to be a victim of MND too.

> But he was always the boss and he's still the boss. That's the way it stayed. He was always very (…) strong mentally. (…) Just demanding. (…) Yeah he's no empathy at all from my side of the problem. That he's the victim in all this. (lines 1416-1434) (…) The biggest thing that I'd say is that he's very demanding. If he wants a thing you must do it now. (…) Or if I wanted to shove him into bed he mightn't want to go.
>
> *Interviewer: Was he like that before the MND?*
>
> Well I'd never have to do anything for him you see. (…) If I wanted to hop off to bed I'd be gone. It didn't matter (…), but he was always demanding. (lines 1473-1481)

Mary's situation may suggest that attitudes towards dysphagia can potentially change over time. In addition, the relationship between the caregiver and the person with MND receiving care appeared to influence the caregiver's perception and management of dysphagia.

So far, the emotions related to experiences of choking have been reported. The majority of caregivers, however, also reported experiencing feelings of stress, worry, and fear in relation to meal times in general. When Kay reflected on her mother's eating abilities she admitted being "uneasy all the time when she was eating because do you know you'd be kind of watching was it going down or was it staying and getting stuck" (lines 252-253).

The emotional impact of dysphagia appeared to be two - fold. On the one hand, the caregivers recognised food as a potential risk to the life of the person with MND, but on the other hand they were aware of the importance of nutrition to maintaining weight, and weight was perceived as one of the main prognostic factors in MND. In the quotation below Jackie illustrated her efforts to "keep the weight on" which made her "think about calories most of the time". When asked if anything had changed in the way she cooks the food for her husband Jackie reported:

> Well I buy cream and double cream on a very regular basis. (…) for calories (…) I might make the soup but I'd put a lot of cream in Chris' (…) to keep the calories because (…) I was told to maintain his weight. On day one the first thing, one of the Motor Neuron nurses she says just keep the weight on. (lines 213-221). If he's having milk I usually buy the super whole milk (line 225). When I go shopping I think of calories most of the time (line 927).

Participants also reported another self - conscious emotion associated with dysphagia which was embarrassment. Sophia, for example, felt embarrassed when eating out with her husband early on in her caregiving experience. Although Sophia felt uncomfortable and embarrassed when her husband was spitting food, coughing, and experiencing choking episodes in public, she aimed to "carry on as normal" and continued to eat out. She reported making some modifications, such as avoiding crowded places and choosing "quiet corners" or no longer visiting restaurants which could not offer softer food.

> Now the food in the golf courses wouldn't be suitable. They wouldn't cater for soft food so we couldn't eat at the golf course. We'd have to get a place to eat (…). We eat out early most evening because not to have too many people

> in a restaurant if anything happened, you know. People are out for a nice meal and they don't want to see somebody coughing and choking (…). That is the hardest part. I find that is the hardest. (…) I'd be looking quiet corners. (…) I'd go in and look at the menu to make sure they have something on it. (lines 954-965)

Later Sophia admitted that her embarrassment had greatly diminished over time and particularly when her husband began using non - oral feeding. She became less concerned with his nutritional intake and reported feeling more comfortable in restaurants.

Some of the caregivers reported that the person with MND also felt embarrassed while eating out, particularly when they needed full assistance to eat. Mary reported that her husband continued to attempt to feed himself despite it taking much longer. Although Mary was aware of why her husband attempted to self - feed in public, her approach was more practical and she insisted on assisting him to eat in order to prevent the food from getting cold and reduce the spillage of food. Mary reported her lack of patience when feeding her husband and to "putting big spoons up to him" despite being aware that he was unable to manage bigger boluses of food ("he'd be only taking half the spoon).

> I took him out for a meal yesterday and I'd try and feed him then if I could (…) Sundays we go out for a meal as well. So I'd feed him. (…) he tries to eat himself but then I recognised that it isn't going to stay hot so I'd insist on eating. Yeah but he could argue with you. (lines 110-114). (…) I suppose it [feeding] would take ten minutes / a quarter of an hour but I've no patience. I'll be putting big spoons up to him. So he'd be only taking half the spoon.

> I'd be saying, "I can't be here half the day." (Laughs).(…) I'd cut it up but sometimes he wouldn't, em, he wouldn't let you and he'd probably have it cold but eventually I'd catch it and give it to him. (…) Probably embarrassed people looking at him, you know. (lines 825-831)

Mary's reaction to her husband's self - feeding could again be related to her extended caregiving time. She seemed to almost block out the emotional side of eating, feeding, and caring in general. She was immersed in her rigid routines, as this helped her to "get through the now". Mary's caregiving had become almost mechanical and it seemed that she had no energy left to reflect on her emotions.

Guilt was reported by Tom and Kay in relation to eating in front of the person with MND, who was unable to eat any food orally. Tom appeared particularly worried about how he could include his wife in celebrations which involved food. He referred to his wife's dysphagia as a "sentence" indicating the significant impact it had had on him and the whole family.

> She's [his wife] sort of there not able to eat (…). I feel very bad about that. (lines 43-44) (…) There's been a complete change (…) socially as well like (…) we're sort of confined to the house and we can't really go anywhere. We used to go out for the odd meal and that sort of thing at the weekend (…) It's very difficult because like Christmas is coming up and (…) it would be very sad to be asking Gran to come over for Christmas dinner and we all sitting down eating and she can't eat, (…) so we don't know what to… We have to get over that problem (…). I feel guilty when I'm eating too. So it's really a sentence (…). It's very difficult for Orla [his wife]. It's a bit difficult for me too, you know. (lines 550-555)

In addition to the feelings of guilt, Tom described "a complete change" in their social life as a result of dysphagia. It emerged that the caregivers of people with MND who had severe dysphagia (total non - oral feeding) were unable to jointly dine out any longer (Kay and Tom). Maintaining previous eating routines emerged as being important to the participants. Tom, for example, reported how, despite his wife's complete inability to eat, he continued to offer her food as a gesture of love and as a way of preserving their previous normality. The couple appeared to enjoy maintaining special rituals associated with their life pre - MND. In the quotation below Tom compares their old routines, with their new routines which are dictated by the medically recommended feeding regime.

> I suppose it's irrelevant but I bring Orla in a little bit of toast which she doesn't eat. She gives it to the dog because the dog expects it. (Laughs). She's always waiting for it so we have to do it because we've been doing it for years and she's used to it. (lines 146-148)

The feeling of frustration appeared to be shared in this group and it was noticeable that the continuous and unstoppable progression of dysphagia led to the caregivers' being unable to implement any management strategies successfully. Despite their greatest efforts to manage dysphagia a point was reached where only non - oral feeding could ensure adequate nutrition and hydration for the person with MND. Jackie, for example, indicated that diet modifications were no longer sufficient to compensate for her husband's swallowing impairment: "Even if you have everything mashed up (…) it gets caught and it's not easy. It's not easy watching him …(lines 313-315). Kay reported that it was very hard for her when she was "trying to cope with different changes that we would see every day" when "even a boiled egg was getting

difficult to eat" (lines 223-226). She illustrated her attempts to manage each stage of dysphagia by adapting her mother's diet:

> So we'd kind of say bread is out, and then it would be yoghurts, when she was kind of going on to the yoghurts. Yoghurts were going out then because she [mother] was coughing a lot more with those. (…) So there was basically ticking off food saying no, no, no and then it basically came down to… (…) very liquidised potatoes. (lines 268-277)

It emerged that attempting to manage dysphagia was like a battle which could never be won. Despite providing food high in calories and modifying the consistency of food, the caregivers were unable to stabilise the weight of the people with MND or prevent them from choking in the long term. As MND continued to progress so too did dysphagia, deterioration of the respiratory function, and muscle waste. Their frustration was also apparent when the caregivers reported how the people with MND had lost their interest in food. "There is more and more food he doesn't like" said Jackie about her husband (line 270). This greatly affected her ability to increase her husband's weight and potentially prolong his life. Eating was compared to a struggle. For some participants, like Sophia, Tom, and Jackie, it was the biggest struggle they faced in relation to MND. When reflecting on the time before her husband had opted for a gastrostomy Sophia reported:

> The eating was a struggle. Anything else was no problem. He [husband] could go anywhere and we did a lot of walks along cliffs and down to the beaches and we did a lot (…) played golf and no bother. It was just the eating. (lines 952-954)

5.2.2 Subordinate Theme 2: Focus on Safety

Participants maintained their focus on the safety of the person with MND during meals by implementing diet modifications and providing supervision during meals. These strategies appeared to change the caregivers' own approach to food. Participants' focus on safety is explained by presenting three emergent themes: (1) Diet Modifications, (2) Supervision, and (3) Changed approach to food.

Emergent Theme 1: Diet Modifications

Almost all caregivers (apart from Jack) reported that they had had to modify the way they previously prepared food, for example, they began cooking the food for a longer time to soften the consistency. In addition, some types of food, considered by the participants as presenting a high risk of choking, were typically excluded from the diet of a person with MND by the caregiver. The main reason provided for applying these modifications was to ensure the safety of the person with MND. Sophia believed that harder food textures would not be suitable for her husband and that extra cream should be used to moisten his food. When preparing meals at home she appeared able to find a compromise between her traditional method of cooking and the needs in relation to her husband's dysphagia. For example, when baking a pudding she separated some parts for her husband and prepared them in a different way:

> I made bread and butter pudding now and I didn't put any fruit in it because usually I put sultanas or maybe a bit of peel and I did a little bit around the edges for myself because I couldn't give him the edged bits (…), because they were too hard. He was able to eat that as well with cream. (lines 348-351).

Sophia also appeared to pay particular attention to the size of the food particles and aimed to "make sure that he [husband] takes small pieces at a time and just mash a very small piece at a time and then bring over the next piece" (596-598). This strategy was also reported by other caregivers (Kay, Una, Helen, Angela, and Jackie) as being successful in optimising the safety of the person with MND.

Emergent Theme 2: Supervision

The majority of caregivers reported providing close supervision during meals in order to monitor the swallowing ability of the person with MND. In addition, the participants often verbally encouraged the person with MND to, for example, take smaller bites, eat slowly, not overfill the mouth, or to apply specific swallowing techniques. Sophia, for example, demonstrated how she supervised her husband's way of drinking by reminding him: "Make sure you take a drink, take a small sip, take a small sip" (line 853). Interestingly, the supervision was sometimes provided covertly. Helen, for example, attempted to supervise her father discretely while "looking at him without trying to stare" (line 920). She reported closely monitoring the size of the bolus taken by him as she was worried that bigger mouthfuls could cause him to choke. It is possible that Helen's dad may not have been aware of her supervision throughout the meals.

> So in terms of swallowing (…) I'm making conversation and looking at him without trying to stare (laughs) to make sure everything is okay, (…) he's putting a load of stuff on the fork. I'm there going please don't choke on that (laughs). (lines 918-921)

Safety appeared to be the priority for the caregivers, particularly when they perceived food as presenting a high risk for choking. Providing constant supervision during meals was considered to be important and it was implemented regardless of the eating environment. For example, when eating out the participants ensured that the person with MND made appropriate food choices and they were often required to provide an explanation regarding the dysphagia - related needs of the person to restaurant staff. For example, Sophia communicated her husband's swallowing difficulties to restaurant staff and carefully supervised food consistencies to ensure they were appropriate. Sophia demonstrated that she had very good insight into her husband's dysphagia.

> I just say he's a difficult swallow. He had to have a soft diet and they say okay and they're [restaurant staff] probably looking at me like, can he eat? Now I tell what he can eat and (…) and I'd say don't bring any vegetables. Just bring mashed potato and salmon. That will be fine. (…) then he [her husband] would always have a dessert, always. (…) We love desserts. Anything, as long as it's not jelly. I wouldn't give him jelly now because it would melt too fast in his mouth. (lines: 978-988).

Emergent Theme 3: Changed Approach to Food

As the caregivers attempted to manage dysphagia, the meaning of food to them and their approach to food appeared to transform (overarching theme). Their main focus was placed on food consistency in order to optimise the safety of the person with MND. Jackie reported "approach[ing] food differently" as she now had to maintain her focus on choosing food of the appropriate consistency:

> I approach the food differently. I'd always be thinking of what he [her husband] could eat. What would suit him? There would be some things I wouldn't put down. I would always think of what's soft and (…) I'd put the chops into the oven at a low heat for a very long time and I would totally change everything. (lines 894-899)

It appeared that dysphagia also affected previous eating routines within the families. For example, Kay's mother, who was unable to tolerate any oral intake, became upset when someone consumed food in front of her or even mentioned food. This resulted in many changes within the household. Eating and cooking was done almost secretly in an attempt to protect Kay's mother's well - being and food became a prohibited topic in the house. Food, which had once been considered a source of pleasure, became a source of many negative emotions.

> Nobody can eat in front of her. (…) even when I'm cooking here I have to keep that door closed and this door open to let the smell when I'm going cooking because she goes cracked. She starts crying then because she'd go oh I'd love… (…) She'd start crying. I can never eat it, I can't. (…) even if people are (…) kind of saying oh I went out for dinner there and I had this and I had that and she's there… She doesn't want to hear it. (lines 953-962)

The approach to food appeared to transform as the participants developed many new eating routines to help them manage dysphagia in an efficient manner. For example, in Mary's house the schedule in relation to meals was very well established. The family ate chicken and fish on alternate days. Mary decided to follow this routine for many practical reasons, mainly to save the time required to plan what to cook and to shop for food. In addition, by choosing chicken and fish Mary was also able to

control the consistency of the food and its nutritional value. The food and taste preferences of the other members of the family did not appear to be considered. Although Mary appeared to be very satisfied with this solution as she considered having a rigid meal schedule to be very effective, it is unknown how this limited and repetitive diet affected Mary's daughter and husband.

5.3 Superordinate Theme 3: Professional Management of Dysphagia

This section focuses on the participants' reports in relation to the professional input they received to manage dysphagia. It is divided into two subordinate themes: (1) Professional recommendations to manage dysphagia, and (2) Service provision. Each subordinate theme is divided into sub-sections corresponding to the relevant emergent themes.

5.3.1 Subordinate Theme 1: Professional Recommendations to Manage Dysphagia

The caregivers reported receiving various recommendations in relation to managing dysphagia. Four emergent themes were identified: (1) Diet modifications, (2) Non - oral feeding, (3) Adjustment and adherence, and (4) Contradictory recommendations.

Emergent Theme 1: Consistency Modifications

All participants described their experiences in relation to being given the recommendation to alter the consistency of fluids and / or the texture of food for the person with MND as part of dysphagia management at home. It emerged that

modifying the consistency of fluids evoked many emotions within the majority of participants, particularly as it was viewed as having a negative impact on the quality of life of the person with MND.

Modifying the texture of food and the consistency of fluids was reported to greatly transform their appearance, which was reported as being difficult to accept by the people with MND. This resulted in additional stress for the caregivers as they attempted to ensure an optimal level of hydration and nutrition for the person with MND. Therefore being asked by professionals to alter the consistency of the food was reported to be challenging, as it potentially decreased the intake of the person with MND, as they were reported to often refuse to eat modified textures. Kay, for example, described how she attempted to make the food of modified texture look appealing to her mother, however despite her best efforts she felt that it always "looked horrible" like a "horrible slop". Kay also reported her unsuccessful attempts to convince her mother to accept thickened fluids. Kay stated that her mother "hate[d]" these modifications, "got turned off" food and finally "gave up".

> (…) I liquidised food and then of course she [mother] got turned off because it would be slop, (…) potatoes and say fish that I would have poached for her (…) and you'd liquidise it and it would still looked…(…) It looked horrible (…) I was using the thickener for the drinks and she hated those. (…) She absolutely hated them. (…) She just gave up. (…) She didn't like the taste even though (…) I tasted one time and I said it's not too bad. (…) but mam had this thing in her head they were thickeners. (…) And they were going to adjust the taste so she said no. (lines 279-301)

The reluctance of the people with MND to accept thickened liquids was commonly reported. Sophia's husband, for example, opted to stop drinking completely and use his gastrostomy tube to ensure appropriate hydration rather than accept thickened fluids:

> He wouldn't touch it [the thickener]. He just wouldn't. He just couldn't go there. No. (…) I held onto it in case that he might like (…) maybe even a drink of orange or anything (…), but no he doesn't really. He doesn't miss the drinking at all (…) so that's totally gone. You know, they're gone yeah. He doesn't seem to mind that at all (lines 414-429)

The refusal to accept thickened liquids appeared to have emotional consequences for most participants. Jackie blamed herself for her husband's reluctance to accept thickened fluids and she considered adding the thickener covertly, as she knew that the thickener was prescribed to protect her husband from aspirating. As Jackie's main priority was to keep her husband alive, she wanted him to use the thickener to protect his respiratory status.

It emerged, that if the caregivers believed that the recommendation to alter the consistency of fluids or the texture of food would be beneficial for the person with MND, they attempted to negotiate with the person with MND in order to convince them to accept these changes. Kay, for example, attempted to encourage her mother to accept thickened fluids by ensuring her that the thickener was "just a bit of stuff": "She'd [mother] grabbed the thing [thickener], no I don't want that thickener. And I'm there: it's just a bit of stuff, you think it's, it's just a bit thicker than what you normally have it". (lines 305-308).

Technical difficulties, such as problems with mixing the thickener, were also reported by some participants. Una stated: “In cold water it [thickener] doesn’t dissolve so it is manky” (line 46).

Emergent Theme 2: Non - oral Feeding

All caregivers provided some information regarding non - oral feeding and all agreed that the decision regarding the acceptance or refusal of non - oral feeding should be taken by the person with MND themselves, as it involved “their body”. It emerged that there was a shared belief that non - oral feeding could potentially prolong the life of the person with MND. In addition, there was a strong awareness of the risks associated with the gastrostomy procedure itself, and the participants were aware of the relationship between this procedure and the respiratory status of the person of MND. Some caregivers expressed their concern that the person with MND may become too unwell to tolerate the gastrostomy down the line. Jackie appeared very worried and nervous when reporting that they were still waiting for the gastrostomy procedure over six weeks after her husband had finally made this “monumental decision”: “And here we are now six weeks later and after coming to this monumental decision in our lives we’re still waiting. (lines 449-450)

In this study three caregivers (Sophia, Tom, and Kay) were involved in caring for of a person with MND who had undergone a gastrostomy procedure prior to this study. Una’s father had a gastrostomy performed during this research and three other participants (Mary, Martha, and Jack) reported that they were not currently focused on considering non - oral feeding as their loved ones presented with milder dysphagia and were able to eat relatively well. Two other participants (Helen and Angela) rejected

the idea of a non - oral feeding as a possible option for the person with MND under their care. According to Helen her dad was unlikely to experience significant swallow deterioration requiring non - oral supplementation as his health status was already compromised and he was advanced in age. Angela's mother, on the other hand, had clearly declined having a gastrostomy, which Angela reported that she fully accepted. Finally, one participant's (Jackie) husband was engaged in the decision-making process in relation to non - oral feeding at the time of this research.

For Jackie, the decision to accept non - oral feeding was a "monumental decision". She appeared to experience an internal conflict, as she considered the tube as "a good thing" which would "prolong life", therefore she really hoped her husband would consent to it. Although Jackie reported that she respected her husband's choice and tried to avoid pressurising him, she was keen to motivate him to opt for non - oral feeding. She approached a number of professionals hoping to get their support for artificial feeding for her husband, which she hoped might indirectly influence his final decision. However, Jackie reported that she had received many conflicting opinions from the professionals, which made her journey particularly difficult and emotionally draining.

Mary openly reported that "she didn't care whether he [her husband] takes it [gastrostomy] or not" and that this decision belonged to her husband only: "I don't think very deeply about it because I wouldn't be someone like to push him to do things. I say look that's what you want. (…) You've only one life so..." (lines 1269-1271). In addition, Mary perceived the gastrostomy as an additional duty for her and appeared unsure of her own ability to manage it. She stated that gastrostomy tubes involve certain risks, such as infections. These concerns could also have influenced Mary's decision not to attempt to encourage her husband to undergo a gastrostomy.

> He [husband] didn't want it [gastrostomy] and then I didn't care whether he took or not. (…) I felt jeekers what will I be drawing on here because I believe you can get infections (…) you've to be very careful cleaning it, (…) so it would be another job for me. That's the way I saw it. (…) Hygiene must be very, very good with it, you know. Flip it sure I won't half wash it. (lines 1236-1245)

Four caregivers looked after their relative with MND, who had a gastrostomy tube in situ. These participants generally perceived non - oral feeding to be the safest option of managing dysphagia in MND and none expressed any regrets in relation to the person with MND having undergone the gastrostomy. In fact it emerged that non - oral feeding positively influenced these caregivers' sense of hope particularly when some participants (Tom, Kay, and Sophia) reported that they had noticed benefits associated with non - oral feeding, such as increased energy levels and increased weight of the person with MND. The caregivers of the people with MND who were eating non - orally, reported that they were unable to imagine their life without the gastrostomy tube. Kay stated: "Like if you think about it now if she didn't have it [PEG tube], how in the name of God, (…) no it was definitely the right time" (lines 755-756). Tom reported feeling delighted with non - oral feeding, as according to him it saved his wife's life.

In order to manage non - oral feeding at home appropriately, the participants had to acquire new skills, which was often perceived as one of the most difficult duties. Kay, for example, reported: "That's what I find the most difficult is trying to get used to all that medical side. When you're not trained, do you know that's what I find hard" (lines 190-191). For Tom, getting used to the medical terminology was like learning a foreign language and, although he attempted to do it well, he perceived it as generally

overwhelming. When describing his management of non - oral feeding, Tom was noted to experience difficulties remembering the terminology used in relation to the measurement units: "What do you call it… millilitres. Or milligrams! Or… whatever, anyway, or sorry not twelve, twelve hundred so it takes, eh, sixteen hours for that" (lines 166-169). Tom also had difficulties recalling the type of gastrostomy in situ: "The, MIG, the…the… what do you call the little thing?" (line 343). Kay reported her feeling of intense fear in relation to managing the gastrostomy at the beginning, which had improved with time. She highlighted that maintaining an appropriate level of hygiene was crucial to preventing infections. Although Kay admitted that she now felt comfortable managing non - oral feeding, she reported that her father still experienced great difficulty administering his feeds. She described him as a person who has difficulty with "fiddly" tasks as he was more familiar with operating heavy machinery. In addition, Kay stated that her father generally avoided dealing with the medical side of MND. Kay had however gradually managed to train her father in how to administer his medication through the gastrostomy tube which was very important to her, as it allowed her to spend evenings in her own home.

> He [her father] took a bit of training but he got there (…) in the end. (…) He's…(…) a big man like and he'd prefer to be outside underneath a digger do you know. (..) working on a hose on the digger not, do you know, rather than fiddling with a tube but he's after…(…) He picked it up like. (lines 1066-1076).

When reflecting on his gastrostomy-related experience, Tom reported that the establishment of the correct feeding regime had taken a long time and that non - oral feeding had caused many side effects for his wife. It emerged, however, that although some caregivers experienced difficulties at the beginning of non - oral feeding at

home, they managed to adjust to these with time. Moreover, the observed benefits of non - oral feeding appeared to outweigh any difficulties associated with it.

Emergent Theme 3: Adjustment and Adherence

It appeared that adjustment was an important factor for half of participants. Adjusting to progressive MND contributed to ongoing transformation (overarching theme). This adjustment also emerged when considering the participants' approach to the professional recommendations received to manage dysphagia.

The process of adjusting to professional recommendations to manage dysphagia appeared to occur gradually and in parallel with other factors, such as the caregivers' adjustment to the diagnosis of MND overall, their life priorities and their way of coping with their caregiving duties. It emerged that some coping strategies, such as avoidance, negatively influenced caregivers' engagement with professional recommendations. Participants who prioritised quality of life over medial safety were more reluctant to follow some dysphagia management strategies (such as diet alternation or non - oral feeding). The desire to prolong the life of the person with MND appeared to motivate the caregivers to implement the strategies perceived to extend survival, even if they could negatively impact on the quality of life of the person with MND.

It emerged that adequate time was required to adjust to non - oral feeding once it had been professionally recommended and subsequently commenced. Kay appeared to require some time to adjust to the thought of non - oral feeding when her mother underwent a gastrostomy. Initially, Kay reported that she had hoped that the gastrostomy had been performed "just in case" and that perhaps her mother would

never need to avail of it. Gradually, Kay realised that the progression of dysphagia was unavoidable and irreversible. When Kay reflected on her experiences with non - oral feeding, she reported having to psychologically adjust to accepting it as part of her and her mother's lives:

> Psychologically, that she [her mother] has to have this [non - oral feeding], the headset at that time, it was just in case. But sure… Only in your head you're kind of saying oh it's just in case but when that day actually came that you knew it was, this is it now that you have to use that tube for as long as we can, that was the hard part trying to get that around. (…) That adjustment, do you know, because when she first got it I thought (…) it's down the line. You'd be thinking oh its way down the line we're going to have to use it but as soon as she got it, it was only a couple of months like that we were using it like. (lines 206-216)

Caregivers' own opinion with regard to the appropriateness of a particular dysphagia management strategy may have been a factor which influenced their level of adherence to this recommendation at home. In this study, one participant (Angela) reported that she had been given professional recommendations in relation to altering the consistency of her mother's food, which she perceived as inappropriate and unnecessary for the mother: "They [professionals] were just telling (…) to mash stuff and chop it up and you know, blend it. You could blend it but she's [her mother] not at the blending stage or anything like that" (lines 736-738). Angela chose not to follow this recommendation and reported that her mother had been managing well on a regular diet. Sophia reported "We decided [to undergo a gastrostomy] because I felt he [husband] was losing weight" (line 270) implying that although it was a joint

decision, it was based on her opinion regarding her husband's inability to maintain his weight.

Emergent Theme 4: Contradictory Recommendations

All caregivers attended more than one dysphagia - related service at some stage in their journey, which sometimes led to them obtaining contradictory professional recommendations, as indicated by Jackie and Tom. Jackie, for example, consulted many professionals when attempting to decide if non - oral feeding would be necessary for her husband. This left her feeling confused due to hearing contradictory opinions in relation to the overall benefits of non - oral feeding in MND and the safety of the gastrostomy procedure. When reflecting on the process of meeting various professionals in relation to non - oral feeding Jackie reported:

> Well it depends who you speak to. Some say it's very straightforward [gastrostomy procedure], others say it's not at all straightforward. One person says it's easy to get a chest infection from a tube as it is to get from swallowing. So I don't know. I don't know really. (lines: 422-428)

When Jackie's husband decided to opt for non - oral feeding Jackie reported feeling very pleased as she hoped it would prolong his life. However, soon after his decision was made the couple met with a professional who clearly stated that non - oral feeding would not bring about any of the expected benefits (including the expansion of the life span). Instead, they were informed that the commencement of non - oral feeding at Jackie's husband stage of MND could create many ethical issues. Jackie reported feeling devastated following this consultation, particularly as she was aware that if her

husband's respiratory status deteriorated further he may not be well enough to survive the gastrostomy procedure: "I worry sometimes if he is still alright to get it [non - oral feeding] but I hope he is" (line 442). This consultation appeared to have been very stressful for Jackie and it was observed that she had great difficulty speaking about it. She was devastated at having experienced a difference in professional opinion, particularly once her husband had finally consented to a gastrostomy.

> Yesterday he [a doctor] said it will not prolong life. (…). That was kind of devastating (short pause). I don't know. Like we have, as I said we've gone up to [names place] about it. We have looked it up. We have spoken to a nurse in [names town] who deals with a lot of PEGs in [name] Hospital. (…) It was only yesterday. It's only sinking in. (lines: 52 - 88)

Tom experienced contradictory recommendations in relation to the therapy for his wife's dysphagia. It was recommended that she carry out swallow strengthening exercises by one therapist, however, another therapist advised her to cease the exercise programme: "She [SLT] gave her [Tom's wife] a chart with exercises to do for her mouth and she's actually a speech therapist I think. (…) And they told us not to do that in [names a service]". (lines 533-536)

5.3.2 Subordinate Theme 2: Service Provision

It emerged that the dysphagia - related services were typically provided by a number of different professionals, predominantly SLTs, but also Dietitians, Neurologists, and Gastroenterologists. Participants' experiences of services received to manage dysphagia varied. One of the main factors identified, which appeared to

greatly influence their experiences of dysphagia - related services, was the geographical location of the participants as this determined their access to these services.

All participants engaged with more than one service in relation to dysphagia and the majority (7/10) attended dysphagia - related services in both MND Clinics and in the community. The remaining participants (3/10) availed of community and palliative care services. Attending more than one service appeared to be more frequent at the earlier stages of MND and as the disease progressed the participants typically engaged with one service only.

Six emergent themes were identified in relation to participants' experiences of dysphagia - related services: (1) Accessibility, (2) MND clinic, (3) Involvement of the caregiver, (4) Professional approach, (5) Domiciliary service, and (6) Training.

Emergent Theme 1: Accessibility

The majority of participants reported being generally satisfied with their access to dysphagia - related services. "(…) she [SLT] was there if you ever have any problem just ring me up with anything" reported Kay (line 637) indicating her satisfaction with access to SLT. The caregivers reported that they appreciated having the mobile phone numbers of the leading professionals as this provided an increased sense of security. It emerged that this sense of security was particularly important for the caregivers of people with MND who were receiving non - oral feeding, as problems related to the gastrostomy tube (such as a blockage, leaking or dislodgement) were always perceived by the participants as an emergency. Moreover, as transport was reported as being an issue especially at the later stages of MND, the caregivers greatly appreciated having

the option of accessing indirect support. It needs to be acknowledged, however, that despite difficulties related to travelling, the caregivers willingly attended professional consultations if they considered them beneficial for the person with MND.

Angela's experiences in relation to dysphagia - related service accessibility and efficiency appeared to be unique in comparison with other participants. Transporting her mother to see a professional was particularly difficult as the family lacked any special equipment to make the transfers more comfortable and safe for all. In addition, Angela's mother was reluctant to leave the house and often refused professional help. Angela had difficulties recalling their last dysphagia - related appointment and reported that it had occurred a few years previously. She observed that her mother's ability to swallow had deteriorated since that last appointment, however, she did not perceive there to be any professional help available in the absence of domiciliary services:

> I mean for someone like mum no one is willing to come out, no one wants to come here to see her. They want her to go like to go there. (…) even in the palliative care unit there isn't anyone like that.(…) no one will come out to see her and the way she is she doesn't want to go in which is difficult so she just doesn't get seen by anyone really. (lines 722-728)

Emergent Theme 2: MND Clinic

The majority of participants experienced MND clinic as part of dysphagia management and most reported their satisfaction with this form of service provision. It emerged that having an opportunity to meet various professionals on the same day was perceived as a time saving option. Mary reported: "So you see all the services in

the one day so that's good." (line 478). However, some participants reported their dislike of the clinics, as they found meeting other people with MND at various different stages of the disease to be disturbing and distressing. Kay, for example, reported how the clinic provoked her to compare her mother's status to that of other people with MND:

> I don't particularly like it [MND clinic] (…) Because that is basically what you are doing. You are looking around at everybody that comes in. You're there oh my God they sound like mam or (…) they worse than mam, and you're walking so you're not. (…) Like sometimes I nearly prefer to say if you had an appointment and bypass all that and just straight into the team and that's it. (lines 561-570)

Jack indicated his experience of delays in the MND clinic:

> Well we're going back there [MND clinic] again in February like, (…) you'd hope that (…) that whatever has to be done would be done quicker in that and not the same amount of hanging around, you know. (lines 305-308)

Emergent Theme 3: Involvement of the Caregiver

Dysphagia - related services involved the caregivers being seen jointly with the person with MND. However, over the half of the caregivers in this study reported the potential benefits of availing of individual appointments. The main reason for this related to raising questions about the status of the person with MND (particularly in relation to the prognosis), which the caregivers reportedly found uncomfortable in the presence of the person with MND. Therefore, the participants admitted that they often refrained from asking questions, which could potentially have a negative impact on

their caregiving and on their ability to be adequately prepared for the upcoming change in the status of the person with MND. Instead, some of the caregivers reported that they carefully observed the professionals during appointments in an attempt to gleam any indication of further deterioration in the status of the person with MND. Sophia reported how she related all information received to her husband: "Somebody might say (…) does he [husband] get any chest infection? I just say to myself, well that's coming, eventually that will come. He will get chest infections." (lines 744-747)

Some caregivers (4/10) reported that their perceptions of the health status and abilities of the person with MND differed from the individual's own perceptions. Therefore separate appointments for the caregivers were perceived as providing a potential opportunity to clarify the information provided by the person with MND and provide further insight into the issues experienced at home. For example, Sophia suggested potential changes to the way the services are currently provided:

> I think if a partner could get to talk they [professionals] might see a different side. (…) you don't want to make it worse but you'd like to give the truth. (…) The truth is all you need really. (…) Well you won't get to the next stage fast enough I feel if you don't give the proper information (…). Paul [husband] may not have had the PEG maybe (…). They'd think he was fine, you know. (…) whereas he would have been losing weight. (lines 1025-1153)

Emergent Theme 4: Professional Approach

A few issues arose in relation to how the professionals were perceived to approach the caregivers as part of the dysphagia - related service. The issue of timing and

consideration for caregivers' level of adjustment to MND emerged as being important for the majority of caregivers.

It emerged that when professionals were perceived as being intrusive it left a long lasting negative impression and affected the caregivers' willingness to engage with the service again. Martha reported being asked to consider non - oral feeding for her husband at their first visit to a palliative care centre. Martha reported that a SLT attempted to convey that this decision needed to be taken soon due to husband's poor respiratory status. Martha described this SLT as being inappropriate and intrusive. She reported being pushed into making an important decision while both she and her husband had not had adequate time to adjust to the diagnosis of MND.

> Before we left we saw a speech therapist (…). I just wanted her to go away because (…) she was talking about the future and if you can't swallow you have to think about having (…) the PEG feeding and all of this and I'm just like: go away. I just thought we don't need it, do you know? (lines 222-230). (…) She [SLT] was going on about (…) the fact she said you can't leave it. She was quite forceful in that she was sort of saying you can't leave it. You have to make this decision because you're breathing is bad and you won't go through an operation having the tube fitted (…) so don't leave it to the last minute. You need to make a decision. (lines 1057-1061)

Over half of participants reported being overwhelmed with the level of information provided shortly after the diagnosis of MND, especially as they were only attempting to adjust to this disease. The preferred timing of professional intervention appeared to be related to the caregivers' adjustment phase. Sophia stated:

> You're very emotional in the very beginning because (…) you just feel you're going to be on tough road and everything is going to change. And you just know things are not going to be the same (…) gradually (…) everything is going to change. (lines 1031-1035)

Martha appeared much calmer at her second interview, which was carried out just over two weeks following the initial one. She reported feeling more adjusted to her husband's diagnosis and able to talk about MND. She reflected on the previously met SLT and suggested what input would have been more appropriate. Martha would have preferred not to have been approached by an SLT at the early stages post - diagnosis, unless it was just for a brief introduction. She was not ready to hear about a gastrostomy and the expected deterioration of the swallowing. She was also focused on protecting her husband from hearing information regarding his prognosis; especially considering he had absolutely no knowledge of MND. The initial contact from the SLT was unscheduled and therefore unexpected, meaning that the couple were not given the opportunity to prepare for this consultation. When approached by a different SLT a month later Martha appeared more satisfied, as she had had more time to adjust to having to live with MND and this SLT was reported to utilise a gradual approach when providing information about dysphagia.

> I just felt that she [first SLT] didn't need to happen (…) at that particular time, because it may have been hi, I'm the speech lady from palliative care and I will be seeing you at some point would have been enough. (…) I think she could have just introduced herself if she wanted to, or not even bother at all. I think the lady [SLT] today was just about the right time, doing the baseline, explaining everything. She's also going to post on loads of information about warning signs, (…) to look out for just in case any weakness starts to happen

> and I think that's all we needed. (…) I think it was good. It is good but not to go into details of PEG feeding. I don't think that was needed yet.(…) I think if things do start to deteriorate, (…) then maybe we need the PEG feeding talk. (lines 1297-1328)

Some participants reported that they had experienced professionals being disrespectful towards the person with MND, ignoring the preferences of the person with MND, and appearing unable to adapt their communication style to engage with the person with MND. This was emotionally upsetting for the caregivers and was perceived as a lack of sensitivity and understanding of people living with MND. Una became quite animated and she raised her voice when describing a visit from a therapist, who she reported to "hammer the powder [thickener] to dad". Although Una understood the rationale for using the thickener, she aimed to respect her father's choices and quality of life priorities. For her, the recommendation to thicken fluids impacted on her father's quality of life as he did not appear to enjoy it. Una appeared unhappy that the physiotherapist criticised her father for taking a drink without a thickener on a particularly difficult day, when he had only just returned home from being in hospital.

> No offence to Adam [physiotherapist], but the day he came I went oh now here, enough. Because it was the same hammering of the powder to dad and he had a full day in [hospital] and I just felt he could do without it for one day, that bloody powder. Sorry. (lines 56-59)

It appeared that the caregivers valued when the professionals provided information in an accessible format and when they spent time explaining about dysphagia. In addition, the participants did not appreciate when they felt pressurised

by a professional to follow a specific dysphagia - related recommendation. Instead, they reported that they preferred being offered the choice of accepting or declining a recommendation. When Kay expressed her satisfaction with dysphagia - related services she also explained what she valued most: feeling comfortable and not forced to follow professional recommendations; being well supported and informed; and having good access to the service (being able to ring for help).

> I think the lads above [names clinic] are fantastic (…). They give you advice. (…) it's up to you whether you take it on board (…) they're not forcing you. They don't say you have to… You have to do this. (…) I feel comfortable with that situation and if there's a problem you ring for the help. (..) That's what I prefer about it (laughs). (lines 600-612).

Emergent Theme 5: Domiciliary Service

Two participants referred to the domiciliary service related to dysphagia and both highlighted different issues associated with it. Tom reported that having many professionals visiting his home disturbed his sense of privacy, although he understood the importance of these visits. In the quotation below Tom indicated his wish to be given appropriate notice in relation to home visits in order to give him sufficient time to prepare.

> Actually there's very few mornings that we don't have somebody coming (lines 423-424). (…) It was very quiet here (…), but now we don't know when there's somebody coming in the door (…) it's okay once we know, we can handle it then. We know it's for the good of our problem. (lines 456-460)

Angela, on the other hand, expressed her strong dissatisfaction with the lack of domiciliary services for her mother, which was already reported under Emergent theme 1: Accessibility. Angela perceived that her mother was deprived of necessary support, as she refused to leave the house and had no access to domiciliary care.

> They're [professionals] not coming out to check her throat or they're not coming out to see if there's anything they can give her (…) Why can't any of these services be provided in the home for someone that won't go out of the home? (…) There hasn't been anyone for a long-time. (…) When would she [an occupational therapist] have last been here though? (…) That's three years ago since she was here. (lines 862-887)

Emergent Theme 6: Training

One participant (Jackie) reported attending specific training related to the management of dysphagia in MND. However, she was dissatisfied with this training for a number of reasons. Firstly, she reported that she had not gained any new knowledge from the training. Also, she reported feeling isolated as she was unable to identify with the other caregivers at the training, as they cared for people with MND who were much older than Jackie's husband and were at more advanced stages of the disease. Hearing their experiences appeared to deeply upset Jackie. She referred to the training as a "waste of time" and time was particularly significant for her. During the interviews she referred to the time spent with her husband as "golden" and she aimed to appreciate every minute they could spend together.

I felt I hadn't learnt much new there because I had been (…) with [husband] the previous week or two [in speech therapy]. (…) I just felt I hadn't much in common with them [other caregivers]. (…) I suppose every case is just so different. I just felt coming home that was just day I didn't get a lot out of it. (…) and I just came home upset. (…) I was there that day now without Chris and I didn't feel any better. (…) I felt it was a waste of my time being away from Chris to be honest. (…) Yeah, next question (laughs). (lines 1071-1096)

5.4 Chapter Summary

The overarching theme revealed through the process of IPA was transformation. This transformation was evident in various aspects of caregivers' lives including dysphagia. The participants immersed themselves in the continuous process of caregiving, which affected them both physically and emotionally. Caregiving was reported to be a continuous duty and compared to a full time job. The participants appeared to be constantly concerned with the needs of the people with MND, and therefore unable to take a break from their caregiving duties. The participants described their attempts to find and maintain the right balance in various aspects of their lives, such as balancing medical safety with quality of life considerations, attempting to be positive while witnessing the ongoing deterioration in the person with MND, and balancing caregiving duties with protecting their own needs. The transformation of participants' lives and their ongoing balancing was metaphorically compared to an act of slacklining.

Time appeared to be important when considering the caregivers' journey. It emerged that once the participants came through the initial phase of shock and panic,

they attempted to adjust to living with MND by developing various coping strategies. Having to cope appeared to be perceived almost as a necessity, with no alternative available. The caregivers had to accept the transformation of their lives and focus on the person with MND in order to adapt their level of care to the progressive nature of MND.

Participants' experiences of dysphagia and their attempts to manage dysphagia at home were reported to have an emotional impact on them. In order to optimise the safety of the person with MND the caregivers reported altering their previous ways of preparing food, and implementing various adaptations to the diet to minimise the risk of choking. Constant supervision was usually provided during meals regardless of the eating environment. It was noted that participants' approach to food transformed and they seemed to be predominantly focused on appropriate food consistencies and nutritional values. The establishment of safer eating practices or feeding routines appeared to be considered as a priority when managing dysphagia as part of MND.

Participants were generally satisfied with the services they received for dysphagia; however, some suggested a desire to avail of individual consultations. The caregivers in this study wished for the professional input to be guided by their current ability to take new information on board.

Non - oral feeding appeared to be associated with many positive emotions and the participants perceived it as the best option for managing severe dysphagia. In contrast, modifications of food and liquid consistencies were related to significant levels of stress and not perceived to have a positive impact on the quality of life of the person with MND.

Finally, Sophia compared her caregiving journey to having a new baby: “It’s like having a child, people would be telling you different things and when you have a child you just have to go through the different stages, you know, yourself, yeah” (lines 1038-1040). The stages experienced by the caregivers were related to the stages of MND progression, and also to the stages of coping and the development of new routines. This appeared to also apply to participants preferences in relation to dysphagia - related services. “People would be telling you different things” – this could be compared to hearing various professional advice. However, as the caregivers had “to go through different stages” themselves, they valued a gradual approach and being provided with professional recommendations without feeling forced to follow them.

CHAPTER 6: DISCUSSION

Introduction

This study aimed to investigate the experiences of dysphagia in MND from the perspectives of both people diagnosed with MND and caregivers of people with MND. In order to answer the main research question: *How do people living with MND*[16] *understand and experience dysphagia?* multiple interviews were completed with the two groups of participants: people diagnosed with MND and caregivers of people with MND. Data analysis from a total of 20 participants (10 in each group), through the process of IPA, revealed one overarching theme in each group: Living in the here and now (people with MND) and Transformation (caregivers). Three superordinate themes emerged in each group. The first superordinate theme for the people with MND was: MND as a life changing disease, and for the caregivers: Immersion in the caregiving as an act of slacklining[17]. Both superordinate themes (as named above) related to living with MND in general and were not specific to dysphagia. They were, however, included in the data analysis as they represented the issues perceived as important by the participants and reflected what the participants were really interested in talking about. The two subsequent superordinate themes, for both groups, were focused on dysphagia. For the people with MND superordinate theme two was: Living with dysphagia, and superordinate theme three: Professional management of dysphagia. In the group of caregivers' superordinate theme two was: Transformation of mealtime experience, and superordinate theme three: Professional management of dysphagia.

[16] The term: "People living with MND" refers to both people diagnosed with MND and their caregivers throughout this chapter

[17] Slacklining - walking or balancing along a suspended length of flat webbing that is tensioned between two anchors and bounces almost like a long and narrow trampoline (Utton, 2015)

Cross - case data analysis, carried out separately for each group, revealed that the experiences of dysphagia were strongly embedded in how the participants' approached and coped with MND in general and therefore could not be investigated in isolation. This finding may not be surprising considering that dysphagia is rarely the only symptom of MND. In the bulbar onset of the disease, speech and swallowing are affected together (Grocher & Crary 2010, Luchesi et al., 2014; Ruoppolo, et al., 2013). In the spinal onset, the limbs are typically more affected before a person develops any signs of a swallowing impairment (Kuncl, 2002). All participants in this study had to cope with other concomitant disabilities in addition to dysphagia.

The people with MND and the caregivers in this study approached and managed dysphagia in a different manner and dysphagia appeared to affect each group differently. This chapter will discuss both perspectives in relation to the wider literature. This discussion is divided into six main sections: (1) The symbiotic relationship between the person with MND and their caregiver, (2) Was dysphagia an issue for people living with MND? (3) How people living with MND wished to manage dysphagia, (4) The impact of dysphagia, (5) Professional management of dysphagia and (6) Reflections for professionals providing dysphagia - related services in MND. The above sections were selected for discussion to highlight the main findings of this study, as interpreted by the researcher.

6.1 The Symbiotic Relationship between People with MND and their Caregivers

This section focuses on the unique symbiotic relationship, which was apparent between the people with MND and their caregivers. Symbiosis is generally understood as a mutually beneficial exchange between two beings (Fitremann, 2017). This

symbiosis emerged for various aspects of participants' lives. For example, all people with MND aimed to continue their engagement in previous routines which they considered to be important, such as routines carried out to support the family. Foley et al. (2013) previously reported the reciprocal nature of family caregiving in MND and noted that people with MND take their families into consideration when making decisions about their own care. Another Irish study (Foley, 2016) highlighted the strong sense of responsibility people with MND feel towards their families. However, the current study indicated that maintaining previous routines (including those carried out to support the family) was usually only possible due to adaptations made by the caregivers. For example, the caregivers ensured adequate access to technology or other assistive equipment, which was necessary to enable the person with MND to carry out a specific routine.

It strongly emerged in the current study, that people with MND and their caregivers were emotionally bounded together. The psychological well-being of the participants with MND appeared to affect their caregivers' mood and ability to cope with their burden of care, which has also been recognised in other studies in MND (Chiò et al., 2005; Boerner & Mock, 2012; Jenkinson et al., 2000; Olsson et al., 2010a; Pagnini et al., 2011; Peters et al., 2012; Rabkin, Albert, Rowland, & Mitsumoto, 2009). In addition, one caregiver in the current study reported a strong connection between her mood and her perceived lack of appreciation from the person with MND to whom she provided care. This is also consistent with the findings of Rabkin et al. (2009) and Boerner and Mock (2012) who investigated the factors which influence the mood and psychological wellbeing of caregivers of people with MND. Both studies found a positive association between caregivers' mood and how supportive the person with MND was towards them. This current study also demonstrated that the attitude

of the person with MND towards the care received from a caregiver could have a strong effect on the well - being of this caregiver.

Interestingly, Parker (1993) highlighted the importance of the quality of the relationship between the caregiver and the person with MND prior to the disease when considering their ability to cope with the strain of caregiving following diagnosis. This was also observed in the current study where the majority of caregivers expressed their gratitude in relation to what the person with MND had done for them prior to their ill - health. This appeared to add meaning to their caregiving, helping them somewhat in their role as caregiver. One participant, however, expressed some negative emotions and experiences related to her relationship with her husband pre-MND, which also seemed to shape her entire caregiving journey. The current research demonstrated that when investigating the relationship between people with MND and their caregivers, the nature of their relationship pre-MND should also be considered.

6.2 Was Dysphagia an Issue for People Living with MND?

Dysphagia was not perceived as an issue by every participant in this study (in both groups). It emerged that the people with MND appeared to be generally less concerned about their swallowing impairment than their caregivers. Their level of concern did seem to increase, however, at the later stages of dysphagia when choking episodes became more frequent, maintaining an adequate oral diet more challenging, and self – management strategies were not sufficient anymore. Although the caregivers did express their concerns in relation to dysphagia, they also spoke about other aspects of living with MND which they found more challenging, for example, the deterioration of breathing.

The people with MND in the current study often denied experiencing difficulties with eating, drinking, and swallowing during the data collection process despite clear evidence that they did have dysphagia (such as observed coughing during meals or according to caregivers' reports). It emerged that the symptoms of dysphagia were perceived as manageable when compared to other impairments caused by MND, therefore for many they were not worth labelling and discussing in - depth. In fact, dysphagia was not reported as being the primary concern by any participant with MND in this study. Dysphagia was not perceived as an issue until it became impossible to self-manage and control. Therefore, for the people with MND in the current study, similar to the people with head and neck cancer in the Tong et al. (2011) study, the presence of a gastrostomy tube was the factor which ultimately led to them admitting that they had dysphagia.

In contrast to the people with MND in this study, the caregivers had no difficulty acknowledging the presence of dysphagia during the data collection process, as they found the symptoms to be visible and obvious, which again confirms that the two groups had different perceptions of dysphagia. The caregivers considered dysphagia as having the potential to cause the death of the person with MND. Moreover, the fear of health complications related to dysphagia was reported as increasing the burden of care, which is in agreement with Trail et al. (2004), who distinguished dysphagia to be one of the three main stressors for the caregivers of people with MND, along with worries about MND progression and worries about emotional and physical well - being of a person with MND. The caregivers in the current study aimed to ensure the safety of the person with MND by providing constant supervision during meals and applying various compensatory techniques, both self - developed and professionally recommended. In order to avoid causing the person with

MND additional distress, the caregivers reportedly refrained from discussing dysphagia with them and often provided discreet supervision or covertly implemented dysphagia management techniques, such as cutting food into smaller pieces or adding small amounts of thickening powder to the liquids prepared for the person with MND.

It emerged that the caregivers who had previous knowledge of dysphagia, were more concerned about the presence of dysphagia in comparison to those caregivers who had not. This appeared to be related to having a more advanced awareness of the health - related consequences of dysphagia, such as chest infections or choking. This finding is potentially significant for clinical practice, especially that caregivers' training and education is understood as important when aiming to increase patients' compliance with dysphagia recommendations (King & Ligman, 2011). The literature concerned with researching connections between patients' / caregivers' knowledge and their level of anxiety appears inconclusive (no studies including people living with MND were found). For example, Zhang, Liao, Liao, Wu, Wan, Wang, & Ma (2014) investigated over 360 people with Chronic Obstructive Pulmonary Disease [COPD] and stated that the lower level of knowledge of COPD increased anxiety. Contrary, Emery, Schein, Hauck, & MacIntyre (1998) in their randomised trial did not find benefits of education in reducing anxiety for people with COPD. The findings of current study indicated that professionals delivering dysphagia - related services should be aware that dysphagia education may potentially increase the level of caregivers' distress.

Adelman et al. (2004) previously reported that people with MND and their caregivers differed in relation to their perceptions of their level of distress and burden of care. In their study, for example, people with MND rated the caregivers' burden higher than the caregivers themselves did, while the caregivers perceived the people

with MND to experience a greater level of fatigue and discomfort than the people with MND themselves indicated. Trail et al. (2003) researched the major stressors for people with MND and their caregivers and observed that caregivers were more concerned about the swallowing ability of the person with MND than the person with MND was. The current study also confirms a disparity between the perceptions of people with MND and their caregivers in relation to the severity of dysphagia. The subsequent sections provide further insight into the perceptions of both groups.

6.2.1 What People Living with MND Wished to Talk about - People Diagnosed with MND

Interviewing people with MND in relation to their experiences of dysphagia was not straightforward, as the majority of participants in the current study denied having dysphagia. This minimising of one's own dysphagia was also reported in a population of people who had experienced a cerebrovascular accident (Klinke et al., 2013). Klinke et al. (2013) observed that their participants were overwhelmed by the impact of the cerebrovascular accident and therefore did not consider their swallowing difficulties to be significant ("Eating difficulties is just a part of the whole package", pg. 257). Similar attitudes emerged in the current study, where the people with MND avoided focusing on one particular aspect of their disease, such as dysphagia. In another study investigating the experiences of dysphagia in head and neck cancer (Tong et al., 2011), participants did not remain focused on the topic of dysphagia either. Instead, they wished to talk about their primary concern which was the re - occurrence of cancer. It would appear that the underlying cause of dysphagia (such as a cerebrovascular accident, cancer, or MND) may be the predominant concern for some people, rather than the presence of dysphagia itself. Moreover, if people are

experiencing multiple comorbidities associated with their illness, along with the prospect of an early death, they may not perceive dysphagia as their priority at all. In order to provide more insight into this phenomenon it is important to consider how people with dysphagia approach and cope with their primary disease in general, rather than focusing solely on dysphagia.

The people with MND in this study frequently redirected the conversation in order to express their concerns with regard to the ongoing deterioration caused by the disease, the loss of their independence and various skills. It emerged that losing their voice and ability to speak was frequently reported as being more of a concern to participants than dysphagia, as it appeared to have much more of a profound impact on their lives. Communication impairment (dysarthria) has already been identified as one of the most significant stressors by people with MND, along with muscle weakness and worry about how the progression of the disease will impact on their independence (Trail et al., 2003). Additionally, in the current study dysarthria and dysphonia were reported as being impossible to conceal. Some participants also reported their reluctance to use an AAC device. Although the current study did not investigate the barriers for using AAC devices, Murphy (2004) identified two main issues with accepting an AAC device in MND: the perceived need for social closeness (which can be impaired by the use of a device), and technical difficulties related to using the device itself.

The people with MND often commented on their changed flow of life and each person recalled the experience of receiving the diagnosis of MND. This appeared to be an extremely traumatic time, in line with Hogden, Greenfield, Nugus, and Kiernan (2012b), especially considering that there is no cure for MND (Ferguson & Elmann, 2007). However, despite the negative prognosis and inability to control the trajectory

of MND, some participants conveyed that they maintained a positive attitude. The importance of positive thinking on person's mental well - being has already been established (Hu, Zhang, Wang, Mistry, Ran, & Wang, 2014; Nowlan, Wuthrich, & Rapee, 2014), and has previously been reported in the literature concerned with terminal illness (Gum & Snyder, 2004; Johnson, 2007; Youll & Meekosha, 2013). In the current study, having a positive approach to life appeared to contribute to the people' with MND attempts to remain independent, continue to support their family, and apply a problem - solving approach to dealing with the symptoms of their disease. Although the trajectory of MND was understood as being outside of participants' control, they perceived that they had a choice regarding how to live and cope with MND. As maintaining their engagement in previous meaningful routines emerged as a priority for every person with MND in the current study, participants strived to find solutions to manage their MND - related problems in order to continue their participation in various routines and maintain their independence. This suggests that they maintained a pragmatic approach to dealing with their own illness. At the same time however, the overarching theme (Living in the here and now) implies that once a solution to a particular problem had been identified, the problem became part of their past, something not worth reminiscing about. The people with MND in the current research wished to talk about the present and any conversation pertaining to the future or past appeared to evoke emotional distress. There is evidence that focusing on life in the present moment rather than the future can lessen the feeling of stress in MND and promote better mental well - being (Centers, 2001).

6.2.2 What People Living with MND Wished to Talk About - The Caregivers

The caregivers in this study frequently spoke about feeling under enormous time pressure due to juggling their caring duties with other work and family commitments, and they perceived having no break from their caregiving duties. The reports regarding the average amount of time spent caregiving per day in MND vary, ranging from 5 hours (Chiò et al., 2006b), 11 hours (Krivickas, Shockley, & Mitsumoto, 1997), 13 hours (Alankaya & Karadakovan, 2015), to even 15 hrs in some studies (Chiò et al., 2006b); and this time is generally reported to increase as MND progresses (Aoun et al., 2012). Interestingly, this average time spent caregiving in MND is greater than the caregiving time established for Alzheimer's disease (Davis et al., 1997) or following a cerebrovascular accident (Hickenbottom et al., 2002). In the current study it emerged that caregiving duties extended well beyond the provision of direct care and in order to gain a more thorough understanding of the amount of time caregivers spend on their caring duties in MND, we must look beyond their direct contact with the person with MND and also consider their stress and strain. Worry and tension appeared to be a constant in the caregivers' lives and seemed to be related to the unpredictability of life with MND in general, similarly to the findings of other studies (Galvin et al., 2016; Ray & Street, 2006; 2007; Trail et al, 2003). In the current study, psychological distress was reported even when the caregivers were away from the person with MND, therefore no "real" breaks from caregiving were perceived as being possible.

The current research, in line with other studies on caregiving in MND (Adelman et al., 2004; Chiò et al., 2005; Jenkinson, Fitzpatrick, Swash, & Peto, 2000; Ray & Street, 2006; 2007), indicated that the caregivers lived under enormous pressure

and experienced significant fatigue leading to inability to undertake other activities (outside work and caregiving duties). The majority of caregivers in the current study reported coping by avoiding self - reflection and suppressing their own feelings, which was often done in order to fully immerse themselves in the needs of the person with MND. Half of the group attempted to maintain a positive attitude and stay focused on the here and now, as MND disabled them from making future plans, which also emerged in Ray & Street (2007). All of the caregivers in the current study provided evidence of their impaired psychosocial and emotional well - being resulting from providing care to a family member with MND with the feeling of frustration being particularly strong. Although caregivers' frustration was previously reported by Galvin et al. (2016) and Ray and Street (2006), this frustration was predominantly associated with communication impairment of the person with MND. In the current study, frustration emerged as related to caregivers' perceived inability to influence the trajectory of MND despite their best efforts. Ray and Street (2006) referred to caregiving in MND as a "continual discovery of new losses" (pg.38), which mirrors the findings of the current study. Apart from frustration, caregivers in the current study reported other signs of psychological distress, such as the level of unpredictability related to how and when the disease would progress, the worry associated with the various challenges brought about by MND, their own ability to cope, and thoughts about the future, which appears in line with the existing literature (Aoun et al., 2011; Galvin et al., 2016; Herbert, Lacomis, Easter, Frick, & Shear, 2005; McCabe, Firth, & O'Connor, 2009; Miyashita et al., 2009; Mock & Berner, 2010; Ray & Street, 2006; 2007) reporting that the transformation of life due to MND resulted in exceptional strain and psychological distress in the caregivers. In addition, the caregivers in the current study admitted that they were concerned with regard to their own health in line

with the findings of Pagnini et al. (2010), especially in relation to constant fatigue and disturbed sleep. Fatigue is a recognised hallmark symptom of caregiving in MND (Gauthier et al., 2007; Ray & Street, 2006, 2007; Rabkin et al., 2007) along with insomnia or reduced quality of sleep (Oliver & Turner, 2010; Ray & Street, 2006; Van Teijingen, Friend, & Kamal, 2001; Dawson & Kristjanson, 2003). However, Pagnini et al (2010) highlighted that disrupted sleep can be symptomatic of somatic depression and it generally deteriorates in line with the progression of MND. In the current study, caregivers who had not retired continued with their employment in addition to their caregiving duties, which they reported to contribute to their level of exhaustion. Although the caregivers' desire to maintain their employment may have been primarily for financial reasons, work was reportedly perceived as providing a change in environment and an opportunity to engage in social interaction, which mirrors the findings of Cheung and Hocking (2004 a,b), who investigated the experiences of spousal caregivers of people with Multiple Sclerosis. This appeared to be important considering that some participants reported feeling lonely due to being constantly immersed in the caregiving duties. It has previously been documented that maintaining social networks can be challenging for caregivers of people with MND (Trail et al., 2003; Love et al., 2005; Ray & Street, 2006), however, having positive social support can reduce the burden related to caregiving and improve quality of life (O'Connor, McCabe, & Firth, 2008; Pagnini et al., 2010; Love et al., 2005). Similar to the findings of other studies (Chiò et al., 2005; Gauthier et al., 2007; Mitsumoto & Rabkin 2007; Pagnini et al., 2010) some caregivers who took part in the current study reported doing "very little" or "nothing" for themselves.

The participants frequently mentioned that the caregiving role had been thrust upon them unexpectedly due to particular life circumstances. If a caregiver was a

spouse of a person with MND the provision of care was perceived as an obvious duty that had to be fulfilled by them. However, when a daughter of a person with MND became the main caregiver, this appeared to evoke additional stress and a sense of unfairness for some participants. One person reported feeling expected to become the main care - provider based on her gender only. Also, personal circumstances, such as working part - time or not having had children, appeared to dictate which sibling was expected to become the main caregiver. Schulz, Beach, Cook, Martire, Tomlinson, & Monin, (2012) surveyed over 1300 family caregivers of older adults and reported that 44% perceived a lack of choice in taking on caregiving duty. This appears significant, as the authors suggested that this lack of choice can negatively influence caregivers' coping and can increase their level of stress.

The literature reports that over 90% of caregivers of people with MND perceive that they are not receiving adequate help from other family members (Alankaya &Karadakovan, 2015; Navaie-Waliser et al., 2002) which appears to be consistent with the findings of the current study. However, the current study indicated that accepting help from other family members is complex, as some caregivers did not consider other family members capable of providing the same level of care as they did. This perception was observed to result in the caregivers being reluctant to allow other family members to take over their caring duties for good or simply to enable them to have a break.

6.2.3 How People Living with MND Understood the Term Dysphagia?

It seems important to report on the differences which were noted between the understanding of the people with MND and that of the professional literature in

relation to the terms “dysphagia” or “eating / drinking / swallowing problem”. The majority of people with MND in the current study did not associate any of the above terms (“dysphagia, eating / drinking / swallowing problem”) with experiencing problems in the oral phase of swallowing. This lack of association between oral stage difficulties and the presence of an eating or swallowing impairment was evident even if it had led to the elimination of particular types of food from the diet due to, for example, reduced chewing ability. In contrast to the people with MND, the caregivers in the current study were very aware of the presence of dysphagia even if the difficulties were localised in the oral stage of swallowing only. When people with MND in the current study experienced symptoms of pharyngeal stage dysphagia however, such as difficulties with passing the bolus through the pharynx or coughing when swallowing, their own swallowing ability was more often reported as being compromised. This again mirrored the findings of Tong et al. (2011), confirming that some people with head and neck cancer and people with MND tend to relate the term dysphagia or eating / swallowing impairment to difficulties in the pharyngeal stage of swallowing only. In contrast, people with Parkinson’s disease did report their perceptions related to their impaired chewing ability (Miller et al., 2006). Jacobsson et al. (2000) also found that people who had experienced a cerebrovascular accident did describe their eating related discomfort in their oral cavity (and in their pharynx), which they referred to as, for example, a “nasty feeling” or “pain” (pg.261). Parker et al., (2004) reported, however, that 16 out of 27 participants presenting with dysphagia following a cerebrovascular accident in their study were unaware of their dysphagia - related symptoms when directly asked about the presence of any “swallowing problem” (pg.31). Parker et al. (2004) concluded that asking a standard clinical

question about the presence of a swallowing problem has little clinical relevance, which is similar to the findings of the current study.

6.3 How People Living with MND Wished to Manage Dysphagia?

6.3.1 Managing Alone – People Diagnosed with MND

It has been found that some people with MND, similar to some people who post a cerebrovascular accident (Parker, et al. 2004), are generally able to adapt to their dysphagia during the earlier stages of the disease and compensate for their impaired ability to swallow (Ball, Wright & Lewis, 2013). This was largely confirmed in the current study, for example, participants were aware of particular types of food which caused swallowing difficulties and admitted to gradually excluding these foods from their diet. However, the inability to eat favourite food due to dysphagia can be associated with a sense of loss and emotional suffering (McQuestion, Fitch, Howell, 2011). If a particular type of food was perceived as being important to people with MND in the current study, they were determined to continue to eat it and they modified the consistency of it in order to ease the effort related to swallowing (for example, soaking bread in tea).

It strongly emerged that the people with MND were focused on developing their own dysphagia management strategies when they perceived them to be necessary and beneficial at that time. This approach was consistently used rather than attempting to seek professional help. This appears to mirror the findings of Jakobsson, Larsson, Nordin, Askmark, and Nygren's (2014), who identified independence as one of the most commonly used coping strategies in MND. In their study, the ability to cope was assessed using the MND Coping Scale (Lee, Rigby, Burchardt, Thornton, Dougan, &

Young, 2001), where independence was rated as: (1) aiming to be as independent as possible, (2) leading as "normal" a life as possible, (3) deciding about own life, and (4) taking each day as it comes. In the current study, participants also wished to maintain their independence and appeared motivated to stay in charge of their own lives despite their inability to control the ultimate progression of MND. They aimed to manage dysphagia if it disrupted their "normal" life often by identifying their own strategies. Once the strategy was applied they no longer focused on it and no longer perceived dysphagia to be an issue. Their drive to focus on the present moment appeared to be very strong, which has also been found in the recent literature pertaining to MND (Gale, 2015; Greenaway et al., 2015).

6.3.2 Wishing for More Professional Help – The Caregivers

Providing care for a person with dysphagia requires adequate training in order to develop relevant skills (Aziz & Campbell-Taylor, 1999). It has been recognised that professional services play an important role in supporting the caregivers of people with MND and that the satisfaction with professional services is related to the caregivers' ability to cope with their burden of care (Goldstein et al., 2006; Mockford et al., 2006; Pagnini et al., 2010). In the current study, the caregivers wished to be able to access professional supports as they required them and in relation to particular aspects of the management of dysphagia only, such as, their role in the management of choking and non - oral feeding, the management of dysphagia in the later stages of the disease (particularly in the absence of gastrostomy), and techniques for making a modified diet more appealing for the person with MND. The above findings largely mirror previous findings in the literature, for example, the need for increased support for carers of people managing non - oral feeding at home has already been highlighted

for people with MND (Stavroulakis, 2016). Ray and Street (2006) also previously reported that caregivers were fearful that the person with MND may die from choking and that the management of choking is positively associated with caregivers' distress. However, the caregivers in the current study expressed strong selectiveness in relation to professional input they wished to obtain for dysphagia and appeared to have own beliefs as to which areas of dysphagia should be professionally managed.

The value associated with teaching caregivers relevant skills has been already recognised, particularly in relation to problem - solving skills which could assist the caregivers in adjusting to the changing needs of the person with MND (Alankaya & Karadakovan, 2015; Mock & Berner, 2010; Murphy et al., 2009). Alankaya and Karadakovan (2015) reported findings similar to the findings of this study, whereby the caregivers of people with MND expressed the need for education in some dysphagia - related areas, such as managing oral feeding when dysphagia is present, managing secretions, and facilitating self - feeding. However, addressing the training needs of caregivers in MND may not be straightforward particularly if the training was to be provided for a group of caregivers rather than individually, and also considering that increased awareness of dysphagia can evoke more distress in the caregivers. Locock and Brown (2010) concluded that for some people meeting other caregivers can be very difficult, especially when they are at different stages in their journey with MND. Similar issues were reported by one participant in the current study, who expressed her distress following dysphagia training which did not meet her individual needs. This distress was reported as being related to meeting caregivers of people with MND at a more advanced stage of the disease and not gaining any new knowledge from the training.

The caregivers in the current study considered that they had an important role to play in the management of dysphagia. In A. Johansson and U. Johansson (2009) the family caregivers of people who experienced a cerebrovascular accident and traumatic brain injury also considered themselves to be experts in the needs of the person under their care in relation to dysphagia. The caregivers in the current study often referred to the people with MND as being unable or unwilling to admit the extent of their dysphagia. Although previous studies reported that the perspectives of people with MND and their caregivers differ (Adelman et al., 2004; Trail et al., 2003), the current study suggests that the caregivers are aware of those differences in relation to dysphagia and consider their own views to be more "correct".

Over half of the group of caregivers expressed their wish to have an opportunity to consult with a professional with regard to dysphagia management options without the person with MND being present. This can be understood considering that they perceived their knowledge in relation to the person's with MND dysphagia to be superior and more objective than that of the person with MND. Lerum, Solbrække, & Frich (2016) observed that the conflict between the needs of people with MND and their caregivers can also influence whether the caregivers feel able to voice their own opinions and needs. The current study highlighted that caregivers' awareness of the discrepancy between their own views and the views of people with MND in relation to dysphagia influenced their desire to access professional services without the presence of the person with MND. Lerum et al. (2016) highlighted that caregivers were concerned with "doing the right things" (pg. 6) for the people with MND and in the current study these "right things" included notifying the professionals about the issues experienced in relation to dysphagia at home.

6.3.3 Approaching Professional Recommendations to Manage Dysphagia

This study revealed that the timing of professional recommendations was considered as important by participants in both groups, who preferred to receive the input when they considered themselves ready to accept it and deal with it. For example, in the early stages post diagnosis of MND participants (both groups) admitted to being unable to process professional information and frequently reported that they had felt overwhelmed. It also emerged that the optimal timing for input from dysphagia - related professional services was perceived as being dependent on the severity of dysphagia experienced by the person with MND. Professional input offered in the early stages post-MND diagnosis (within two months) was perceived as being "intrusive" or "unnecessary" by some participants, who presented with mild dysphagia (or provided care for a person with MND and mild dysphagia). However, if participants were experiencing severe dysphagia (or caring for a person with severe dysphagia), dysphagia - related services were viewed as "life savers" even if they were received shortly after the diagnosis.

In their longitudinal study on strain and needs in caregivers in MND, Bruletti, Comini, Scalvini, Morini, Luisa, Paneroni, and Vitacca (2015) distinguished four stages: (1) Early coping and adjustment, (2) Maintenance, (3) Transition to end stage, and (4) Coping with change / loss. Although establishing the duration for each phase appears to be impossible considering that MND affects people in a very individualised way, some participants in the current study provided data suggesting that the early coping and adjustment phase lasts at least two months. Martin et al. (2016), in their study investigating the decision - making process in relation to gastrostomy and non - invasive ventilation in MND from the perspective of health care professionals, stated

that in order to accept a professional intervention people must first accept their diagnosis. In relation to dysphagia in MND this finding implies that in order to comply with a specific professional recommendation targeted at the management of dysphagia, a person must first recognise and accept their dysphagia. This again reinforces the importance of the timing of professional input, particularly considering that in the current study this timing appeared to influence participants' future engagement with dysphagia - related services. If the timing of professional support was perceived as inappropriate it had a negative impact on the development of rapport which led to less desire to engage with this service again. In the current study, people living with MND appeared unable to accept their diagnosis of dysphagia if they were only beginning to adjust to the diagnosis of MND itself.

The issue of compliance with dysphagia related professional recommendations appeared to be unique for the people with MND in the current study. A number of studies established the reasons for patients' noncompliance with dysphagia recommendations, for example, denial or minimization (Colodny, 2005; King & Ligman, 2011; Leiter & Winsor, 1996). However, none of them appeared to be truly mirrored in the current study. When participants with MND were given a professional recommendation to manage their dysphagia, they engaged in a careful process of consideration, during which they tested the usefulness of this recommendation and attempted to personalise it by developing / applying own modifications. Moreover, the people with MND expected to clearly see the benefits related to following a professional recommendations, for example, to experience less coughing during swallowing after the fluid had been thickened. This appears consistent with the health belief model, where the patients' perceptions of benefits outweigh the negatives associated with a particular intervention (Trick, 1993). In the current study, if the

recommendation was not considered to have obvious benefits it was rejected. However, if it was considered to be potentially beneficial, participants engaged in a process of "experimentation", whereby the recommendation was tested and a decision was made regarding whether they would follow it or not. Interestingly, data received from one participant suggested that after he had engaged in the process of evaluating and internalising a professional recommendation, he began perceiving this recommendation as being self - developed. In addition, it was frequently noted during the interviews that participants reported a number of self - developed dysphagia management strategies which in fact significantly resembled compensatory dysphagia management strategies documented in the professional literature (Cichero & Murdoch, 2006; Groher & Crary, 2009; Logeman, 1998; Murry, Carrau, & Chan, 2016, Steele et al., 2015). It is not known, however, whether these recommendations, which were perceived as being self - developed, had in fact been professionally recommended in the first instance.

The caregivers approached professional recommendations to manage dysphagia in a similar manner; however, they did not appear to engage in any prolonged testing of their usefulness. It emerged that the majority of caregivers considered that they possessed the most in - depth and accurate knowledge in relation to the swallowing ability of the person with MND, more superior in fact than the person themselves and the professionals involved in the management of dysphagia. When a professional recommendation was made, the caregivers appeared to assess or judge whether or not it was appropriate for the person with MND. If the recommendation was perceived to be unsuitable or unnecessary the caregivers did not make any attempt to follow the recommendation at home. Similar findings were reported by the caregivers of people with an intellectual disability (Chadwick et al., 2006), who did not comply with

dysphagia recommendations if they did not recognise their relevance for the care - receiver. In the current study, however, if the caregivers regarded a specific dysphagia management strategy (such as non - oral feeding) as being important for the person with MND, they made every attempt to support the person with MND and convince them to follow this recommendation.

The caregivers of people with MND have previously been recognised as key people responsible for the continuity of care between the home and healthcare environments (Pagnini et al., 2010). It seems imperative, therefore, to recognise their views and opinions in relation to the management of dysphagia, as ultimately having the support of the caregivers may increase the likelihood of recommendations being followed in the home.

In the current study, participants reported that modifications to fluid consistency and food texture and non - oral feeding were the most common professional recommendations received to manage dysphagia. These recommendations are discussed in the sections below.

6.3.3a) Texture Modifications

A texture modified diet is frequently recommended in the management of dysphagia (Cichero & Murdoch; Groher & Crary, 2009; Logeman, 1998; Murry et al., 29016; RCSLT, 2014; Steele et al., 2015). However, this management strategy is often considered to be disliked by the people with dysphagia (Chadwick et al., 2006; Colodny, 2005; Garcia et al., 2005; King & Ligman, 2011; McCurtin et al., 2017; Macqueen, et al. 2003), which was confirmed in the current study. It emerged that both people with MND and their caregivers approached the modification of food and

that of fluid in a different manner, in line with the findings of Low et al. (2001), who reported that their participants were more likely to comply with recommendations to alter foods textures rather than to thicken fluids. In the current study, the majority of participants not only adjusted to the alternations required to food textures (if at level A or B, Appendix 1) but also perceived them as being self - developed. Some of the caregivers reported that they had already begun modifying the diet of the person with MND prior to the recommendation being made by a professional. This appeared to be related to the caregivers' awareness of the risk of choking associated with dysphagia in MND. Despite difficulties reported with textures C and D (Appendix 1), modifications to the texture of food were perceived as being more natural, especially considering they required the use of regular domestic equipment, such as a blender. In contrast, the thickening powder for fluids had to be professionally prescribed and was considered to be a medical intervention. The majority of caregivers (9/10) had not had any previous experience of thickener at all. However, they understood the thickening of fluids to be an intervention that may help the person with MND to maintain better health.

Changes to liquid consistency were always reported as being very difficult to accept by people with MND. One participant with MND in the current study openly admitted that he would choose non - oral feeding over having to use thickening powder in his fluids. Although six people with MND in the current study reported receiving a professional recommendation to thicken fluids, only one participant opted to implement it fully and reported that this decision had required a significant amount of time to process and adjust to. The same person highlighted their fine motor difficulties related to opening the tin and using the scoop to measure the correct amount of thickening powder. These fine motor difficulties, if not managed professionally in

advance, could potentially contribute to a reduction in the independence of a person with MND.

The caregivers reported that they and the person with MND may have different opinions regarding the optimal texture of food or consistency of fluids. This finding is in line with the findings of studies by Adelman et al. (2004) and Trail et al. (2003) which highlighted differences in the views of the two groups in relation to the perceived needs of the person with MND. In the current study, these differences were described as causing increased stress and worry, as the caregivers aimed to stimulate the appetite of the person with MND in the hope that good oral intake would increase their survival. The caregivers found that when the texture of food demanded a great deal of transformation it became impossible to prepare food which was visually appealing. Smooth pureed or liquidised diets (textures C or D; Appendix 1) were reported as being the most problematic due to their unappealing appearance. For example, one caregiver reported that despite her efforts the meals always resembled "a horrible slop". Although all caregivers in the current study reported receiving professional recommendations in relation to the required level of modification of food textures for the person with MND, none of them remember obtaining assistance in making the food more appealing. Reilly, Frankel, & Edelstein (2013) investigated the role of molecular gastronomy in improving the aesthetic appearance of food of modified textures for people with dysphagia. The authors concluded that using specific molecular gastronomy techniques (such as spherification, gelification, and emulsification) can make the food more appetizing for those with dysphagia.

The implementation of thickened fluids at home was also seen as problematic by the caregivers as it was perceived as being unpleasant by the people with MND. Similar issues were reported in a study by Chadwick et al., (2006), which focused on

a population of caregivers of adults with an intellectual disability. The caregivers in both studies therefore faced a dilemma, in that they understood that thickened fluids may bring medical benefits, but they also wanted to respect the wishes of the person under their care. In the current study, some caregivers attempted to encourage the person with MND to accept thickened fluids and if this was unsuccessful some admitted to adding the thickener covertly. Other caregivers perceived thickened liquids as being very unpleasant and having a negative impact on the quality of life of the person with MND and therefore they decided not to use it at all, as in their opinion, quality of life values were more important, which again mirrors the attitude of the caregivers in the Chadwick et al. study (2006).

6.3.3b) Non - oral Feeding

Participants in the current study appeared to have individual expectations related to non - oral feeding, such as hope for prolonged survival, improved nutritional status leading to preserved independence and better quality of life, and overcoming the medical risks related to dysphagia, such as chest infections. The caregivers believed that non - oral feeding may prolong the survival of a person with MND despite the limited evidence to support this belief (Katzberg & Benatar, 2011; Simons, 2005). The current study revealed that the two caregivers of older people with MND (over 70 years of age) and the caregiver who had been providing care for the longest time (over 6 years) were less supportive of life prolongation options such as non - oral feeding.

The perceptions of people with MND in relation to non - oral feeding also appeared to influence their desire to undergo a gastrostomy. People with MND, who

perceived non - oral feeding as being an option which may prolong their independence and quality of life, seemed to opt for it relatively quickly once it was professionally recommended (a few weeks). For others, the decision was more complex and demanded a few months of consideration (up to 9 months). Participants reported that this process of contemplation was negatively influenced by hearing conflicting opinions pertaining to the benefits of non - oral feeding from professionals delivering dysphagia - related care. Worryingly, two participants perceived that they had not been adequately informed about what was involved in the gastrostomy procedure, what the associated risks were, and what care was required afterwards. For example, one lady was convinced that the feeding tube would be inserted in her throat rather than her abdomen. Another person reported not being made aware of any possible complications which may occur post-gastrostomy procedure, and was shocked when they developed. The above findings are in line with Stavroulakis et al. (2014), as people with MND in their study also perceived that they had not been adequately informed about the gastrostomy. Also, people with MND in a study by Greenaway et al. (2015) reported feeling pressurised and unsupported by professionals during the decision making process in relation to non - oral feeding.

One person with MND in the current study never came to terms with the fact that he had undergone a gastrostomy. For this man, the presence of the feeding tube appeared to be almost symbolic, in that it signified his disease as being terminal with no hope of recovery. The gastrostomy tube was a constant reminder of MND and its associated prognosis, as it required daily care. This person never consented to the commencement of non - oral feeding even at the advanced stage of dysphagia. This case potentially highlighted not only the need for clear and succinct communication between clinicians and the person with MND, but also the need for emotional support

for people with MND post gastrostomy. Educational support for people with MND and their caregivers in the first few weeks of non - oral feeding at home was previously reported as being necessary by Stavroulakis et al. (2016), who deemed the training provided in acute settings to be insufficient. This current study, however, also highlighted the potential need for psychological support in addition to the provision of practical information pertaining to the management of non - oral feeding in the home.

When attempting to understand the experiences of non - oral feeding in MND, the issue of optimal timing for undergoing gastrostomy insertion and subsequently for commencing non - oral feeding should be considered. Although evidence in relation to the timing of the introduction of non - oral feeding in MND is considered to be insufficient (Katzberg & Banatar, 2011), NICE guidelines (2016) support the early introduction of this topic to provide people with adequate time to consider it (as, according to the authors, gastrostomies performed at a late stage of MND can be associated with significant health risks). Although the importance of psychological readiness to accept non - oral feeding has already been recognised in MND (Benstead et al., 2016), the current study revealed potential discrepancies between the psychological readiness of people with MND and their caregivers. It emerged that the majority of caregivers of people with MND were informed about the possibility of non - oral feeding while the person with MND was still managing an oral diet relatively well. However, the caregivers tended to avoid thinking about non - oral feeding for as long as they felt able to manage the symptoms of dysphagia without it. When dysphagia progressed and eating became “a struggle”, they understood non - oral feeding to be necessary. In contrast, this understanding typically emerged in the group of people with MND at a much later stage.

Another factor influencing personal readiness to accept non - oral feeding appeared to be the changed relationship with food, which emerged as a consequence of progressive dysphagia. However, discrepancies between the two groups were also evident in this regard, and these are discussed in section 6.4.5 below.

Although the majority of caregivers in this study reported that the decision as to whether or not to accept non - oral feeding should lie with the person with MND, they did attempt to influence this decision. These findings are consistent with those in a study by Martin et al. (2016). In their study, both the people with MND and their caregivers were perceived as being actively involved in the decision - making process.

In this study, in line with the findings of a study by Patterson et al. (2013), it emerged that the caregivers of people with MND required time to adapt to managing non - oral feeding at home, and that this management was perceived as being easier when they noticed the benefits related to it (such as increased energy levels). Similar to the findings of a study by Stavroulakis et al. (2016), all caregivers in the current study reported being unable to imagine living without the support of non - oral nutrition and hydration despite admitting to a number of issues related to gastrostomy care and its negative impact on the social aspects of meals (which is further discussed in section 6.4.1. below).

6.4 The Impact of Dysphagia

6.4.1 The Social Aspects of Meals

Living with dysphagia as part of MND affected the social aspect of meals for both the people with MND and the caregivers. Although both groups continued to eat out despite the awareness of dysphagia and the necessity to adjust the textures of food

or eating techniques, the caregivers appeared to be much more concerned about the safety of the person with MND when eating in public.

As the people with MND attempted to self - manage their dysphagia, they reportedly felt less at risk of choking and aspirating in a distraction-free environment. However, ensuring a distraction - free environment during meals was problematic, as the people with MND wished to maintain their previous social routines, such as going to restaurants. Dining with others was prioritised by some people with MND, who also reported having to be very careful and concentrate during meals. It emerged that participants with MND preferred to engage in eating with others for as long as they were able to tolerate any oral intake, and despite having a gastrostomy tube in situ. They appeared able to overcome the potential embarrassment associated with, for example, assisted feeding in public or decreased ability to control the food in the oral cavity. Moreover, some people with MND continued dining out despite having to eat food of modified consistency and appearance, which seems extraordinary considering the evidence regarding the social importance and influence of food. For example, the literature states that the social influences related to the appearance of food are powerful and pervasive (Higgs & Thomas, 2016). It has also been established that who we dine with dictates our food choices and other food - related behaviours, such as decisions regarding portion size (Cruwys, Bevelander, Hermans, 2015; Herman, Roth, & Polivy, 2003; Herman, 2015, Vartanian, 2015). Eating with family members also affects our choice of food and the diets of people who are related to each other are considered to be correlated (Robinson, Otten, & Hermans, 2015; Guidetti, Cavazza, Graziani, 2014). However, MND does not appear to conform to these links between social norms and diet, as the food choices made by participants with this disease were often dictated by their own perception of their swallowing ability rather than by any social influences.

When people with MND and dysphagia have a meal with others, their main focus appears to be on ensuring that the food is safe to swallow, rather than considering what food would be socially acceptable. Although this finding may be expected considering that participants with MND preferred to manage their dysphagia on their own and desired to continue their usual routines, it is still interesting to note that they prioritised eating with others despite dysphagia and despite having to break food related social norms, which could potentially cause psychological distress, for example, embarrassment.

The caregivers were often concerned when eating out, as they felt responsible for ensuring the appropriate texture of food for the person with MND in the hope of eliminating the risk of choking. They reported carefully searching for suitable foods on the menu and also double - checking the textures of food by consulting with restaurant staff. A lack of understanding of dysphagia in restaurants was often highlighted, and the caregivers perceived it necessary to provide detailed and clear instructions pertaining to appropriate food types to the restaurant staff. Although these findings generally mirror those in a study by Nund et al. (2014b) which researched the eating - out experiences of caregivers of people with dysphagia related to cancer, a difference was noted in relation to the reported reasons for ceasing going out to eat. Some participants in the Nund et al. study viewed eating out as too challenging due to the level of preparation required to ensure appropriate food textures, such as checking the restaurant menu in advance. In contrast, the caregivers of people with MND in the current study when justifying their decision to stop eating out, provided reasons unrelated to dysphagia, such as the physical deterioration of the person with MND which resulted in their transportation outside the home being too difficult.

A. Johansson and U. Johansson (2009) reported that caregivers experienced discomfort associated with having meals with a person with dysphagia in the home environment. In the current study, caregivers' discomfort or embarrassment only emerged in relation to eating out with a person with MND. In addition, the caregivers seemed to overcome this feeling with time. One caregiver reported choosing darker restaurants and quieter times to avoid upsetting the meal experiences of others. However, in time she became less concerned with the reactions of other people. Instead, her focus was maintained on supervising her husband during meals to ensure his safety.

Eating out was also reported by some caregivers as a potential source of embarrassment for the person with MND, particularly if assistance with meals was required. None of the caregivers stated, however, that this feeling of embarrassment negatively influenced the person's with MND desire to continue eating out.

The commencement of non - oral feeding brought significant changes to the social aspects of meals in both groups particularly when the person with MND became unable to tolerate any oral intake. The lost ability to share a meal appeared to have significant psycho - social consequences for the caregivers as well as for the people with MND. However, in the current study it emerged that these psycho - social consequences were more significant for the caregivers. In line with previous studies on non - oral feeding in MND (Mayre-Chilton et al., 2011; Progas, 2015, Stavroulakis et al., 2016), the caregivers in the current study reported that the increased duration of feeding time led to them feeling confined to the home. The results of the current study suggest that the type of non - oral feeding in situ can influence the severity of the impact on caregivers' lives. It emerged that continuous gastrostomy feeding was perceived as significantly more challenging in comparison to bolus feeding, as it took

a greater amount of time and ultimately confined the person with MND and / or the caregiver to their home for longer.

6.4.2 Transformed Relationship with Food

In order to understand the impact of dysphagia on participants' relationship with food, we must first consider the average person's relationship with food. Although eating is considered to be one of the most fundamental of human activities (Capaldi, 1996), food is not simply viewed as a necessity in order to sustain life (Conner & Armitage, 2002). It is considered to be a source of pleasure and a source of relief from stress as we use it to regulate our behaviour (Conner & Armitage, 2002). Daily life is structured by our eating routines and our choices of food are considered to be a way of self - presentation to others (Conner & Armitage, 2002). Food preferences can be associated with expressing both personal and group identity (Kenny, 2015), for example, people (particularly women) tend to manage other's impressions of them by selecting specific types and amounts of foods (Conner & Armitage, 2002). Expressing group identity through food can manifest by eating specific types of food, such as vegetarian, or selecting foods based on religious or cultural traditions (Hauck-Lawson, 2004; Kittler, Sucher, & Nelms, 2012). The visual appearance of food is considered to be very important, particularly as cognition plays a role in our perception of food and influences our decision whether or not to eat it (Conner & Armitage, 2002). The connection between food and emotional wellbeing has previously been established (Gibson, 2006). Eating affects the mood and emotions of human beings as it is considered to be a fundamentally rewarding behaviour (Vögele & Gibson, 2010). Positive effects of food are associated with whether or not our expectations have been met regarding its size and composition (Gibson, 2006).

The current study identified a number of differences in how participants perceived food following the diagnosis of MND and subsequently following their experiences of progressive dysphagia. These are discussed in the subsequent sections.

6.4.3 Nutrition and Weight

Changed meaning of food was already reported in the population of people with head and neck cancer and dysphagia (McQuestion et al., 2011). This study revealed that MND transformed the relationship that participants in both groups had with food prior to this disease. Both people with MND and the caregivers reported approaching food in a different manner shortly after the diagnosis of MND, regardless of the presence and severity of dysphagia. Gradually, they began to prioritise the more medical properties of food, such as nutritional values and calories, rather than its taste and appearance. The key factor driving this change was identified as participants' awareness of the important role which nutrition plays in prolonging the survival of the person with MND, which is in line with the scientific evidence (Desport et al., 1999; Jawaid et al., 2008; Körner et al., 2003; Lacomblez, et al., 1996; Limousin et al., 2010; Stambler, Charatan, & Cedarbaum, 1998). However, the maintenance of weight in people with MND is considered to be a difficult task not only due to cachexia, but also hypermetabolism and loss of appetite (Holm, et al., 2013, Körner, et al., 2013), which in the current study emerged as being particularly stressful for the caregivers.

The presence of dysphagia has been shown to disrupt the lifestyle of caregivers and cause multiple challenges for them (Arai et al., 2005; Nund et al., 2014b; Patterson et al., 2013; Penner et al., 2005). In the current study the majority of caregivers reported feeling under constant pressure to increase the weight of the person with

MND, especially as dysphagia progressed. Over time, attempting to ensure high calorie food intake for a person with MND became almost an obsession for some caregivers, who reported being continuously worried about the weight loss of the person with MND. Additionally, some caregivers considered providing adequate nutrition as the only manner in which they could have some influence on the progression of MND, which appeared to motivate them to change the previous eating habits of the person with MND. However, the caregivers considered the high calorie diet to be unhealthy and unsuitable for the rest of the family and therefore they often aimed to maintain their own eating routines, which challenges the theory of social modelling of eating which states that people tend to eat similar sorts of food (Cruwys et al., 2015; Guidetti et al., 2014; Herman et al., 2003; Herman, 2015, Vartanian, 2015, Robinson et al., 2015). This finding is also in contrast with Nund et al. (2014b), who reported that caregivers' of people with head and neck cancer frequently chose to only eat foods which were suitable for the person with dysphagia.

In the current study, changes in the eating routines of the caregivers emerged, however, when the person with MND became unable to tolerate any oral intake and was relying exclusively on non - oral feeding. These caregivers often reported feeling guilty when eating and aimed to eat in isolation to avoid causing further distress to the person with MND. The negative impact of non - oral feeding on the psychological health of caregivers' in MND has also been reported by Stavroulakis et al. (2016). It appears that for as long as the person with MND is able to tolerate some oral diet, even if that diet is significantly modified, the impact of dysphagia on the caregivers' own diet is not immense. However, when the person with MND ceases an oral diet completely, it appears to have a strong negative effect on the caregivers' eating routines and psychological well - being.

6.4.4 Eating as a Struggle

As dysphagia progressed, the relationship participants in both groups had with food appeared to change. In the current study, as the people with MND began to experience the loss of control over their own swallowing ability they reported that eating gradually became an unpleasant experience and compared it to a "struggle". The effort required during meals resulted in increased exhaustion. The taste and appearance of food became insignificant as the main role of food was understood to be the provision of nutrients, almost like getting petrol for a car. Food had to be consumed in the hope of prolonging life or at least achieving a better quality of life. Although life extension did not appear to be the priority for everyone, having a good quality of life did and this was seen as being associated with having more energy. Meals began to resemble a schedule of medications.

The fact that the caregivers considered dysphagia as a serious risk was reported to influence the emotions they experienced during meals. The atmosphere during mealtimes shared with someone with MND was described as tense. Instead of enjoying the food, the caregivers provided constant supervision in order to observe and address any risky behaviour of the person with MND, such as overfilling the mouth or eating too fast. They remained focused on observing signs of dysphagia and the occurrence of any potential choking episodes. None of the caregivers reported enjoying their food nor commented on the pleasure associated with eating meals. While none directly stated that eating became unpleasant, it was observed that the caregivers own behaviour during meals was almost automatic, as their attention was solely focussed on the needs of the person with MND.

The changed relationship with food, decreased pleasure related to eating and drinking, and considering meals to be a "struggle", appeared to influence the willingness and readiness of participant's to accept non - oral feeding, which is discussed in the next section (6.4.5).

6.4.5 Readiness to Accept Non - oral Feeding

This study indicated a connection between participants approach to food, relationship with food, and their readiness to accept non - oral feeding, similarly to Johnson et al. (2012). The current research indicated that this readiness emerged earlier in the group of caregivers in comparison to the people with MND. The people with MND attempted to continue an oral diet for as long as they were able to swallow the food and despite experiencing multiple symptoms of dysphagia. When their relationship with food changed and their food related pleasure diminished, they began to detach from food and were more likely to accept non - oral feeding. The caregivers, however, would prefer the person with MND to avail of non - oral feeding as soon as they felt unable to maintain their weight by means of an oral diet.

In the current study, the majority of people with MND who had a gastrostomy tube in situ, appeared to have started using it when their pleasure from eating food had diminished or when they perceived their own swallowing ability to be compromised and risky. One person with MND, who was in the process of considering non - oral feeding during this study, did not consent to this procedure for nine months. He appeared to require this time to fully acknowledge the impact of his dysphagia, which had manifested in an inability to maintain his weight by using oral diet and nutritional supplements alone, poor appetite, and decreased enjoyment from food.

It emerged that non - oral feeding was perceived as a "last resort" by the majority of people with MND, something to be considered only if all other techniques have failed. This finding may help to elucidate the results of studies by Stavroulakis et al. (2014, 2016) and Greenaway et al. (2015), who noted that people with MND, who were offered a gastrostomy while their eating and drinking was still well preserved, were reluctant to or did not consent to this procedure. Stavroulakis (2016) observed that people with MND had a tendency to opt for the late insertion of a gastrostomy, which was also observed in the current study. The NICE guidelines state that late gastrostomy insertion can increase the risk of medical complications and therefore it should be recommended prior to the onset of severe bulbar symptoms (NICE, 2016). Other reports, however, support the late insertion of a gastrostomy tube (Czell et al., 2013; Kak et al., 2017; Sarfaty et al., 2013; Spatro et al., 2011), which from a psychological perspective could potentially be a less traumatic option for a person with MND.

Caregivers' readiness to accept non - oral feeding appeared to be influenced by their ability to control the intake of the person with MND. The preparation of food for a person with dysphagia has been recognised as a complex task demanding increased effort and time, and therefore causing disruption to the lives of caregivers' (Nund et al., 2014b; Patterson et al., 2013). In the current study, the caregivers described their attempts to find techniques to compensate for progressing dysphagia as ongoing. The preparation of meals for a person with MND was complex and three main concerns emerged in the group of caregivers: (1) ensuring high calorie diet, (2) ensuring food is relatively safe to swallow, and (3) attempting to make food as appealing as possible for people with MND in order to stimulate their appetite. When

the caregivers perceived themselves as unable to maintain an appropriate diet for the person with MND, they were more likely to support the idea of non - oral feeding.

6.5 Professional Management of Dysphagia

The management of MND is palliative from the moment of diagnosis (Bede et al., 2011). It is recognised that the perspective of people with terminal illness, such as MND, in relation to their care can differ to that of the professionals involved in that care (Rodriguez & Young, 2006). Roe and Leslie (2010) investigated challenges related to palliative care services and recommended setting the goals from the outset. However they also highlighted that during the course of the illness these goals may change. The next sections discuss the experiences of dysphagia - related services as reported by both people with MND and their caregivers.

6.5.1 MND Clinics

In this research, participants reported availing of multidisciplinary dysphagia services in both hospitals and in the community and 90% attended MND Clinics at some stage in their journey with MND. The benefits of multidisciplinary care in MND are widely documented (Calzada-Sierra & Fernandez, 2001; Lee, Annegers, & Appel, 1995; NICE, 2016; Traynor et al., 2003) and the literature suggests that this model of care prolongs survival (Chiò et al., 2009; Hardiman, van den Berg, & Kiernan, 2011; Higo, Tayama, & Nito, 2004; Lee et al., 1995; Oliviera & Pereire, 2009; Rooney et al., 2013; Traynor et al., 2003). Despite these findings, Foley, Timonen, and Hardiman (2012) noted a paradox between positive outcomes related to a multidisciplinary approach to care in MND and patient dissatisfaction with services. This paradox was

associated with the use of objective outcomes to evaluate the benefits of multidisciplinary care (for example, cost and survival) as opposed to service - related satisfaction which is subjective and inclusive of both the cognitive and emotional perceptions of service users. Although it has been recognised that the perspectives of service - users should form the core of the care process (Oliver, Borasio, & Walsh, 2006), Foley et al. (2012) highlighted that in MND service users' experiences of services are poorly understood. The current study appears to be the first investigating the perceptions of people living with MND specifically in relation to dysphagia - related services.

McCabe et al. (2008) investigated the satisfaction of people with progressive neurological diseases in relation to general health services, and concluded that integrated service delivery (similar to MND Clinics) was considered to be the most useful. This is generally consistent with the findings of the current study, as all participants expressed their overall satisfaction with the services received for dysphagia from MND Clinics. Despite this satisfaction, however, participants also reported multiple negative feelings associated with their attendance at MND Clinics.

It emerged that participants' distress was not only related to meeting a number of professionals on the same day and often simultaneously, but also to the amount of information participants received on the day. Hughes et al. (2005) highlighted the importance of delivering an appropriate level of professional input at the right time. The authors stated that people with MND tend to independently look for facts themselves if there is a mismatch between their needs and the level of information provided by the services. This finding could suggest that if professional management does not match the individual needs of a person with MND it may potentially have a negative impact on their future engagement with professional services.

In addition to feeling overwhelmed during MND Clinics, the people with MND reported the emotional distress associated with meeting other people at various stages of the disease. However, the opportunity to avail of multiple professional inputs in one day, thus reducing the need to travel and ultimately decreasing the burden on families, was seen to counterbalance any emotional distress experienced.

6.5.2 Wishing for a Positive Experience

No professional can cure MND. No clinician can stop the progression of MND. Potentially, this awareness may lead to limited belief in the value of professional input and transform the expectations of health services. When participants in both groups reported on their experiences of dysphagia - related services, they frequently expressed satisfaction when professional consultations were positive and pleasant, when they were given some "good news" in relation to their swallowing status, and when they perceived that they had a good rapport with the professionals. Both people with MND and their caregivers reported disengaging or being reluctant to attend a professional consult with a clinician, who was perceived as disrespectful and unfriendly. None of the participants spoke about their expectations in relation to obtaining dysphagia - related therapy or even professional recommendations which could improve their swallowing ability at home. Instead, it emerged that participants prioritised the rapport with professionals delivering dysphagia - related care, and wished to feel comfortable and respected during professional visits. The people with MND in the current study strongly valued being given a professional recommendation to manage dysphagia without feeling pressured to adhere to it.

Hughes et al. (2005) highlighted that despite obtaining professional advice, people with MND are aware that they have to work through their own needs themselves. In the current study, the participants (in both groups) appeared to perceive that they were the main person responsible for the management of dysphagia. In addition, some people with MND in the current study perceived there to be limited knowledge and experience of MND amongst some professionals involved in the management of dysphagia in the community, in line with Hughes et al (2005). This appeared to be associated with participants' feelings of disappointment and wasted time, and a reluctance to engage with these services in the future.

Consistent with the findings of Locock and Brown (2010), domiciliary services were perceived to be "quite disturbing" by some caregivers of people with MND in this study. This was particularly evident when home visits were provided by multiple professionals and when the timing of domiciliary services was not agreed in advance, therefore increasing the pressure experienced by the caregivers. Other caregivers, however, preferred domiciliary input and did not report any distress related to this service model.

This study revealed an array of emotions related to experiencing dysphagia and confirmed the necessity of psychological support for the caregivers during their journey. Noticing the progressive symptoms of MND has already been reported to increase caregivers' burden (Aoun et al. 2012), and this also applies to dysphagia. The caregivers in the current study identified being particularly stressed and worried when: (1) they were unable to follow a professional recommendation despite understanding the rationale for it (for example, when a person with MND refused to take thickened fluids and the caregiver was aware of the risk of aspiration), (2) when they were unable to prevent a person with MND from losing weight (while being aware that this is an

important prognostic factor), (3) when they were introduced to the idea of non - oral feeding for the first time, and (4) when they had to manage non - oral feeding at home.

6.5.3 Suggestions for Change

Interestingly, despite reporting some negative experiences and emotions regarding dysphagia - related services, the majority of participants stated that they were unable to think about any potential improvements which could be made to these services. Only two people with MND directly expressed their suggestions for change with regard to dysphagia - related services.

The first suggestion related to an improvement in the communication between the professionals involved in the management of dysphagia. Miscommunication was perceived to exist between hospital and community services reportedly leading to decreased satisfaction with services and increased frustration. In addition, contradictory professional recommendations regarding non - oral feeding were reported, which not only contributed to increased distress, but ultimately led to a delay in participants consenting to this procedure.

The second suggestion related to a change in the referral system, where the frequency of professional reviews is dictated by the person with MND rather than the professional. The existing system was perceived as problematic for two reasons. Firstly, it was reported that professional and personal perceptions of dysphagia can vary. Therefore, it was highlighted that people with MND may misjudge their own swallowing ability and ultimately not report any issues related to it. Secondly, people with MND may consider their attendance at a professional consultation as increasing the caregivers' burden, for example, due to the time required to travel. Foley et al.

(2016) stated that people with MND resisted being a burden to their families and aimed to minimise the strain imposed on their families. Therefore, there could potentially be a risk of people concealing their own swallowing impairment and not seeking professional input in order to avoid increasing the burden on the caregiver.

6.6 Reflections for Professionals Providing Dysphagia - related Services in MND

A number of potential practical considerations for professionals delivering dysphagia - related care in MND emerged in this study. Firstly, this research indicated that dysphagia in MND is strongly rooted in how people with MND and their caregivers experience and cope with MND in general. In order to provide professional support for managing dysphagia, it seems necessary to recognise how MND affects the lives of people diagnosed with this disease and their caregivers, what they prioritise in their daily life, and what they expect from attending a professional service. When dysphagia is managed in a multidisciplinary setting it may be possible to divide roles and establish effective methods of communication between team members to ensure a holistic approach is provided. The following sections present a number of specific considerations, which emerged during this study. These considerations are representative of the views of the twenty participants who took part in this research, and are interpreted from a SLT's perspective.

It is recognised that the management of dysphagia should be in accordance with evidence based practice (IASLT, 2015). SLTs, as key members of an MDT team delivering services for dysphagia (ASHA, 2004; IASLT, 2015; RCSLT, 2014),

sometimes however lack clear evidence to support their recommendations when dysphagia is related to MND. For example, the guidelines in relation to the optimal timing of non - oral feeding appear to be unclear.

It seems imperative to ensure effective communication between all professionals involved in the management of dysphagia to avoid contradictory recommendations being provided to the people living with MND. If robust clinical evidence for a particular issue is not available, a team consultation should be carried out in order to agree on the management plan. This consultation should be conducted prior to meeting the person with MND and the caregiver. This type of effective communication, potentially leading to clear recommendations, may not only decrease the levels of distress and frustration experienced by the service users, but may also have a positive influence on their future engagement with the professional service.

As the number of professionals involved in managing dysphagia in MND can be high, the selection of one professional who would act as a key-worker could be considered (this has already been supported by Bakker et al., 2015 in relation to the management of MND in general). This person could act as the main point of contact and the main coordinator of the care provided. Currently in Ireland there are only three MND nurses providing domiciliary services to people with MND and acting as a link between them and the professionals involved in the three MND Clinics (Dublin, Galway, and Cork). This does not appear to be a sufficient resource considering the complexity of the needs of the 350 people who are living with MND at any one time in Ireland and their caregivers.

This study confirmed that an individual's involvement with dysphagia services should be carefully planned from the moment of diagnosis until discharge. The model

of service provision should be mutually agreed while acknowledging that as MND progresses service delivery may need to be reviewed and modified accordingly. Roe and Leslie (2010) indicated that SLTs delivering palliative care should prepare patients and their families for the possibility that priorities in relation to professional management may change as the disease progresses in order to minimise the shock when those changes occur. Palliative service provision requires a skilled therapist delivering an individualised approach and balancing evidence based practice with the personal values of people living with MND (NICE, 2016). In addition, it should be recognised and accepted that dealing with multiple risks is unavoidable when managing dysphagia as part of MND. Therefore, the clinical reasoning should be inclusive of quality of life considerations and risks assessments, which may prove to be a particularly challenging task for a novice professional. This study indicated that therapists with an appropriate level of understanding and experience in providing dysphagia - related palliative care should be engaged in the management of dysphagia as part of MND.

Dysphagia may not always be perceived as a priority for the person affected, and in this study participants with MND were much more concerned about their communication impairment. It also emerged that professionals, people with MND, and their caregivers, may all have a different perspective in relation to the presence and severity of dysphagia. Therefore, directly inquiring about a person's with MND swallowing ability may not lead to adequate information pertaining to the extent of their dysphagia. Alternatively, asking a person to describe the schedule and types of meals typically consumed may provide more accurate and richer information. In addition, it seems important to enquire about eating related pleasure in order to identify

a person's relationship with food, which may affect their oral intake and readiness to approach the topic of non - oral feeding.

It seems important to recognise that personal approaches to food and eating may change as MND progresses. As people with MND began to perceive their meals as a struggle, they may be more accepting of non - oral feeding. The caregivers of people with MND can perceive food as a severe risk due to their fear of choking. As people living with MND experience the changes in their relationship with food, they are likely to develop psycho-social consequences, which should be recognised and managed by an appropriately trained professional (such as a social worker or counsellor). The psychological and emotional status of services users should be considered when delivering professional services related to dysphagia.

The people living with MND in this study appeared to maintain hope and a positive attitude despite being fully aware of the medical prognosis. This should be acknowledged by professionals, who may not themselves perceive there to be any sense of hope related to MND. This study confirmed that participants particularly valued hearing even some minor positives while attending professional consultations.

Participants in this study expressed their wish to feel respected during professional consultations and to be offered a choice rather than being pressured into following a particular dysphagia management strategy.

Although both people with MND and their caregivers may wish to be actively involved in the management of dysphagia and, for example, decide about the appropriateness of a particular dysphagia management strategy, the professionals should consider maintaining a leading role in relation to service provision and not relying on self - referrals.

This research revealed significant differences in the dysphagia - related experiences of caregivers in comparison to the people with MND. The caregivers perceived themselves as having accurate knowledge of the swallowing ability of the person with MND; however, they were often reluctant to share it with a professional in the presence of the person with MND. The caregivers often wished to enquire about the future management of dysphagia, and again refrained from doing so when the person with MND was present. It emerged that some caregivers of people with MND would benefit from being offered individual appointments with professionals delivering dysphagia related services.

The training needs of the caregivers should be addressed, particularly in relation to managing choking episodes, as this emerged as being most stressful for the caregivers. The current study identified, however, that the caregivers attending each training session should be carefully matched based on the status of the person with MND under their care. This would be challenging considering that MND can affect people in an individualized manner. Perhaps the main emphasis should instead be placed on educating people living with MND on how to problem - solve with regard to dysphagia. Previous literature highlighted that a caregivers' ability to problem solve was the best predictor of their QOL in MND (Murphy, Felgoise, Walsh, & Simmons, 2009). The authors suggested that the skills teaching could take place over a specific number of weeks and could be delivered / reviewed indirectly (for example, over the phone). Skills teaching in dysphagia should focus on the practical knowledge necessary in order to manage dysphagia at home. Skills teaching has the potential to maximise the independence of people living with MND and potentially reduce their need to attend professional consultations.

Finally, there does not appear to be any guidelines available in relation to the issue of discharge from dysphagia - related services in MND. This issue has previously been recognised in the delivery of palliative care, whereby discharge is frequently guided by the progression of the illness or by death, as opposed to it being planned in advance (Roe & Leslie, 2010). Interestingly, in the current study participants identified the lead professional involved in the management of dysphagia to be a Speech and Language Therapist [SLT] (for people with MND eating orally) and a Dietitian (for people with MND fully reliant on non - oral feeding). This poses the question as to whether a person with MND should be discharged from dysphagia - related SLT services when they cease all oral intake. Although this study confirmed that people living with MND value having a positive rapport with professionals, establishing when and how this relationship should end requires further consideration, particularly when the withdrawal of services "may signify the beginning of the end" (Roe & Leslie, 2010, pg. 306). Roe and Leslie (2010) suggested that the process of discharge should be well planned from the onset of the disease in order to avoid the risk of imbalance, whereby the most intensive input is provided at the beginning of the individuals' journey with a terminal illness, and the least input at the end prior to discharge. This imbalance was also highlighted by the participants in the current study, who reported receiving intensive professional services soon after the diagnosis of MND when they were only attempting to adjust to the diagnosis and much less professional input at the terminal stages of MND.

6.7 Chapter Summary

This chapter discussed the main findings of this study and their potential relevance for clinical practice. It revealed that people with MND perceive dysphagia in a different manner to their caregivers and professionals involved. It highlighted the psychosocial consequences of dysphagia in MND, which appeared to be particularly severe when the person with MND had to cease an oral diet. In addition, dysphagia affected both the people with MND and the caregivers' relationship with food. The people with MND reported their gradual detachment from considering food as a source of enjoyment and pleasure as their dysphagia progressed. They appeared to focus on the medicinal values of food in the hope that this would improve their survival or at least their stamina, leading to increased energy levels and ultimately improving their quality of life. The caregivers, on the other hand, perceived food as a severe risk to the person with MND, and therefore provided constant supervision during meals. Although both groups of participants reported their fear of choking, this fear was particularly apparent in the group of caregivers, who felt unsure as to how to assist the person with MND during choking episodes.

This chapter also outlined the issues reported in relation to dysphagia - related services along with suggestions as to how these services could consider the individual needs and wishes of participants. It also highlighted the complexity of managing dysphagia associated with MND, particularly in light of the gaps in the current evidence based practice and when working within limited resources.

CHAPTER 7: FINAL REFLECTION – STUDY CONTRIBUTION TO RESEARCH AND PRACTICE

In this chapter the contributions of this study are summarised from the researcher's perspective. The main findings are highlighted with regard to the study results and the methodological approach chosen to complete this study (IPA). Suggestions for future research complete this thesis.

7.1 Study Contribution

This was a qualitative study investigating the experiences of dysphagia in MND by interviewing 10 people with MND and 10 caregivers. The sample in both groups was diverse and included male and female participants who were (1) dealing with different levels of dysphagia (mild versus severe; eating orally versus non - orally), (2) at different life stages (working versus retired, having no family versus having a young family or a grown up family), (3) living in different geographical areas (city versus rural), (4) living with MND for varying lengths of time (a few months to a few years), and (5) presenting with different levels of engagement with professional services. This appears to be the first study providing an in - depth investigation into the dysphagia - related experiences of people with MND and caregivers of people with MND, where each participant was interviewed and analysed individually before a cross - case analysis of each group was conducted. This approach resulted in an in - depth understanding of participants' experiences while revealing both similarities and differences not only between the groups, but also between individual participants.

This in-depth investigation into participants' experiences uncovered issues potentially relevant to a wide spectrum of people living with MND in Ireland. The

study revealed that the people with MND and the caregivers perceive and manage dysphagia differently and often have different expectations from professional services. Dysphagia can potentially have a negative impact on both groups, especially if not managed utilising an individualised approach. The need for greater recognition of the significant role that people with MND and their caregivers play in the management of dysphagia emerged. Professional services should support the people in both groups by making an effort to develop good rapport and provide adequate training and guidance in a planned manner.

General awareness of dysphagia, its associated risks, and possible management options appears to be low in Irish society. Increased public awareness may not only reduce the stress related to having to deal with a completely unknown impairment, but may also result in more adequate recognition of the symptoms of dysphagia.

This study highlighted that dysphagia in MND should not be viewed and managed in isolation, but instead considered holistically.

This research confirmed the psychological and emotional consequences of dysphagia. In addition, it suggested that living with a progressive swallowing impairment can affect a person's relationship with food, which appears to influence the decisions made in relation to dysphagia - management options, such as non - oral feeding.

This study highlighted the views of people living with MND in relation to professional services received to manage dysphagia, which should provoke a reflection on the services currently provided.

It appears that the issue of non - oral feeding in MND has not previously been investigated in an in - depth manner from the perspective of caregivers. Stavroulakis

et al. (2014, 2016) carried out simultaneous interviews with people with MND and their caregivers. However, the pilot study conducted as part of the current research revealed a number of issues associated with interviewing participants in dyads (Chapter 2 section 6), which can potentially influence the data collected. This may be the first study investigating dysphagia related experiences of caregivers of people with MND by interviewing them separately.

The literature indicates that MND continues to have an impact on families following the death of the person with this disease (Hebert et al., 2005; Aoun et al., 2011). Considering that the emotional trauma experienced by caregivers can potentially extend beyond the life of the person with MND (Aoun et al., 2011), it is surprising that in Ireland there appears to be little support being offered to caregivers who have suffered a bereavement.

Finally, this study confirmed IPA as a suitable methodology to use in order to investigate the complex issues related to dysphagia in MND particularly as it allows for multiple in - depth interviews (Smith et al. 2009). It emerged that a single interview design may not provide adequate insight into the phenomenon of dysphagia in MND. IPA could potentially be employed for future research by allied health professionals, for example, investigating individual perceptions of particular therapeutic interventions or the presence of dysphagia in another multisystem disease. The benefits of IPA include a systematic approach to data collection and analysis, while allowing the researcher to make their own interpretations and use their professional knowledge when analysing the findings. The challenges, however, include potential difficulties with accessing the sample (as IPA emphasises particularly important life experiences meaning that participants may be under significant time pressure or distress), and the significant amount of time required to apply the idiographic approach

before moving to cross - case analysis. This study included twenty participants analysed each individually before conducting the cross - case analysis of each group, and finally compiled the findings in the discussion. This precise and multi - level data analysis meant that prolonged time to complete the analysis phase of the study was required. Breaks were necessary to be included, for example in order to transition from the idiographic to the cross - case analysis of each group. In addition, the researcher had to develop creative ways of visually presenting the data, such as through the use of mind - maps.

7.2 Implications for Future Research

This study highlighted a number of opportunities for further research. Firstly, it became clear that the clinical evidence pertaining to the management of dysphagia in MND is sparse, particularly in relation to specific dysphagia management options (both compensatory and therapeutic). Improved clinical knowledge could have a positive impact on the efficiency of dysphagia - related services. Moreover, potential changes in dysphagia management could also influence the lives of people with MND and their caregivers. Therefore it seems imperative to include the views of people living with MND when developing / reviewing dysphagia – related services.

This study demonstrated that the type and level of dysphagia - related services currently provided in Ireland may not address all the requirements of people living with MND in this country, particularly in terms of the timing of professional input. Although this study represented the perceptions of a group of people with MND and a group of caregivers, it seems necessary to conduct research representing the views

of professionals providing dysphagia - related services in order to obtain and analyse all perspectives.

It emerged that both people with MND and their caregivers would benefit from increased skills teaching in relation to managing dysphagia at home. It appears that it would be useful to develop and test a skills teaching programme which could minimise the need for direct involvement with professional services and at the same time optimise the independence of people living with MND. Reflecting the requirements reported by the participants in the current study, this programme should include techniques directly related to dysphagia (such as methods of improving the appearance of modified food textures or strategies to manage choking episodes at home), and also address the management of the psychological consequences of dysphagia (such as the feeling of fear or worry associated with meals). In addition, an effort should be made to carefully explain all dysphagia - related professional terms in order to ensure that both service users and professionals communicate using the same language.

The suitability of molecular gastronomy for people with dysphagia who require modified texture of food should be investigated in a wider manner, including not only cost - effectiveness analysis, but also the views of people with dysphagia.

This study demonstrated that some people with MND may be more concerned with their communication impairment rather than their dysphagia. Therefore more research investigating this phenomenon and which modes of support are perceived as being optimal seems warranted. In addition, it may be informative to investigate the impact of communication impairments on the caregivers of people with MND.

To gain an even more comprehensive understanding of the issue of dysphagia as part of a multisystem neurodegenerative disease, the experiences of people with

other illnesses such as Parkinson's disease or Multiple Sclerosis, could be compared. Similarly, the dysphagia - related experiences of the caregivers of people with such illnesses could also be compared and further explored. In addition, the dysphagia - related experiences of people with a cognitive impairment and MND and their caregivers should be investigated to reveal the presence of any specific requirements in relation to the management of dysphagia.

This study confirmed that the needs of people living with dysphagia as part of MND change over time. This creates potential challenges for the delivery of dysphagia - related services, which should adequately reflect the changing requirements of this population. A dedicated longitudinal research study seems necessary therefore, potentially combining quantitative and qualitative methodologies, and following participants over an extended period of time. Given the variable rate of progression of MND this longitudinal approach may be difficult to conduct on a large scale, however, it may create an opportunity for international collaboration.