

A Feasibility Study of Physiotherapy for Functional Motor Disorder

Glenn Nielsen

PhD Thesis

Sobell Department of Motor Neuroscience and Movement Disorders

UCL Institute of Neurology

I, Glenn Nielsen confirm that the work presented in this thesis is my own. Where information has been derived from other sources, I confirm that this has been indicated in the thesis.

Signature:

Date:

Abstract

Functional motor disorder (FMD) is a common cause of disability and distress amongst patients seen by neurologists and physiotherapists. Patients present with combinations of weakness, tremor, jerks, dystonia and gait disorder. The long term prognosis is poor.

Historically, psychological explanations for FMD have dominated, correspondingly, psychological therapy has been considered the treatment modality of choice, although evidence for its effectiveness is limited. A more recent understanding of FMD, considers symptoms within a broader biopsychosocial framework. This is backed by research into biological mechanisms that suggest FMD is associated with abnormalities in motor planning and agency, related to illness beliefs/expectations and abnormal self-directed attention. This broader conceptual model of FMD provides a rationale for physiotherapy treatment. A systematic review of the literature found that, while promising, the evidence for physical rehabilitation is limited, with a lack of randomised controlled trials.

This thesis describes a specific 5-day physiotherapy intervention that is based on a mechanistic understanding of how functional motor symptoms are generated. The intervention was tested in a randomised feasibility study with an embedded longitudinal qualitative study. Sixty patients were randomised to either the intervention or treatment as usual control. At six months follow up, feasibility was demonstrated by high rates of recruitment, retention and intervention acceptability. The intervention was associated with a significant improvement across a range of physical and quality of life outcome measures, with a moderate to large effect size. A health economic analysis showed evidence of likely cost-benefit. Findings from the qualitative study suggest that helping the patient develop a biopsychosocial understanding of their problem was an important ingredient of the intervention.

The findings from these studies add to the growing evidence for specialist physiotherapy for FMD and support the need for a multicentre randomised controlled trial.

Acknowledgements

The work in this thesis is a result of the inspiration, collaboration, generosity, support and help of many people, most importantly, my supervisors: Mark Edwards, Marta Buszewicz and Fiona Stevenson, thank you.

I would also like to thank,

Collaborators: Eileen Joyce, Rachael Hunter, Jon Marsden, Jon Stone, Alan Carson and Luciana Ricciardi

Kate Holt, for providing outstanding physiotherapy treatment

Magda Dudziec for data collection, data entry and trial management support

Patient and Public representatives on the Trial Advisory Committee, Chrissie McDonald and Hayley Thorne

The participants who volunteered their time to take part in the studies

Kate Hayward, who contributed to the analysis of the qualitative data

Anne Rodger and the Therapy Services Department at the National Hospital for Neurology and Neurosurgery for supporting the research.

Priment Clinical Trials Unit for advice on the study protocol and encouragement

The National Institute for Health Research, who funded this research (NIHR/HEE Clinical Doctoral Research Fellowship, GN, CDRF-2013-04-034).

The National Institute for Health Research Clinical Research Network (NIHR CRN) who provided funding for a research physiotherapist.

Table of Contents

Abstract	3
Acknowledgements.....	5
Table of Contents	6
List of Tables.....	9
List of Figures	10
List of Abbreviations	11
Thesis Aims, Objectives and Overview	12
Papers Published During PhD.....	16
Chapter 1 Introduction to Functional Motor Disorder	18
1.1 Terminology	18
1.2 Incidence and Prevalence of Functional Motor Disorder	21
1.3 The Cost of Functional Motor Disorder	22
1.4 Aetiology	23
1.5 Diagnosis	27
1.6 Malingering and Factitious Disorders	29
1.7 Clinical Presentations	29
1.8 Prognosis	33
1.9 Treatment.....	34
1.10 Summary	36
Chapter 2 A Brief History of Hysteria	37
Chapter 3 Literature Review	46
3.1 Methods	46
3.2 Results	47
3.3 Discussion.....	73
3.4 Conclusions	75
Chapter 4 Development of the Study Intervention	76
4.1 Introduction	76
4.2 Developing and Evaluating a Complex Intervention.....	77
4.3 A Theoretical Aetiological Model for Functional Motor Disorder	77
4.4 The Physiotherapy Treatment Protocol.....	86
4.5 Testing the Intervention: A Prospective Cohort Study	87
4.6 The next stage in the MRC Guidelines for Developing Complex Interventions	88
Chapter 5 Feasibility Study Methods	89
5.1 Introduction	89
5.2 Study Design and Setting	89
5.3 Ethics Approval.....	89
5.4 Participants	90
5.5 Randomisation and masking.....	91
5.6 Intervention Group	91
5.7 Control Group	93
5.8 Assessment.....	94
5.9 Outcome Measures.....	94
5.10 Statistical Analysis	97

5.11	Exploration of the Data	98
Chapter 6	Feasibility Study Results	99
6.1	Recruitment	99
6.2	Retention and Dropouts.....	99
6.3	Fidelity.....	101
6.4	Acceptability of the Intervention	101
6.5	Follow Up Assessment Timeframes	101
6.6	Safety.....	102
6.7	Treatment Received by Control Group	102
6.8	Baseline Characteristics of Participants	103
6.9	Continuous Clinical Outcomes	106
6.10	Categorical Clinical Outcomes.....	114
6.11	Health Economic Analysis: Cost-Utility Analysis	117
6.12	Health Economic Analysis: Client Service Receipt Inventory.....	118
Chapter 7	Feasibility Study Discussion	123
7.1	Key Findings Related to Feasibility.....	123
7.2	Clinical Outcomes.....	125
7.3	Health Economic Analysis	128
7.4	Mechanism.....	129
7.5	Limitations.....	130
7.6	Generalisability	131
7.7	Clinical Implications.....	131
7.8	Future Research	132
7.9	Conclusions	133
7.10	Acknowledgements and Contributions.....	133
Chapter 8	Qualitative Study – Introduction and Methods	134
8.1	Introduction	134
8.2	Methods	136
8.3	Sample Description	143
Chapter 9	Qualitative Findings: Prior to Treatment.....	145
9.1	Introduction	145
9.2	The onset of FMD	145
9.3	Early Interactions with Health Care Professionals.....	146
9.4	Resistance to Psychological Explanations	147
9.5	Feeling Unconvinced by the Diagnosis	150
9.6	Conflict and Marginalisation	151
9.7	Impact of Symptoms	152
9.8	Symptoms Remain Mysterious and the Uncertainty is Frightening	155
9.9	Powerlessness	156
9.10	Summary	156
Chapter 10	Qualitative Findings: Post-treatment	158
10.1	Introduction	158
10.2	Physical Changes	159
10.3	Ingredients of the Intervention.....	159
10.4	I Could See It Working.....	163
10.5	Understanding.....	164
10.6	Less Resistance to Considering Psychological Factors.....	166

10.7	A Work In Progress.....	167
10.8	Hope	168
10.9	A Positive Experience	168
10.10	Outlier	169
10.11	Summary	169
Chapter 11	Qualitative Findings: Six Months Follow-up.....	171
11.1	Introduction	171
11.2	Impact of the Intervention.....	171
11.3	Challenges of Putting it into Practice	178
11.4	Perceived Role of Psychological Factors	179
11.5	Feedback from Participants	180
11.6	Summary	183
11.7	Central Themes Across All Three Time Points	184
Chapter 12	Qualitative Study Discussion	188
12.1	The Impact of FMD.....	188
12.2	Resistance to psychological explanations.....	189
12.3	The importance of understanding	192
12.4	Legitimisation and Validation	194
12.5	From passivity to becoming active participants in rehabilitation	195
12.6	The impact of the intervention	196
12.7	Barriers to recovery and limitations of the intervention.....	197
12.8	Clinical and Research Implications.....	198
12.9	Limitations.....	202
12.10	Future Research	203
12.11	Conclusion	204
Chapter 13	Final Thesis Discussion and Conclusions	206
13.1	Introduction	206
13.2	Summary of Findings by Chapter	206
13.3	Synthesis of Quantitative and Qualitative Data.....	210
13.4	Thesis Conclusions	213
Appendix 1	Quality Appraisal of Studies with Subject Numbers of 10 or More	217
Appendix 2	Treatment Protocol.....	218
Appendix 3	Patient Information Sheet: Feasibility Study	227
Appendix 4	Consent Form: Feasibility Study	230
Appendix 5	Client Services Receipt Inventory (version used in this study).....	231
Appendix 6	Feedback Questionnaire and Results	236
Appendix 7	Brief Illness Perception Questionnaire Results.....	240
Appendix 8	Reflections on the Methodology	242
Appendix 9	Qualitative Study Patient Information Sheet.....	246
Appendix 10	Qualitative Study Consent Form.....	249
Appendix 11	Coding Framework.....	250
Appendix 12	Qualitative Interview Topic Guide	251
References	254

List of Tables

Table 1.1. Terminology related to Functional motor disorder	20
Table 1.2. Biopsychosocial aetiological representation of FMD ⁴¹	26
Table 1.3 Clinical Signs of Functional Motor Disorder ⁴³	27
Table 1.4. Classification of diagnostic certainty for functional movement disorders (Gupta and Lang, 2008).....	28
Table 2.1. Reynolds description of functional neurological symptoms ⁹⁴	40
Table 3.1. Literature Search Strategy.....	46
Table 3.2. Studies describing physical rehabilitation of FMD, listed in order of sample size.....	49
Table 3.3. Comparison of measures of disability at baseline	64
Table 3.4. Commonly described overarching treatment principles	68
Table 3.5: Outcome measures used in the literature	70
Table 3.6. Proportion of patients judged to have had a “good outcome” with treatment.	71
Table 5.1. Clinical outcome measures. Measures were collected at baseline, four week assessment and six month follow up (unless otherwise stated).	95
Table 6.1 Baseline demographic characteristics.....	104
Table 6.2. Baseline clinical characteristics	105
Table 6.3. Short Form 36 outcomes at baseline and six months.....	107
Table 6.4. Continuous outcome measures at baseline and six months	108
Table 6.5. Clinical Global Impression Scale at four weeks and six months.	114
Table 6.6. Contingency table of outcome by intervention at 6 month assessment	114
Table 6.7. Cases of anxiety and depression, as defined by Hospital Anxiety and Depression Scale sub scores of 10 or greater	115
Table 6.8. Contingency table of intervention group outcome at 6 months by anxiety caseness at baseline.....	115
Table 6.9. Contingency table of intervention group outcome at 6-months by depression caseness at baseline	115
Table 6.10. Baseline characteristics of the intervention group by outcome.....	116
Table 6.11. Services used and costs, data from Client Service Receipt Inventory ..	120
Table 6.12. Number of medications taken.....	122
Table 8.1. Purposive sampling criteria (participants could be represented in more than one box)	137
Table 8.2. Participant clinical and demographic characteristics.....	143
Table 8.3. Purposive Sampling Criteria Targets (participants fulfil multiple criteria).	144
Table 9.1. Participant's descriptions of the impact of FMD.....	152
Table 11.1. Central Themes Across All Three Time Points.....	185

List of Figures

Figure 1.1. Cycle of Development, Evaluation and Implementation of Complex Interventions.....	15
Figure 4.1 Tremor distraction and entrainment	78
Figure 4.2. Symptom explanatory model.....	85
Figure 5.1 Symptom explanatory model.....	93
Figure 6.1. Trial Profile	100
Figure 6.2. Hospital Anxiety and Depression Scale, Anxiety Score	109
Figure 6.3. Hospital Anxiety and Depression Scale, Depression Score	109
Figure 6.4. Work and Social Adjustment Scale	110
Figure 6.5. Berg Balance Scale	110
Figure 6.6. 10-Metre Timed Walk	111
Figure 6.7. Functional Mobility Scale	111
Figure 6.8. Disabilities of the Arm Hand and Shoulder (DASH)	112
Figure 6.9. Brief Illness Perception Scale, Total Score	112
Figure 6.10. Simplified Psychogenic Movement Disorders Rating Scale	113
Figure 6.11. Mean EQ-5D-5L utility scores	117

List of Abbreviations

B-IPQ	Brief Illness Perception Questionnaire
BMI	Body Mass Index
CBT	Cognitive Behavioural Therapy
CGI	Clinical Global Impression
CI	Confidence Interval
CSRI	Client Services Receipt Inventory
DASH	Disabilities of the Arm Shoulder and Hand
DSM	Diagnostic Statistical Manual of Mental Disorders
EEG	Electroencephalogram
EMG	Electromyography
FES	Functional Electrical Stimulation
FIM	Functional Independence Measures
FMD	Functional Motor Disorder
fMRI	Functional Magnetic Resonance Imaging
HADS	Hospital Anxiety and Depression Scale
HCP	Health Care Professional
HR	Heart Rate
I	Interviewer
ICD	International Classification of Disease
ICER	Incremental Cost Effectiveness Ratio
IQR	Interquartile Range
MCID	Minimum Clinically Important Difference
MRC	Medical Research Council
MRI	Magnetic Resonance Imaging
MRMI	Modified Rivermead Mobility Index
NES	Nonepileptic seizures
NHNN	The National Hospital for Neurology and Neurosurgery
NHS	National Health Service
PMDRS	Psychogenic Movement Disorders Rating Scale
QALY	Quality Adjusted Life Years
R	Respondent
RCT	Randomised Controlled Trial
SD	Standard Deviation
SF12	Short Form 12
SF36	Short Form 36
S-FMDRS	Simplified Functional Motor Disorders Rating Scale
TENS	Transcutaneous Electrical Nerve Stimulation
WSAS	Work and Social Adjustment Scale

Thesis Aims, Objectives and Overview

The research contained in this thesis aims to build on my previous work developing physiotherapy treatment for patients with Functional motor Disorder (FMD). This work started in July 2012 when, in collaboration with Professor Mark Edwards, I developed a specialist treatment programme for FMD, which was part of an NHS clinical service at the National Hospital for Neurology and Neurosurgery. The programme was assessed and reported in a cohort study of 47 consecutive patients.¹ The promising results led to an expansion of the clinical service and my successful application for a National Institute for Health Research - Clinical Doctoral Research Fellowship, to further develop the physiotherapy treatment and evidence for its efficacy.

Thesis Objectives

The primary aim of this thesis is to determine the feasibility of testing the 5-day Physiotherapy Programme in a randomised controlled trial (RCT).

The specific objectives of my thesis were:

1. To describe a standardised physiotherapy treatment protocol for FMD: the 5-day Physiotherapy Programme.
2. To assess the feasibility of conducting an RCT of the 5-day Physiotherapy Programme.
3. To assess the usefulness of a range of possible outcome measures to evaluate the effectiveness of physiotherapy for FMD and determine a primary outcome for a future trial (including the collection of data in order to perform a sample size calculation for a future RCT).
4. To assess the feasibility of completing a formal cost effectiveness analysis as part of an RCT.
5. To assess the acceptability of the intervention to patients.

6. To explore the lived experience of being diagnosed with FMD using qualitative semi-structured interviews.
7. To explore how participants interacted with the study intervention using qualitative semi-structured interviews.

Thesis Overview

To meet these objectives, I have completed 3 separate projects, (i) a systematic review of the literature for physical rehabilitation for FMD; (ii) a large randomised controlled feasibility study; and (iii) a longitudinal qualitative study of patients undergoing the physiotherapy intervention. This work is described in the following chapters:

Chapter 1 is an introduction to FMD, covering terminology, epidemiology, diagnosis, clinical presentations, prognosis and an overview of treatment.

Chapter 2 is a brief review of the history of FMD. It is argued that to understand the current status of FMD in health care settings, it is necessary to consider the historical context.

Chapter 3 is a systematic review of the literature for physical interventions for FMD.

Chapter 4 presents the theoretical aetiological model for FMD, which underpins the study intervention. The intervention protocol is briefly described.

Chapter 5 describes the methods for the randomised controlled feasibility study.

Chapter 6 reports the feasibility study results.

Chapter 7 is a discussion of the feasibility study results.

Chapter 8 introduces the qualitative study and describes the study methods.

Chapter 9 presents the qualitative findings from interviews conducted prior to treatment.

Chapter 10 presents the qualitative study findings from interviews conducted immediately after treatment.

Chapter 11 presents the qualitative study findings from interviews conducted at six months follow-up.

Chapter 12: is a discussion of the qualitative study findings.

Chapter 13 is the final thesis discussion and thesis conclusions.

The research plan for this thesis was devised with reference to the Medical Research Council's (MRC) guideline for developing and evaluating complex interventions.² The guideline identifies four key stages, (i) Development, (ii) Feasibility/Piloting, (iii) Evaluation and, (iv) Implementation, see Figure 1.1. The double headed arrows in the figure indicate that the stages are considered to be cyclical, so that interventions evolve over time, with continuous development, evaluation and implementation. The cycle starts with the development of an intervention, which involves identifying the evidence base and developing the theory that underpins the intervention, these are the topics of Chapters 3 and 4 respectively.

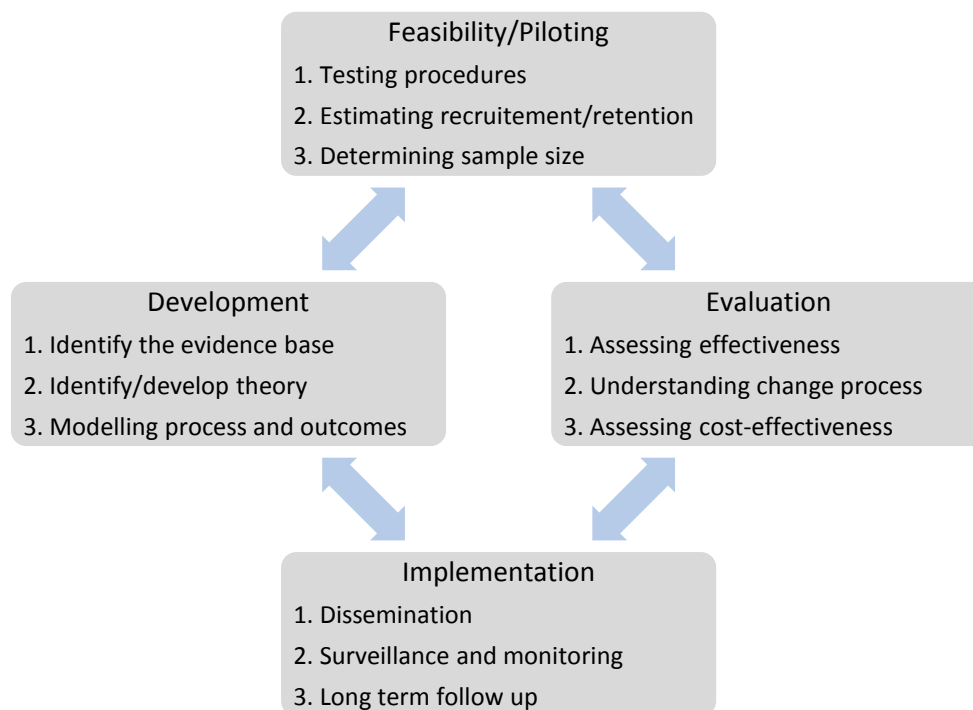


Figure 1.1. Cycle of Development, Evaluation and Implementation of Complex Interventions.

The thesis Feasibility and Qualitative studies (Chapters 5 to 12) aim to determine the feasibility of conducting, and inform the design of, an adequately powered, pragmatic, RCT of the study intervention. That is, to enable the progression to the Evaluation stage of the MRC guideline by conducting a definitive trial. Data from this research will also be used to further refine and develop the intervention.

Papers Published During PhD

March 2015

Nielsen G, Ricciardi L, Demartini B, Hunter R, Joyce E, Edwards MJ. Outcomes of a 5-day physiotherapy programme for functional (psychogenic) motor disorders. *J Neurol* 2015;262:674-681.

October 2015

Nielsen G, Stone J, Matthews A, Brown M, Sparkes C, Farmer R, Masterton L, Duncan L, Winters A, Daniell L, Lumsden C, Carson A, David AS, Edwards M. Physiotherapy for functional motor disorders: a consensus recommendation. *J Neurol Neurosurg Psychiatry* 2015;86:1113-1119.

September 2016 (Online first)

Nielsen G, Buszewicz M, Stevenson F, Hunter R, Holt K, Dudzic M, Ricciardi L, Marsden J, Joyce E, Edwards MJ. Randomised Feasibility Study of Physiotherapy for Patients with Functional Motor Symptoms. *J Neurol Neurosurg Psychiatry* 2016; Online first: jnnp-2016-314408.

December 2016 - Text Book Chapter

Nielsen G. Physical treatment of functional neurologic disorders. In: Hallett M, Stone J, Carson A, eds. *Functional Neurologic Disorders, Vol 139 of the Handbook of Clinical Neurology Series*. Amsterdam: Elsevier; 2016:555-569.

February 2017 (Online first)

Nielsen G, Ricciardi L, Meppelink A, Holt K, Teodoro T, Edwards MJ. A simplified version of the Psychogenic Movement Disorders Rating Scale: The Simplified Functional Movement Disorders Rating Scale (S-FMDRS). *Mov Disord Clin Pract*. *In publication*

2017 (in Publication) - Text Book Chapter

Nielsen G, Edwards MJ, Stone J. Functional disorders presenting to the stroke

service. In: Caplan LR, Biller J, Leary M, et al, eds. *Primer on Cerebrovascular Diseases, Second Edition*. In publication.

Chapter 1 Introduction to Functional Motor Disorder

Functional neurological symptoms are broadly defined as genuine neurological symptoms that lack internal consistency and are unexplained by a defined disease process.³ It is a diagnosis that exists on the boundary between the disciplines of neurology and psychiatry. Symptoms are diverse and can include disorders of movement, sensation and awareness. This research is concerned with functional symptoms affecting movement, which I will collectively refer to as Functional Motor Disorder (FMD). Patients with FMD typically present with one or a combination of weakness, tremor, jerks, spasms, dystonic postures or an altered gait pattern. Despite being amongst the most common diagnoses made in neurology,⁴ having been recognised as a discrete problem for hundreds if not thousands of years;⁵ and causing disability and distress equivalent to neurological disease;⁶ there is very little awareness and understanding of FMD. As a result there are few treatment options available for patients.

1.1 Terminology

Over the years many different terms have been used to describe patients with FMD, including hysteria, psychogenic, conversion disorder, somatization disorder, non-organic, medically unexplained, etc. Many of these terms remain in current use. While there is disagreement over which are most appropriate,⁷ the term ‘functional’ appears to be gaining traction. It was endorsed by the most recent version of the Diagnostic Statistical Manual of Mental Disorders (DSM-5) released in 2013, which updated its terminology from “Conversion Disorder” to “Conversion Disorder (Functional Neurological Symptoms Disorder)”.⁸ In addition, the beta draft version of the International Classification of Disease version 11 (ICD-11), due for release in 2017, has also updated its terminology to use functional.⁹

The term ‘functional’ is used to imply a change in function of the nervous system rather than structure. Its use has a long history in neurology; in the early 19th century, conditions that were classified as functional included chorea, migraine, tetanus,

epilepsy, as well as hysteria.¹⁰ In more recent times, it has come to represent those conditions that exist on the boundary between neurology and psychiatry.^{10,11} Opponents of the term “functional” have suggested it is ambiguous, that it obfuscates the true cause of the problem and can lead to a reluctance or refusal of the patient to accept the diagnosis and psychological treatment.⁷ The counter argument to this is that the current data suggests that psychological factors may not be of primary relevance to the aetiology and/or treatment in every patient with FMD, therefore terms such as psychogenic and conversion disorder are not necessarily appropriate. Perhaps more importantly, “functional” is the preferred term of patients and terms that imply a psychological aetiology are often considered unacceptable by patients.¹²

In this thesis I have opted to use the term “Functional Motor Disorder (FMD)”. This term is inclusive of the range of motor phenomenology seen in patients with functional neurological symptoms and it excludes functional symptoms that are not necessarily amenable to physiotherapy, such as attacks of decreased awareness (including nonepileptic/functional seizures), memory loss and confusion. The term “functional *movement* disorder” is often used in the literature, however a classical definition of a “movement disorder” excludes weakness, which is one of the most common functional motor symptoms.³

The lack of consensus of agreement of terminology reflects the heterogeneity of the patient population, as well the current state of limited understanding and differences of opinions in regards to the aetiology. The lack of consistency in terminology is confusing for patients and clinicians, but it also inhibits accurate clinical coding of these patients, inevitably leading to inadequate provision of health resources. The case for widespread adoption of the term functional is strengthened in Table 1-1 below, which lists common terminology and the associated problems with each term.

Table 1.1. Terminology related to Functional motor disorder

Terminology	Description
Functional	Used to imply a change in function to the nervous system, as opposed to structure. This term was shown to be less likely to offend patients, compared to other terms commonly used to describe these symptoms. ¹² However, it has been criticised for being ambiguous.
Conversion Disorder	Along with “Functional Neurological Symptom Disorder”, Conversion disorder is the term used by DSM-5. Some criticise this term for assuming an aetiology (conversion of psychic angst into physical symptoms) that is not supported by evidence. It has been described as a relic of late 19 th century Freudian psychoanalytic theory. ¹³
Dissociative Disorder	Dissociative (motor, sensory or seizure) disorder, is the term used in the ICD-10. ¹⁴ This term implies that symptoms are aetiologically related to the phenomenon of dissociation, where there is a subjective perception of a disconnection. ¹⁵ This term has been criticised for not representing all patients, as many do not recognise having dissociative experiences. ¹³ This may explain why the term has not been readily accepted by the medical community, rarely appearing in clinical correspondence or the scientific literature.
Psychogenic	In a survey of 519 neurologists, the term psychogenic was shown to be the preferred term for FMD by 83% when talking to colleagues, but only 59% used the term when talking to patients. ¹⁶ This suggests issues with patient acceptability of the term. The term assumes a psychological aetiology, which is not necessarily supported by evidence, or may be overly simplistic.
Pseudo-neurological	Meaning symptoms are not neurological in origin and implies symptoms are of a nonorganic basis. The prefix pseudo may be perceived as pejorative, by insinuating symptoms are not real.
Non-organic	Meaning symptoms do not have an organic basis. There is some argument over what constitutes an organic basis. In addition, this term and other similar terms such as non-epileptic are criticised for describing what the problem is not, which can leave the patient wondering what the problem is. ¹³
Psychosomatic	Originally meant to imply an interaction between mind and body, but is often used or perceived pejoratively. ¹³
Medically Unexplained symptoms	This term is used to describe functional symptoms from multiple body regions including pain, fatigue and gastrointestinal symptoms. It is often criticised for being a “non-diagnosis” and may be interpreted by patients as meaning the doctor does not know what is wrong. ¹³
Hysteria	An historical term, entering the English language during the early 17 th century, a time when FMD was presumed to be caused by “the wondering womb”. ⁵ The term encompassed many different presentations of functional disorders and remained in use into the 20 th century, long after uterine theories were dismissed. The meaning of hysteria has evolved to mean uncontrolled emotion.
Briquet’s Syndrome	Named after French physician Pierre Briquet (1796-1881) who completed a large epidemiological study noting that patients with hysteria were often polysymptomatic. Briquet’s Syndrome was a term used in the DSM synonymously with Somatisation Disorder (see below). It denoted patients with multiple functional symptoms in multiple body symptoms. The term was no longer in use by DSM-4. (1994) ¹⁷
Somatisation Disorder	A term used in DSM-4, the specific criteria for Somatisation Disorder included the patient having at least one “conversion symptom”, four pain symptoms, two gastrointestinal symptoms, and one sexual symptom, all before the age of 30. The term has been replaced in DSM-5 (the current version) with Somatic Symptom Disorder. ¹⁸
Somatic Symptom Disorder	In the DSM 5, Somatic Symptom Disorder replaced Somatisation Disorder. The plain English description includes “excessive thoughts, feelings, or behaviours related to somatic symptoms or associated health concerns”. This diagnostic label is widely criticised for being overly inclusive and pathologising normal behaviour. ¹⁸
Factitious Disorder	A psychiatric disorder distinct from FMD where symptoms are consciously fabricated for the purpose of receiving medical care. ¹³
Munchausen Syndrome	Alternative name for factitious disorder. Often described as a more severe form of factitious disorder, where patients seek hospital admissions and treatments. ¹³
Malingering	Feigning symptoms for material gain. ¹³

1.2 Incidence and Prevalence of Functional Motor Disorder

The incidence of a disease is the rate at which new cases occur in a population during a specified period.¹⁹ Functional motor disorder is generally considered to be common, but high quality studies of incidence are lacking. Epidemiological study of FMD is complicated by problems with definition and case ascertainment.²⁰ Defining FMD is complicated because diagnostic categories are often broad and therefore overlap with other diagnoses (e.g. psychogenic nonepileptic seizures, functional overlay in neurological disease, functional symptoms with organic comorbidity, and other functional or psychological syndromes such as chronic fatigue/myalgic encephalomyelitis). Case ascertainment is problematic as diagnosis often requires expert neurological assessment, which may not have been performed in studies conducted in primary care. Also, studies in tertiary care (where most studies of FMD are conducted) may miss patients who are managed in secondary or primary care.²⁰ Additionally, as previously discussed, many diagnostic labels are used for patients with FMD, preventing accurate coding of cases. Despite the obstacles in examining incidence, results from studies are relatively consistent, ranging from 4 to 12 per 100,000 population per year.²¹ To put this into context, the incidence of multiple sclerosis is estimated to be 4 to 6 per 100,000 in northern parts of North America and Europe²² and estimates of the incidence of Parkinson's disease range from 8-18 per 100,000.²³

Stone et al (2010)³ estimated a minimum incidence of functional weakness in adults of 3.9 per 100,000 population per year. This figure was based on 116 cases from a referral population of 1,261,191 in 28 months. While this study included all NHS neurologists in the referral population for South East Scotland, the authors note it may have missed patients who were not referred to the study or who were referred to other medical specialties (e.g. rheumatology). This figure was therefore considered a minimum incidence. Also the study only included functional weakness, therefore the incidence of all functional motor symptoms in South East Scotland can be presumed to be higher than 3.9 per 100,000.

A study of “disability due to motor conversion disorder” in Sweden found an incidence of 4.6 per 100,000 and 5.0 per 100,000 in two different Swedish district hospitals in 1997.²⁴

The proportion of the population that have the diagnosis at any one time point is referred to as the prevalence.¹⁹ Estimates of the prevalence of “conversion disorder” (inclusive of cases with non-motor functional neurological symptoms such as non-epileptic attacks) start from 50 per 100,000.²⁵

Functional symptoms are extremely common in clinical neurology settings. The Scottish Neurological Symptoms Study recruited 36 out of 38 neurologists working in the Scottish NHS and asked them to rate all new referrals over a 15 month period according to the degree to which the patient’s symptoms could be explained by disease. They found that 30% had neurological symptoms that were either ‘not at all’ or only ‘somewhat’ explained by neurological disease. Functional neurological disorder was the second most common diagnosis made, comprising 16% of patients and second only to headache (19%).⁴

1.3 The Cost of Functional Motor Disorder

Corresponding to the high incidence, FMD is widely considered to be associated with a substantial economic burden, though specific data is limited. The most commonly cited study to support this assertion is one that estimated the economic burden of medically unexplained symptoms in England, which included neurological symptoms as well as other functional symptoms such as gastrointestinal problems, fatigue and pain. The estimated annual total cost was estimated to be £18 billion for the period 2008-2009. This figure took into account healthcare use, quality of life effects and output losses. The cost of additional healthcare was estimated to be £3 billion per year, representing 10% of total NHS expenditure on healthcare services for the working age population.²⁶

Additional data that support the high costs of FMD, is found in a cohort of 1144 cases of functional symptoms from the Scottish Neurological Symptoms Study. Twenty-six percent were found to be unemployed due to ill health and 27% were in receipt of

disability benefits.⁶ In general these values were equivalent to patients with organic neurological disease, however patients with functional symptoms are usually more likely to be of working age.

1.4 Aetiology

Functional motor disorder is often primarily defined by an absence of disease, which does little to help understanding and treatment. When it comes to defining the actual mechanisms explaining symptoms, much remains unknown and there is disagreement over the relative importance of psychopathology.

It has been well established that psychological problems are more common in patients with FMD compared to the general population and patients with organic neurological disease. Kranick et al (2011)²⁷ compared 64 patients with “psychogenic movement disorder” (FMD), with 39 patients with organic focal hand dystonia, and 38 healthy volunteers, using a battery of psychological assessments. They found that patients with FMD scored higher in questionnaires assessing childhood trauma (specifically emotional abuse and physical neglect), as well as self-rated depression and anxiety. However, no difference was found between the groups across a large range of variables including the frequency of sexual abuse, frequency of physical abuse, a measure of the tendency to dissociate, and a self-reported measure of recent life events. There was also no difference between FMD and focal dystonia patients in the presence of interviewer-rated psychiatric disorders of major depression (lifetime), generalised anxiety disorder, phobia and panic disorder. This study and others have shown an inconsistent relationship between functional neurological disorders and psychological factors, for instance, childhood trauma appears to be more common in FMD, but the majority of patients are unaffected.²⁸ Also, average self-reported anxiety scores are higher in groups of patients with FMD compared to other diagnoses, but many patients with FMD still score within the normal (subclinical) range. These data suggest a pure psychological model is insufficient to understand FMD.

Factors other than psychological have become increasingly recognised as an important part of the aetiology. There are several studies that demonstrate a relationship between FMD and physical precipitating events, including pain, injury and neurological disease. In a systematic review, physical injury prior to symptom onset was found in 37% of 869 patients with functional sensory and motor symptoms.²⁹ In a study of 50 consecutive patients with FMD at the National Hospital for Neurology and Neurosurgery, 80% reported a physical event shortly before the onset of their motor symptoms.³⁰ The events included physical injury, infections, drug reactions and episodes of acute or exacerbated chronic pain.

Research into the neurobiological basis for FMD has become a topic of increasing interest. Modern functional imaging techniques hold promise as a method of studying the aetiology of FMD, however, this field of science is still in its infancy. To date, there have been over 20 functional imaging studies of FMD.³¹ While these studies are limited by small subject numbers and differing paradigms, a number of interesting findings have been reported. Patients with FMD appear distinct from those feigning neurological symptoms;³² they have shown abnormal recruitment of limbic areas, an abnormal functional connectivity between the amygdala and the supplementary motor area (a region involved in motor planning)³³ and finally, activity in areas associated with a normal sense of self-agency appear to be different in patients with FMD versus controls, which may be related to the patients' reported experience that symptoms feel involuntary.³³

An interesting finding from neurophysiological studies, that may be related to the lack of sense of agency, is that patients with FMD have been found to have impaired sensory attenuation.^{34,35} Sensory attenuation is where there is a difference in perception of identical sensory input depending on whether the input was self-generated or externally generated. Sensory feedback from self-generated movement is associated with a reduction in the perceived intensity (sensory attenuation), and this is believed to be how the brain labels a movement as self-generated rather than externally generated.³⁵ It has been demonstrated in repeated experiments that

patients with FMD lack normal sensory attenuation, which may explain why they experience symptoms such as a functional tremor as externally generated.^{34,35}

Other novel findings in some patients with FMD are impaired interoception (perception of sensations related to the physiological state of the body),³⁶ and higher rates of alexithymia (an inability to correctly interpret emotions).³⁷ These findings suggest that patients with FMD may show reduced awareness of internal body signals, which may lead to their misinterpretation.

A neurobiological aetiological model for FMD, incorporating the above findings, has been described by Edwards et al (2012).³⁸ The model highlights two key mechanisms that account for functional symptoms, which are (i) an abnormal attentional focus, and (ii) erroneous illness beliefs and expectations. The role of attention can be easily demonstrated, as functional motor symptoms require attention to manifest. When the patient's attention is distracted away from their symptoms, there is a reduction or disappearance of the movement disorder.³⁹ Expectation as a symptom mechanism relates to the patient's expectation or belief that their movement will be abnormal. The role of belief and expectation can be seen with commonly reported instant "curative" responses to placebo treatment, for example an instant response to botulinum toxin injections for fixed functional dystonia.⁴⁰ The mechanism by which belief and expectation may result in functional symptoms has been described in regards to the theory of active inference of brain function.³⁸ In brief, active inference refers to how the brain operates using predictive models rather than attempting to process the potentially infinite amount of afferent and efferent information available when performing functions such as controlling movement. Peoples' predictive models are based on prior experiences of interacting with the world. In the context of FMD, experiences may lead to a particular illness belief (e.g. of paralysis) and an associated expectation of abnormal movement. It is thought that the expectation of abnormal movement influences motor output at a preconscious level. The way in which expectations influence and shape motor output is thought to be related to the process by which the brain uses predictive "pre-programmed" models to generate or control movement. This concept can be likened to the experience of picking up an object that

you expected to be heavy but turns out to be light. The expectation is inaccurate resulting in overshooting the movement.

An alternative, biopsychosocial aetiological representation of FMD, considers the problem in terms of predisposing, precipitating and perpetuating factors. Each of these can be considered in biological, psychological and social categories. This biopsychosocial framework is helpful to understand the broader context of a patient's problem and to formulate the relevant issues within a clinical treatment context. See Table 1.2 below.

Table 1.2. Biopsychosocial aetiological representation of FMD⁴¹

Factors	Biological	Psychological	Social
Factors acting at all stages	"Organic" Disease (including underlying neurological disease)	Emotional disorder Personality disorder	Socio-economic/ deprivation Life events and difficulties
	History of previous functional symptoms		
Predisposing Vulnerabilities	Genetic factors affecting personality Biological vulnerabilities in the nervous system Persistent Pain Fatigue	Perception of childhood experience as adverse Personality traits Poor attachment / coping style	Childhood neglect / abuse Poor family functioning Symptom modeling of others
Precipitating Mechanisms	Abnormal physiological event or state (e.g., drug side effect hyperventilation, sleep deprivation, sleep paralysis) Physical injury / pain	Perception of life event as negative, unexpected Acute dissociative episode /panic attack.	
Perpetuating Factors	Plasticity in CNS motor and sensory (including pain) pathways leading to habitual abnormal movement Deconditioning Neuroendocrine and immunological abnormalities similar to those seen in depression and anxiety Pain and Fatigue	Illness beliefs (patient and family) Perception of symptoms as being irreversible Not feeling believed Perception that movement causes damage Avoidance of symptom provocation Fear of falling	Social benefits of being ill Availability of legal compensation Ongoing medical investigations and uncertainty Employment and financial issues

1.5 Diagnosis

In the past, FMD has been described as a diagnosis of exclusion but in recent times there has been a move towards making a 'positive' diagnosis. A positive diagnosis is generally based on clinical signs, incongruity with recognised neurological disease or internal inconsistency during the physical examination.¹¹ A number of clinical signs have been described to positively identify FMD. Perhaps the most clinically useful is Hoover's sign for functional lower limb weakness, which has been shown to have high specificity (99%) and sensitivity (94%).⁴² Other clinical signs of FMD are described in Table 1.3 below. Examples of incongruity with recognised clinical disease include midline splitting of sensory disturbance, global pattern of limb weakness and a tubular visual field.¹⁵ An example of internal consistency is when the patient is unable to actively plantarflex their ankle against resistance but is able to stand on their toes.

Table 1.3 Clinical Signs of Functional Motor Disorder⁴³

Clinical Sign	Description
Hoover's sign	Hip extension weakness that returns to normal when the contralateral hip is flexed against resistance.
Give-way weakness / collapsing weakness	Muscle power is initially generated on testing which quickly gives way or collapses.
Hip Abductor sign	Hip abduction power is tested bilaterally with the patient supine. Power in both limbs is generated in a positive test.
Dragging leg gait	Gait pattern characterised by dragging a weak leg behind.
Clear signs of inconsistency	For example, weak ankle plantarflexion on testing but the patient is able to walk on their toes.
Hemifacial muscle over activity presenting with unilateral limb symptoms	Over-activity of orbicularis oculi, orbicularis oris and/or platysma giving the appearance of a facial droop.
Sternomastoid test	Weakness of head turning to affected arm and leg in functional hemiparesis.
Drift without pronation test	During a 'pronator drift' test, the forearm may not pronate in a functional hemiparesis.
Global pattern of weakness	Flexors and Extensors equally affected - e.g. wrist flexion and wrist extension.
Tremor entrainment or distractibility	A distraction task, when tapping an unaffected limb at a set frequency, the affected limb entrains to the set frequency of tapping or the tremor stops. ¹¹
Dragging monoplegic gait	The leg is dragged at the hip behind the body. ⁴²
Walking on ice gait	Exaggerated postural responses, pivoting at the waist with a narrow base of support. ¹¹

In addition to clinical signs, key features of the history may support the diagnosis. This can include a history of other functional symptoms, rapid onset to peak disability and physical precipitating factors (such as a viral illness or minor injury).⁴⁴ Psychological problems such as anxiety, depression and recent stress are generally more common in functional disorder than other neurological diagnoses, but they are not universal and therefore are an unreliable basis for the diagnosis.⁴⁴ It is also generally recommended that specific and targeted investigations should be conducted to rule out other potential causes for symptoms, this may include MRI and nerve conduction testing.¹¹ Criteria for degrees of diagnostic certainty were proposed by Fahn and Willams (1988)⁴⁵, and were updated by Lang and Gupta (2009)⁴⁶, see Table 1.4 below. These criteria are useful for defining patient eligibility criteria for clinical trials.

Table 1.4. Classification of diagnostic certainty for functional movement disorders (Gupta and Lang, 2008)

1. Documented
Remittance with suggestion, physiotherapy, psychotehrapy, placebos, “while unobserved”
2a. Clinically established plus other features
Inconsistent over time/incongruent with clinical condition + other manifestations: other “false” signs, multiple somatizations, obvious psychiatric distrubance
2b. Clinically established minus other features
Unequivocal clinical features incompatible with organic disease with no features suggesting another underlying neurological or psychiatric problem
1 + 2a + 2b = Clinically definite
3. Laboratory-supported definite
Electrophysiological evidence proving a psychogenic [functional] movement disorder (primarily incases of psychogenic [functional] tremor and psychogenic [functional] myoclonus

It is often recommended that making the diagnosis of FMD should be the responsibility of a neurologist.⁴⁷ This is because of the potential complexity of ruling out rare neurological conditions that may mimic FMD, such as the alien hand phenomenon of corticobasal degeneration, and the variability seen in stiff person’s syndrome.⁴⁴ In

addition, on rare occasions, FMD may exist as a prodromal state for neurological disease. For example a functional gait disturbance and psychiatric symptoms may occur prior to the onset of firm signs of motor neurone disease, multiple system atrophy or Parkinson's disease.⁴⁴ More importantly, functional symptoms are often co-morbid with neurological (and other) disease, up to 15% of patients with an "organic" neurological disease diagnosis additionally have functional symptoms.⁴⁸ Despite these complexities, the diagnosis appears to be stable when made in neurological practice. A systematic review found that the rate of misdiagnosis is approximately 4%, which is in line with misdiagnosis rates of other neurological diagnoses.⁴⁹

1.6 Malingering and Factitious Disorders

Clinicians commonly report concerns that patients with FMD are feigning their symptoms.⁵⁰ The term malingering is used to describe the situation where symptoms are feigned for material gain and factitious disorders are when symptoms are feigned as part of a psychiatric disorder, for the purpose of receiving medical care.¹³ Many convincing arguments have been made to support the suggestion that FMD is distinct from feigning, this includes evidence from fMRI and laboratory studies.^{35,51} Some clinicians may interpret symptom variability or exaggeration of symptoms as evidence of malingering. Symptom variability can also be explained by the amount of attention invested in a movement or action. It may be true that some patients with FMD exaggerate symptoms to convince others of the need to be taken seriously, but this is different from exaggeration to deceive.¹³

1.7 Clinical Presentations

Functional neurological symptoms rarely occur in isolation. Functional motor, sensory and cognitive symptoms commonly coexist together with subjective reports of fatigue and persistent pain. When taking a history, the patient may focus on one particular problem, such as limb weakness, but on closer questioning, multiple symptoms are usually uncovered.

A number of specific presentations of FMD have been described. The most common are discussed below.

1.7.1 Functional Weakness

Functional weakness most commonly presents as weakness or paralysis of the limbs. It may occur in any combination including monoparesis, hemiparesis, paraparesis, and tetraparesis.⁵² The weakness is characterised by internal inconsistency, which differentiates it from weakness due to neurological disease. Examples of inconsistency include an inability to move a lower limb on testing but the patient is still able to stand. Clinical signs of functional weakness are described in Table 1.3.

Patients presenting with sudden onset of functional weakness are often mistakenly directed through emergency stroke treatment pathways. In this context patients are referred to as stroke mimics (other conditions that can also mimic stroke include migraine and brain tumours). Within specialist stroke services functional stroke mimics represent a reported range of 1.4 to 8.4% of all admissions^{53–57} and 0.5 to 5% of all patients who receive intravenous thrombolysis for presumed stroke^{58–60}.

1.7.2 Functional Tremor

Functional tremor as a dominant functional symptom is one of the more common presentations seen in tertiary specialist Movement Disorder clinics, accounting for up to 55% of patients with functional movement disorders.^{61,62} Functional tremor most commonly affects upper limbs, but can also affect the lower limbs, head and neck, and even the palate.¹¹ Functional tremor is characterised by distractibility and entrainability (see Table 1.3). It may occur at rest or with movement (action tremor). The tremor may be generated by alternating contraction of agonist-antagonist muscles or by co-contraction.¹¹

1.7.3 Functional Jerks (Myoclonus)

Functional jerks are intermittent jerking movements that can affect any part of the body, though jerks affecting the trunk are probably the most common (axial jerks).⁶³ They have been estimated to make up approximately 15% of patients with functional symptoms in specialist Movement Disorder clinics. As functional jerks are intermittent and have variable frequency, it is difficult to assess the effect of distraction, which makes diagnosis complex. A clinical diagnostic test has been devised for functional myoclonus which involves EEG-EMG back averaging to assess for cortical activity before movement. In patients with functional myoclonus and in normal voluntary movement, there is a pre-movement potential (also known as a Bereitschafts potential) arising approximately 1.5 seconds before activity.¹¹ This test is not suitable for all cases as it requires at least 30 recorded measurements and may not be possible when jerks occur at a greater frequency than 3-5 per second.¹¹

Many patients with functional jerks were previously diagnosed with myoclonus that was presumed to originate from the spinal cord, often called Propriospinal Myoclonus. The develop of EEG-EMG back averaging to assess for pre-movement potentials has resulted in many patients being re-diagnosed with FMD, and the existence of organic Propriospinal Myoclonus has been questioned.⁶⁴

1.7.4 Functional Dystonia

Functional dystonia presents as “dystonic” postures and movement that can affect virtually any body part, including the limbs, facial muscles, head, neck and the tongue. Functional dystonia commonly presents with other functional symptoms such as tremor, weakness and altered sensation.

1.7.5 Fixed Functional Dystonia

Fixed functional dystonia typically presents as a fixed (locked) abnormal joint posture (commonly ankle plantarflexion and inversion), accompanied by significant pain. It often overlaps with the diagnosis of complex regional pain syndrome.⁶⁵ Evaluation

under sedation relaxes the muscle tension and may reveal a soft tissue contracture, but often joint range of motion is unexpectedly preserved. Fixed dystonia commonly develops rapidly after a relatively minor injury, such as an ankle sprain.⁶⁶ Another common presentation of fixed dystonia is fourth and fifth finger flexion at the metacarpophalangeal and interphalangeal joints.⁶⁶ There is some disagreement over the aetiology of fixed dystonia and whether or not it is best characterised as a functional disorder.^{65,66} The debate draws on the cross over with complex regional pain syndrome and the inconsistent relationship of fixed dystonia with psychological factors.

1.7.6 Functional Gait Disorder

A functional gait disorder can present as part of a mixed functional movement disorder picture (due to functional weakness, dystonia, tremor, altered sensation, pain, etc) or as an isolated problem.⁶⁷ Common patterns of functional gait disorder have been described including a dragging leg gait, walking on ice gait (exaggerated trunk-sway postural adjustments without falling), astasia-abasia (inability to stand or walk despite normal power when tested on the bed), excessive slowness, non-economic postures, knee buckling and scissoring-leg gait.^{42,67}

1.7.7 Other Categories

Other categories of FMD have been described in the literature and used in clinical descriptions for research cohorts. Mixed Movement Disorder is often used to describe a mixture of motor symptoms including dystonia, weakness and tremor. Functional motor disorder can mimic Parkinson's disease and several case series of functional Parkinson's disease have been described. Patients exhibit a functional tremor (usually at rest and during action), slow effortful movement, abnormal response to postural stability testing and resistance to passive movement.⁶⁸

1.7.8 Non-epileptic Seizures and Other Common Symptoms

Non-epileptic seizures (NES) (also called dissociative/psychogenic/functional seizures) are a specific presentation of functional neurological disorder characterised by episodes of decreased awareness often associated with movement resembling tonic-clonic epileptic seizures.⁶⁹ Non-epileptic seizures are not considered part of the spectrum of functional movement disorders, due to the predominant characteristic of reduced awareness. However, symptom cross over is common and many patients with a primary complaint of NES will also experience functional motor symptoms and vice versa. In general, patients with a predominant problem of NES differ from those with FMD in a number of ways. They are more likely to be younger, more likely to report childhood abuse and more likely to report adverse life events prior to the onset of their symptoms.⁶⁹

It is also common for patients with functional neurological disorders (motor and non-motor) to experience functional symptoms in other body systems. Among the most commonly reported are gastrointestinal symptoms (many are diagnosed with irritable bowel syndrome), bladder retention and chronic fatigue. Many patients will also have other medical conditions and complaints that may need separate diagnosis and treatment in addition to their FMD. These conditions may act as one of several risk factors for the individual for developing FMD. Common comorbidities in FMD include migraine and Ehlers Danlos syndrome. In this context anxiety, depression and other psychiatric diagnoses may also be considered as separate comorbidities.

1.8 Prognosis

The prognosis of FMD is generally considered to be poor; a systematic review of prognosis found that approximately 40% of patients were the same or worse at long term follow up of 7 years and it would appear that the majority of patients remain symptomatic.⁷⁰ This data is based on a broad cross section of patients with FMD, including those with chronic symptoms at baseline assessment. There is limited data

on the prognosis of first episode acute onset FMD, a group that may have a higher rate of symptom resolution.

A number of prognostic indicators have been identified. Longer duration of symptoms prior to diagnosis and the presence of a personality disorder are among the most powerful predictors of poor outcome, while high satisfaction with care has been shown to predict positive outcome.⁷⁰

1.9 Treatment

There is limited evidence for the treatment of FMD. Multidisciplinary treatment is generally considered the gold standard, which may involve neurology, psychiatry, physical therapists (physiotherapy, occupational therapy and speech and language therapy) and psychology (or similar therapists); with the general practitioner at the centre of the team. Additionally, clinical nurses have an important role in inpatient treatment programmes.

1.9.1 Neurology

As described above (section 1.5 Diagnosis), neurologists are usually the clinician responsible for making the diagnosis of FMD. Several authors have presented arguments suggesting that neurologists have a key role in the management of patients with FMD and that this starts with the first step of treatment, which is effective communication and explanation of the diagnosis.^{11,15} Other roles of the neurologist may include treating comorbidity such as migraine and chronic pain; rationalising medications; referral to other medical specialities for assessment of comorbidity that has not previously been addressed (e.g. sleep apnoea); referral for psychiatric assessment and management if appropriate; and referral for multidisciplinary rehabilitation.

1.9.2 Psychiatry

The role of psychiatry will differ between patients and in the NHS may be dependent on the provision and commissioning of local services. The key role of a psychiatrist is to assess and treat psychiatric comorbidity. Psychiatrists may also oversee rehabilitation; the two specialist inpatient multidisciplinary rehabilitation programmes in the UK are led by psychiatrists within the psychiatry departments of tertiary hospitals.^{71,72}

1.9.3 Physical Therapists

The role of physical therapies is increasingly being recognised. In the UK, specialist services for FMD involve physiotherapy, occupational therapy and speech and language therapy.^{71,72} Specialist treatment programmes abroad include other clinicians, such as exercise therapists.⁷³ The evidence for physiotherapy treatment is reviewed in Chapter 3. The evidence for Occupational Therapy is limited to cohort studies of multidisciplinary inpatient rehabilitation programmes,^{71,72,74,75} which report promising outcomes. For Speech and Language Therapy, in addition to their involvement in multidisciplinary cohort studies,^{37,72} there are several cohort studies showing promising treatment outcomes, in particular for functional stuttering.⁷⁶

1.9.4 Psychological Therapies

Psychological therapies can be an important part of treatment for patients who consider that psychological factors are relevant to their problem. A randomised controlled trial of a CBT-based, guided self-help intervention for patients with a wide range of functional neurological symptoms, including motor symptoms, showed benefits in subjective health at 3 months and the physical function domains of the SF36 questionnaire at 3 and 6 months. The number needed to treat was 8.⁷⁷ There is some evidence for psychodynamic interpersonal therapy combined with neurological consultation,^{78,79} though these studies have low numbers and other limitations due to pragmatic experimental designs. There is better evidence for psychological treatment of the specific functional presentation of NES.⁸⁰

1.9.5 Other Treatments

A range of other treatments have been described for FMD. This includes transcranial magnetic stimulation,⁸¹ therapeutic sedation and abreaction,⁸² and hypnosis.⁸³ Controlled evidence for these treatments is currently lacking, however they may prove to be useful treatment adjuncts in some patients.

Alternative therapies such as reflexology are popular amongst some patients and often patients turn to alternative therapies when they have been unable to find support in the NHS. Alternative therapies can be a contentious issue, particularly in a cohort of patients that are vulnerable to iatrogenic harm and financial exploitation by unscrupulous providers.

1.10 Summary

Functional motor disorder is a common cause of disability for which there are few evidence based treatment options currently available. The aetiology is multifactorial, but historically it has been considered a problem predominantly of psychogenic origin. In the past 10-15 years however, neurobiological mechanisms for functional motor symptoms have been described and supported by clinical and laboratory evidence. This has led to a biopsychosocial understanding of FMD and opened the door to the development of physical as well as psychological treatments.

Chapter 2 A Brief History of Hysteria

Functional motor disorders have a long and colourful history, full of charismatic characters, many of whom have been remembered as the early pioneers of neurology and psychiatry. Below I will review significant milestones that have led to the current day understanding and status of FMD within the health care system.

It is often reported that the first descriptions of symptoms now recognised to be FMD come from ancient Egyptian papyrus, dating from 1900 BC.^{5,17,84} The symptoms included choking, mutism and paralysis, and were attributed to spontaneous movement of the uterus within the body.⁵ FMD was also described in ancient Greek and Roman texts. It was believed that the symptoms were caused by a lack of normal sexual contact, which resulted in the uterus migrating upwards causing multiple bodily symptoms. This is recorded in Plato's *Timaeus* (360 BC):

“... the matrix or womb, as it is called, which is an indwelling creature desirous of child-bearing, remains without fruit long beyond the due season, it is vexed and takes ill; and by straying all ways through the body and blocking up the passages of breath and preventing respiration it cases the body into the uttermost distress, and causes, moreover all kinds of maladies; until the desire and love of the two sexes unite them.”^{85,86}

The association between FMD and the uterus was commonly accepted until well into the 19th century. This obviously confined the diagnosis to women and gave rise to the label *hysteria*, derived from the Greek and Latin words for uterus. The term hysteria came into usage in the English language around 1615 but was used in French literature around 1568.⁵

During the Middle Ages (5th to 15th century) and the early Modern Period (16th to 19th century), beliefs about the uterus were replaced by beliefs of demonic possession and witchcraft. “Hysterical” women were subjected to exorcisms in an attempt to cure them or punish them for sorcery.⁸⁴ Often confessions were

obtained under torture, before the victim was put to death. With the Renaissance and emergence of scientific thinking, the uterus once more became the focus of descriptions of hysteria. However, examples of accusations and belief in demonic possession continued to appear, perhaps most famously in the witch trials of Salem Massachusetts in 1692.⁸⁴ Cultural and religious beliefs remain influential in how some cultures treat people with FMD today.

Edward Jordan an English physician and chemist (1569-1633) produced what is regarded as the first English work on hysteria in “The Suffocation of the Mother” (1603).¹⁰ He argued that hysteria was a natural disease, within the scope of medical study, rather than a theological issue. He suggested the uterus was the source of the problem, which in turn could affect organs such as the brain, heart and liver.^{10,87} Uterine theories of hysteria remained dominant until the 17th century, when the influential English physicians Thomas Willis (1621-1675) and Thomas Sydenham (1624-1689), promoted the brain and nervous system as “the seat of hysteria”, thus controversially suggesting that men could also be affected.^{87,88} Sydenham used the phrases “nervous distemper” and “animal spirits” when postulating on the mechanism of hysteria.

The naming of the brain and nervous system as the seat of hysteria led to the concept of nervous disorders.⁵ George Cheyne (1671-1743) a physician, mathematician and philosopher who was born in Scotland and practiced medicine in Bath wrote the influential “English Malady or a Treatise on Nervous Diseases of all kinds, as Spleen, Vapours, Lowness of Spirits, Hypochondriacal and Hysterical Distempers” in 1733.⁸⁹ Cheyne suggested that nervous symptoms arose from the moist climate, rich food and nervous culture of wealthy English society.⁵ It is considered that this suggestion may have provided a basis for a fashionable nervous culture, where fainting, swooning and attacks of the vapours became a fashionable way to behave,⁵ an interesting contrast to the stigma attached to mental illness in the present day. It was widely considered that hysteria only affected the upper classes, the lower classes were considered to be insufficiently impressionable and of too coarse a sensibility to fall victim to the disorder. In contrast, one hundred years

later the French neurologist Charcot and his contemporaries considered hysteria to be a disorder of the lower classes.⁵

The 19th century is often described as the heyday of Hysteria, when great interest and time was devoted to the subject by the pioneers of neurology and psychiatry. The French neurologists were particularly influential and much of the research occurred at the Parisian hospital La Salpêtrière. A comprehensive historical catalogue of French psychiatric dissertations written during the 19th century indicates that 20 percent were devoted to hysteria, and this was more than any other subject in psychiatry at the time.⁹⁰

Pierre Briquet (1796-1881) was also a neurologist and early pioneer of functional neurological symptoms. He became the chief physician at Charité hospital in Paris and reportedly undertook a study of hysteria as a matter of duty, on account of the frequency of cases.⁹¹ His published work “Traité clinique et thérapeutique de l’hystérie” (Clinical and Therapeutic Treatise on Hysteria) of 1859 was an epidemiological study of 430 patients with hysteria over a 10 year period.⁹² He noted that the patients were often poly-symptomatic and he believed that their symptoms were caused by the brain and related to nervousness caused by the emotional part of the brain.⁸⁵ He considered predisposing factors to be female sex, affective and impressionable temperament, family history, low social class, and situational difficulties.⁵ His work remained influential and he lent his name to the DSM diagnostic label “Briquet’s Syndrome”, characterised by multiple medically unexplained physical complaints in multiple body systems. This term remained in use until the release of version four of the DSM in 1994,⁹³ when it was replaced with Somatization Disorder, which has since been replaced with Somatic Symptom Disorder in version five of the DSM, released in 2013.¹⁷

Perhaps the next influential figure in the history of hysteria is Englishman John Russell Reynolds (1828-1896), a much respected early neurologist and professor of medicine at University College London.⁸⁵ Reynolds wrote a paper that was published in the British Medical Journal in 1869 titled “Remarks on paralysis, and

other disorders of motion and sensation, dependent on idea”.⁹⁴ In this paper he describes disorders of movement and sensation that would *appear* to be caused by disease of the brain or spinal cord, but instead “may depend upon a morbid condition of emotion, of idea and emotion, or of idea alone”. This paper presents an aetiological understanding that is strikingly similar to current biopsychosocial models for FMD.^{11,39} Similarities include highlighting the importance of belief as part of the symptom mechanism; that symptoms are often associated with biological events (e.g. physical precipitating factors) but are distinct from definite diseases of the nervous system; suggesting that psychopathology is common but not a necessary part of the aetiology; that symptoms are distinct from malingering; and that a positive diagnosis can be made based on characteristics of the motor symptoms. Reynolds suggested that treatment should be “A real, earnest dealing with the case, as one of grave character”. The doctor should give a confident expression of hope and positive reinforcement, facilitate early weight bearing “with support on each side, the amount of support [should] be gradually diminished day by day”. He also suggested the use of electrotherapy “partly as a moral and mental agent”, friction and passive movements. Reynolds’ summary of this condition, which he considered was neither neurological disease nor hysteria, insanity or malingering is outlined in his words in the table below. Reynolds’ paper was cited as an important influence by the French neurologist Jean-Martin Charcot.⁸⁵

Table 2.1. Reynolds description of functional neurological symptoms⁹⁴

<ol style="list-style-type: none"> 1. That some of the most serious disorders of the nervous system, such as paralysis, spasm, pain, and otherwise altered sensations, may depend upon a morbid condition of emotion, of idea and emotion, or of idea alone; 2. That such symptoms often exist for a long time, appearing as complicated disease of the brain or spinal cord; 3. That they resist many different kinds of treatment [such as pain not responding to morphine as one would expect]..., but that they disappear entirely upon the removal of the erroneous idea; 4. That they occur independently of anything that could be called either insanity of mind, hysteria, hypochondriasis, or malingering; 5. That they are often, but not constantly, associated with some bodily weakness or general debility;

6. That they sometimes associated themselves with distinct and definite disease of the nervous centres, so that it becomes very important to know how much of a given case is due to organic lesion, and how much to morbid ideation;
7. That it is possible to make a diagnosis with regard to them in many instances; and,
8. That the principles upon which their treatment should be conducted are simple, and their application marvellously successful.” (Reynolds 1869).

Jean-Martin Charcot (1825-1893) was an extremely influential figure in the history of FMD and has also been described as the premier clinical neurologist of the 19th century.⁹⁵ He spent his career at the Salpêtrière Hospital in Paris, where he built a much respected centre of neurological research. People came from around the world to hear his famous lectures and study with him. Among his students and collaborators were Georges Gilles de la Tourette, Pierre Janet, Joseph Babinski and Sigmund Freud, who all went on to make their own mark on the field of neurology and hysteria.⁹⁵⁻⁹⁷ During the 19th century, hysteria was a specific and defined neurological diagnosis largely due to the work of Charcot.⁹⁵ Outside of FMD, Charcot pioneered the classification of neurological symptoms according to anatomical lesions, based on post mortem studies, and he also established a category of neurological disorders, the “névroses” (neuroses) which were neurological conditions that were clinically characterised but still had no identifiable physical lesions, and included epilepsy, migraine and hysteria.⁹⁸

Charcot believed that a neurological basis for hysteria would eventually be found. He developed the concept of a dynamic lesion to explain paroxysmal symptoms,⁹⁷ a concept that some authors have likened to the findings from fMRI studies of abnormal neural circuitry.⁹⁵ He believed that suggestibility and susceptibility to hypnotism were pathognomonic of hysteria, although the pathology, in his opinion, remained grounded in neuroanatomy.⁹⁵ He used hypnosis to induce hysterical attacks during lectures and demonstrations as well as for treatment. Charcot had his detractors, including the British neurologist William Gowers, who practiced at Queen Square, and suggested that the displays of Charcot’s patients in his famous

lectures had been nothing to do with mental disorders, but were displays initiated by suggestion or feigned by the patient to please Charcot and his students.⁹⁷

After Charcot's death, his student the French neurologist Joseph Babinski (1857-1932) distanced himself from some of Charcot's work, in particular that there was a neuroanatomical basis for hysteria. He considered hysteria to be a psychological problem in which suggestion was important.⁹⁹ His views were influenced by his involvement in the treatment of soldiers suffering shell shock from the Battle of the Somme.⁹⁹ Babinski's contribution to hysteria included describing methods to differentiate hysteria from neurological disease.⁹⁶ Pierre Janet (1859-1947), another neurologist and student of Charcot's, emphasised the importance of psychological factors in hysteria and introduced the concept of dissociation as part of the explanatory mechanism.^{97,99} With the growing conceptualisation of hysteria as a psychological disorder, and the emergence of Sigmund Freud and psychoanalysis, hysteria started to move towards the field of psychiatry and became excluded from neurology.⁸⁵

The neurologist and founder of psychoanalysis Sigmund Freud (1856-1939) may be the most famous early clinician in the field of hysteria. Charcot is often credited with sparking Freud's interest in the area, after he spent time studying with him at the Salpêtrière in 1885.¹⁷ Despite being a neurologist by training, Freud's work made hysteria a psychiatric illness and his psychoanalytical paradigm dominated psychiatry's thinking for over 50 years.¹⁰⁰ Freud's ideas centred on repression of a traumatic idea and its conversion into physical symptoms. He highlighted the role of the unconscious to differentiate symptoms from feigning and initially considered sexual abuse to be an important trigger, although this became less prominent in his later work.¹⁰⁰ Together with the Austrian physician Josef Breuer (1842-1925), he published his famous manuscript "Studien über Hysterie" (Studies on Hysteria) in 1895, which included 5 case studies, including Breuer's famous case of Anna O. This work was seminal for the development of psychoanalysis and the "talking cure".¹⁰¹ Freudian theories about hysteria remained part of the DSM diagnostic criteria for conversion disorder until the most recent iteration in 2013.¹⁰⁰ In a recent textbook

chapter, psychiatrist Richard Kanaan (2016) suggests that with the removal of Freudian criteria, no obviously psychiatric features remain, once again hysteria is returning to the field of neurology where it had sat under the influence of Charcot.¹⁰⁰

The onset of World War I in 1914 saw an epidemic of functional neurological symptoms, with what became known as 'shell shock'. Soldiers presented with a variety of physical and psychological symptoms, including gait disorders, tremors, nightmares and panic attacks, that were the consequence of trench warfare.⁹⁹ Shell shock renewed the debate over whether hysterical/functional symptoms were conscious (malingering) or subconscious.¹⁰⁰ Treatments were influenced by the need to send soldiers back to the front, and often involved painful electrotherapy (Faradic currents). It is widely reported that the treatment was often delivered abusively, and that treatment often became to be considered as a form of punishment.¹⁷ The National Hospital for Neurology and Neurosurgery (at the time known as the National Hospital for the Paralysed and Epileptic) took in many patients with shell shock, with up to one third of the hospital allocated to military casualties.¹⁰² Lewis Yealland (1884-1954) a Canadian-born physician treated a large proportion of these patients. He described his treatment regimen as including walking exercises, re-education, suggestion, complete rest, isolation and strong faradic currents.¹⁰² This electrical therapy was often extremely painful and some historians have judged Yealland as having dealt out cruel punishing therapies. However, more recently Linden et al (2013)¹⁰², suggest that Yealland has been unfairly singled out, arguing that paternalistic and often painful treatment was standard practice for the time. Yealland played on soldiers' fear of being accused of malingering, by explaining to patients that, *if you recovery quickly, then it is due to disease, if you recover slowly, ... then I shall decide that your condition is due to malingering.*¹⁰² Interestingly, a similar deceptive approach to rehabilitation was described as current practice in a paper from Canada, published in 2004.¹⁰³

Following World War I, there was a common perception that hysteria had disappeared from medicine. It was argued that the florid symptoms described by

Freud and Charcot were a product of the repressed Victorian era and had since been replaced with more elusive symptoms such as fatigue.¹⁰⁴ However, Stone et al (2008)¹⁰⁴ noted that, although there is limited data on the incidence of FMD over time, the best evidence suggests that the proportion of patients presenting with FMD in neurology has remained strikingly similar over time, but that these patients became effectively invisible. Stone suggested a number of factors which might have contributed to the disappearance of FMD/hysteria from medical history, including the divergence of neurology and psychiatry leaving patients in a virtual 'no-man's land'; both neurologists and psychiatrists being notoriously uninterested in seeing patients with FMD;¹⁰⁴ and also the reported findings of the British psychiatrist Eliot Slater.

Eliot Slater (1904-1983) was a Psychiatrist at the National Hospital for Neurology and Neurosurgery.¹⁰⁵ Shortly before retiring, he gave a lecture, the transcript of which was published in the BMJ in 1965,¹⁰⁶ which described a follow up study of 112 patients diagnosed with hysteria over nine years. He reported that over half of the patients diagnosed with "hysteria" developed an alternative clear neurological or psychiatric diagnosis at follow up. Slater concluded, "The diagnosis of hysteria is a disguise for ignorance and a fertile source of clinical error." The paper has since been widely criticised for numerous flaws and poor methods of data collection.^{105,107} Unfortunately Slater's study was very influential and it has been suggested that it contributed to the almost complete disappearance of the concept of functional neurological symptoms from neurological curricula and text books.⁹⁹ The "disappearance" of functional neurological symptoms from scientific interest appeared to last until around the mid 1990's, when a resurgence of the topic can be seen in the scientific literature.⁹⁹ Modern imaging techniques, such as fMRI, have helped to develop an understanding of FMD to include theoretical and empirically testable neurobiological models, allowing for a biopsychosocial explanation for symptoms with no apparent organic basis.

There is however ongoing debate in the scientific literature about the classification of some symptoms as either functional or organic. Until the early 1980's, focal and

task specific dystonias were often considered psychogenic in origin. The British neurologist David Marsden (1938-1998) is credited with firmly establishing these as organic neurological conditions.¹⁰⁸ The syndrome of fixed (functional) dystonia has only relatively recently been widely recognised as a functional symptom,^{65,108} although many still consider this an 'organic' problem due to the diagnostic cross over with complex regional pain syndrome and the absence of a clear link to psychopathology. The development of modern neurophysiological assessments, has led to the condition known as propriospinal myoclonus, characterised by repetitive abdominal jerks, being considered a functional movement disorder in most cases.¹⁰⁹

In summary, the history of FMD dates back to the earliest medical records, yet these patients have never found a 'secure home' in medicine. In the late 19th century patients were embraced by neurology, before being moved towards psychiatry, where they sat uncomfortably, until recently when the pendulum has started to swing back towards neurology. This uncertain territory, where patients have found themselves, has no-doubt influenced the lack of awareness, understanding and status of FMD. Things, however, appear to be changing with the recently increased interest in FMD as a topic of research and with collaborations between neurology, psychiatry and rehabilitation specialists.

Chapter 3 Literature Review

A Review of the Literature of Physical Rehabilitation for Functional Motor Disorder

This review builds on my previously published systematic review of physiotherapy for FMD.¹¹⁰ Since publication, several new studies have been reported and the literature has been reviewed in light of this new evidence.

The research was reviewed in respect to the questions: (i) what is the evidence for physical rehabilitation of FMD? and (ii) what are the important ingredients of physical rehabilitation, as described in the published literature?

3.1 Methods

A systematic search of the literature was conducted. The search strategy from the earlier literature review (1950 to 5 September 2012) was extended to include the period of September 2012 to 15 March 2015. Condition terms were combined in an “AND” search with treatment terms. Output was limited to English language papers and studies of paediatric subjects were excluded due to potential differences in aetiology and corresponding treatment. The search was executed on databases: Medline (OVID and Pubmed), Embase and CINAHL. See Table 3.1 for search strategy. Additional searches were conducted using Google Scholar and the reference lists of published papers.

Table 3.1. Literature Search Strategy

Database	Results	Search Strategy (01/09/2012 – 15/03/2015)
Medline (OVID)	83	<u>Condition terms:</u> Exploded MeSH terms: “Conversion Disorder”, “Somatoform Disorder”, “Dissociative Disorder”. <u>Keywords:</u> “psychogenic”, “medically unexplained”, “hysteri*”
Medline (Pubmed)	88	<u>Treatment terms:</u> Exploded MeSH terms: “Physical Therapy Modalities”, “Rehabilitation”, “Physial and Rehabilitation Medicine”. <u>Keywords:</u> “physiotherapy”, “physical therapy”, “exercise”
Embase	507	
CINAHL	44	<u>Condition specific terms:</u> Exploded MeSH terms: “somatoform disorders”, “dissociative disorders”.

		<p><u>Keywords</u>: “psychogenic”, “medically unexplained” and “hysteri*”.</p> <p><u>Treatment terms</u>: Exploded MeSH terms “physical therapy”, “rehabilitation”. Keywords: “physiotherapy”, “physical therapy” and “exercise”.</p>
--	--	---

Being aware of limitations in the number and methodological quality of published studies on this topic, the inclusion criteria were kept deliberately broad. Studies were included if they described physiotherapy or physical rehabilitation of patients with an established diagnosis of FMD. Peripheral electrical stimulation aimed at improving movement was included in the definition of physical rehabilitation. Studies of multidisciplinary treatment inclusive of physiotherapy were also included. Studies were not excluded on the basis of low methodological quality. The abstracts (or full text when unclear) of all studies returned from the search were reviewed for inclusion. The original paper was retrieved for all studies fitting the inclusion criteria.

The following data were extracted from each study: (i) study type/methodology; (ii) treatment type; (iii) sample size; (iv) demographic characteristics of subjects (sex and age); (v) clinical characteristics of subjects (symptom phenotype, symptom duration and measures of disability); (vi) treatment parameters (setting, duration and frequency; (vii) treatment outcome; (viii) follow up duration; (ix) treatment ingredients; and (x) use of outcome measures. As part of the literature review process, a quality appraisal checklist designed for case series was applied to studies with subject numbers of 10 or more. It was felt that the appraisal criteria were not consistently applicable to small case series and case studies. This checklist and the scoring of studies with 10 subjects or more is given in Appendix 1 (page 217).

3.2 Results

The search criteria returned 722 hits including duplicates, from which 5 new studies were included in this review. An additional study, published after the search date was identified and included. Combined with the previous search, a total of 35 studies describing physical interventions for FMD were identified, published from

1970 to 2016, these are presented in Table 3.2. These studies represent a pooled population of 564 subjects. The majority of studies took place in an inpatient setting, with only 4 studies set in outpatients. There was only one RCT, which was a cross over design.⁷³ There was one cohort study with a historical control group,⁷⁵ the remaining studies were case series (n=25) and single case studies (n=8). Meta-analysis was not considered possible due to limitations in the reporting of the studies and the inconsistent use of outcome measures. A narrative analysis was therefore conducted. Studies with subject numbers greater than 10 (n=12 studies) are described in more detail.

Table 3.2. Studies describing physical rehabilitation of FMD, listed in order of sample size

Reference	Type	Symptoms	Treatment	n	Setting	Symptom Duration	Treatment duration	Outcomes	Follow up
Demartini et al 2014 ⁷¹	Prospective cohort study	Mixed movement disorder including weakness	4 week inpatient multidisciplinary rehabilitation involving physiotherapists, occupational therapists, cognitive behavioural therapists and nursing overseen by psychiatry with input from neurology.	66	IP	4.8 years	4 weeks	Two-thirds of patients rated their general health as better or much better on a 5 point CGI at discharge, which was maintained at 12 month follow up. 45% of patients were lost to follow up.	12 months
Jordbru et al 2014 ⁷³	Randomised cross over trial	G (100%)	3 week inpatient rehabilitation described as adapted physical activity within a cognitive behavioural frame work. Intervention carried out by physicians, physiotherapists, occupational therapists, nurses and an educator in adapted physical activity.	60	IP	9.5 months (12.1 SD)	3 weeks	Treatment resulted in significant improvement in physical function (FMS and FIM) and quality of life (SF12). Improvements were sustained at 1 month and 1 year follow up, except for the mental health domains of the SF12.	12 months
Czarnecki et al 2012 ⁷⁵	Retrospective cohort study with historical control group	G (40%) MD(43%) W (10%)	Physical therapy and occupational therapy with a motor reprogramming approach.	60	OP	17 month median (range 1-276)	5 days	68.8% markedly improved or nearly normal at end of treatment, 60.4% at follow up (self rated). This group was compared to a historical control group of 60 patients who did not receive treatment. 21.9% of this group were markedly improved at follow up.	25 month median (range 10 -64)

Nielsen et al 2015 ¹	Prospective cohort study	G (34%) D (25%) T (19%) W (9%) MD (13%)	5-day specialist physiotherapy based programme consisting of education and movement retraining with a self-management focus.	47	IP	5.5 years (SD 6.7)	5 days	65% rated their symptoms as “very much improved” or “much improved” on a 7 point CGI scale at the end of treatment. This reduced to 55% at 3 month follow up. This corresponded with significant improvement physical scales and self-reported outcome measures.	3 months
Moene et al 2002 ⁸³	2 arms of a randomised controlled trial (considered here as an uncontrolled cohort)	W (84%) G (56%) MD (42%) T (16%) D (18%) *	Hypnosis and MDT rehabilitation compared with MDT rehabilitation alone.	45	IP	2 months-22 years (Mean 3.9 years)	12 weeks	Both groups improved significantly, the addition of hypnosis did not affect outcome. 65% of patients were substantially to very much improved post-treatment and 83.7% at follow up.	6 months
Shapiro and Teasell 2004 ¹¹¹	Prospective cohort study	W (72%) G (31%) *	Behavioural intervention including physiotherapy. A standard programme is compared to a strategic double bind where patients are told that recovery constituted proof of an organic aetiology and failure to recover was proof of psychiatric aetiology. Outcomes were reported as recovery, significant improvement or no improvement.	39	IP	9 Acute (less than 2 months) 28 Chronic (more than 6 months)	Not stated	Standard Programme was effective for acute patients only – recovery was seen in 8 of 9 acute patients and 1/28 chronic. Patients who did not improve with the standard programme transferred to the strategic protocol. 14/22 recovered. 2 patients who had the strategic protocol only recovered.	Nil
Matthews et al 2016 ¹¹²	Prospective cohort study	G & W	Inpatient physiotherapy that included an explanation of	35	IP	2 week median	Mean length of	Patients had a mean of 11.2 physiotherapy sessions (range	Nil

			the diagnosis, demonstration of reversibility of symptoms and movement retraining. Half the patients also had intervention from a neuropsychologist or liaison psychiatrist.			(range 1 day-10 years)	stay 18 days (range 2-62 days)	2-43). The mean MRMI went from 20 to 37 post-treatment (max score 40).	
Heruti et al 2002 ¹¹³	Retrospective cohort of consecutive patients	W (100%)	MDT rehabilitation	34	IP	Not stated	Not stated	26% complete recovery, 29% partial recovery, 44% unchanged, 5 patients were re-diagnosed as malingering.	Nil
McCormack et al 2013 ⁷²	Retrospective cohort of consecutive patients	Motor conversion disorder	Multidisciplinary rehabilitation on a specialist neuropsychiatric unit, median length of stay was 101 days. The core treatment was from neuropsychiatrists, psychologists, physiotherapists, occupational therapists and speech therapists if needed.	33	IP	48 months medium (IQR 19–72)	Median length of stay 101 days	Significant improvement in Modified Rankin Scale. There was also an increase in the proportion of patients mobilising unaided and an increase in proportion of patients independent in personal activities of living.	Nil
Saifee et al 2012 ⁷⁴	Retrospective cohort of consecutive patients	MD (68%) W (16%)	MDT rehabilitation	26	IP	68% had symptoms for more than 36 months	Mean 24 days	At long term follow-up 58% of patients reported that the programme had been helpful or very helpful.	Mean 7 years
Ferrara et al 2011 ¹¹⁴	Prospective cohort study	T (68%) D (63%) G (37%) *	Daily TENS to produce a tingling sensation without muscle twitch or pain.	19	OP	Mean 46 months, 66 (SD)	Not stated	Statistically significant improvement in the PMDRS after an average of 6.9 months (SD 4.7) of treatment. Symptom resolution occurred in approximately 25% of subjects.	Nil

Dalocchio et al 2010 ¹¹⁵	Prospective cohort study	MD (75%) T (25%)	Group walking exercise programme, 3 times/week.	16	OP	3-44 months (median 13)	12 weeks	Marked improvement in 62% of patients.	Nil
Espay et al 2014 ¹¹⁶	Proof of concept, prospective cohort study	T (100%)	Tremor "retraining" using visual biofeedback	10	OP	4.3 years (2.9 SD) range 0.8-10 years	Training sessions over 1-3 days, equating to 2-6 hours	Resolution of tremor in 3 subjects which remained at follow up of 4-5 months. Two subjects remained improved at 4-6 months. The remaining 5 patients relapsed.	3-6 months
Speed 1996 ¹¹⁷	Retrospective cohort study of consecutive patients	W (100%)	MDT rehabilitation	10	IP	0.5-112 weeks	4-22 days (mean 12)	All had Improved FIM-gait score post-treatment. 7 out of 9 patients maintained improvement at follow up.	7-36 months (mean=29)
Moene et al 1998 ¹¹⁸	Retrospective cohort study of consecutive patients	W (50%) D (25%) G (12.5%) D (12.5%)	MDT rehabilitation including hypnosis.	8	IP & OP	1.5-19 years (Mean 9 years)	4 weeks-6 months IP, OP sessions 2-66	One patient dropped out, the remaining 7 had good recovery. 3 had relapsed on follow up, 2 of these resolved with outpatient hypnosis.	7-0.5 years (mean 2.6 years)
Weiser 1976 ¹¹⁹	Retrospective cohort study of non-consecutive patients	W (100%)	Persuasion, suggestion, general rhythmic exercises and emotional support provided by a physiatrist.	7	OP	1 week-1 year	1-3 sessions over 2 weeks	Symptom removal in 5 of 7 patients.	1 month-7 years
Delargy et al 1986 ¹²⁰	Retrospective cohort study of non-consecutive patients	W (100%)	Physiotherapy focused rehabilitation.	6	IP	1-24 yrs	2-10 weeks	All improved and were independent in activities of daily living at follow up.	8-14 months
Withrington and Wynn Parry 1985 ¹²¹	Retrospective cohort study of non-consecutive patients	W (100%)	MDT rehabilitation	5	IP	1-6 yrs	1-6 months	Improved	6 months-3 years

Cardenas et al 1986 ¹²²	Retrospective cohort study of non-consecutive patients	W (100%)	MDT rehabilitation with behavioural shaping.	4	IP & OP	1-4 years	Unclear in each case. Subject 1, 12 weeks.	Improvement but not resolution of symptoms.	2-5 months
Fishbain et al 1988 ¹²³	Retrospective cohort study of non-consecutive patients	W (100%)	EMG Biofeedback with MDT rehabilitation.	4	IP	3.5-13 years	2 weeks-3.5 months	Functional improvements and relinquished assistive devices for walking.	4 months-4 years
Silver 1996 ¹²⁴	Retrospective cohort study of non-consecutive patients	W (50%) MD (25%)	MDT rehabilitation	4	IP	Acute to 3 months	Not stated	Improved	Nil
Watanabe et al 1998 ¹²⁵	Retrospective cohort study of non-consecutive patients	W (100%)	MDT rehabilitation	4	IP	Acute	Mean 11 days	All improved.	Nil
Teasell and Shapiro 1994 ¹⁰³	Retrospective cohort study of non-consecutive patients	W 1 MD 2	MDT rehabilitation with "double bind".	3	IP	2.5-10+ years	5 weeks-4 months	Some improvement, one "completely well".	2 months-2 years
Behr 1996 ¹²⁶	Retrospective cohort study of non-consecutive patients	W (100%)	MDT rehabilitation	3	IP	2-4 yrs	2-4 months	All improved	Nil
Ness 2007 ¹²⁷	Retrospective cohort study of non-consecutive patients	MD 2 W 1	MDT rehabilitation	3	IP	1 day-2 months	6-9 days	All patients improved.	3 months
Trieschmann 1970 ¹²⁸	Retrospective cohort study of non-consecutive patients	W (100%)	MDT rehabilitation	3	IP	18 months-6 years	4-6 weeks	Improved, 2 patients had relapses that resolved with treatment.	2 years

Vatine et al 1996 ¹²⁹	Retrospective cohort study of non-consecutive patients	W (100%)	MDT rehabilitation	2	IP	6 weeks-6 months	4-6 weeks	Subject 1 mild residual weakness and symptom free at follow up; Subject 2 symptom free.	4 weeks-6 months
Glennon 2011 ¹³⁰	Retrospective case study	T	Physiotherapy with a focus on gait re-education.	1	IP	4 weeks	Not stated	Ongoing improvement	Nil
Atan et al 2007 ¹³¹	Retrospective case study	W	Physiotherapy with TENS and faradic stimulation	1	IP	Approximately 6 weeks	7 days	Complete resolution, maintained at follow up.	1 month
Oh et al 2005 ¹³²	Retrospective case study	G	Physiotherapy with a focus of functional training.	1	IP	Not stated	5 weeks	Improved	Nil
Sullivan and Buchanan 1989 ¹³³	Retrospective case study	W	MDT rehabilitation	1	IP	23 years	4 weeks	Improved mobility and independence in ADL. Dysphonia did not respond to treatment. Sustained at follow up.	1 year
Khalil et al 1988 ¹³⁴	Retrospective case study	W	Functional electrical stimulation and MDT rehabilitation.	1	IP	4 years	2 weeks (3 sessions a week)	Resolution of symptoms, sustained at follow up.	3 years
Findlater 1986 ¹³⁵	Retrospective case study	W	MDT physical and behavioural treatment programme.	1	IP	2 years of physical symptoms	35 weeks	Independent in mobility without aids and all activities of daily living.	7 months
Klein et al 1985 ¹³⁶	Retrospective case study	G	MDT rehabilitation	1	IP	16 years	11 weeks	Attained all goals including normal gait pattern.	2.5 years
MacKinnon 1984 ¹³⁷	Retrospective case study	W	Physiotherapy with psychiatric treatment	1	IP	1 week	6 weeks	Return to walking	Nil

Abbreviations: W=Weakness/Paralysis; MD= Movement Disorder (mixed); T=Tremor; D=Dystonia; G=Gait; IP=Inpatient; OP=Outpatient; FMS=Functional Mobility Scale; MRMI=Modified Rivermead Mobility Index; FIM=Functional Independence Measure; SF12=Short Form 12; CGI=Clinical Global Impression; PMDRS=Psychogenic Movement Disorders Rating Scale; MDT=Multidisciplinary Team; TENS=Transcutaneous Electrical Nerve Stimulation; EMG=Electromyography. * Unable to classify the primary motor disorder as frequency of symptoms were listed.

3.2.1 Randomised Controlled Trial of Physical Rehabilitation

Jordbru et al 2014

Jordbru et al⁷³ conducted the only randomised trial reported in the literature. In this study they tested a 3-week inpatient rehabilitation programme on 60 patients with a functional gait disorder. Patients were randomised to receive either the treatment or a 4-week waiting list control. After the 4 weeks, the controls crossed over into the intervention group. The exclusion criteria were: age over 69 years, symptom duration greater than 5 years, presence of a comorbid medical condition and if the patient did not want to take part in active rehabilitation. Patients were also excluded if they “needed inpatient psychiatric treatment”, a criterion that was not operationalised. The intervention was described as adapted physical activity with an educational and cognitive behavioural frame of reference. The treatment team consisted of a physician, physiotherapist, occupational therapist, nurses and an educator in adapted physical activity. Treatment included sporting activity such as bicycling, canoeing and indoor rock climbing. The authors describe using a behavioural approach in treatment, where improvement and desired behaviours were positively reinforced and positive reinforcement was withheld where no improvements were made.

Of the 60 randomised patients, 46 were followed up at one month and 40 at one year. Eighty percent were women, 20% were in receipt of a disability pension, 64% employed and the mean symptom duration was 9.5 months (SD 12.1). Subjects were assessed using the Functional Mobility Scale, the Functional Independence Measure (FIM) and the Short Form 12 (SF12). Data were collected at baseline, admission, discharge, 1 month and 1 year. Data were analysed using a covariance matrix model, to determine a treatment effect (the difference between treatment and no treatment within subjects, as each patient was exposed to a period of treatment and no treatment) and a carry over effect (treatment effect at one year follow up). The Functional Mobility Scale gives a score between 3-18, based on assistive device required to walk over 5, 50 and 500 metres, with a higher score indicating better function. The mean difference between treatment and no treatment was 6.9 Functional Mobility Scale units (95% CI 5.5, 8.3) and the carry over effect was 8.1 units

(95% CI 5.9, 10.3). The FIM is an 18-item scale of physical and cognitive disability, scored according to the level of assistance required to complete various activities of daily living. Each item is scored from 1-7, giving a score range of 18-126, with higher scores equating to more independence. The treatment effect for the FIM was 8.4 units (95% CI 5.2, 11.7) and the carry over effect was 9.2 (95% CI 5.4, 13.1). The SF12 is a condensed version of the Short Form 36, a health related quality of life questionnaire. The questionnaire yields a physical health and a mental health composite score, each out of 100. For the physical health score, there was a significant treatment effect (11.7 units, 95% CI 7.2, 16.1) and carry over effect (14.1 units CI 5.9, 22.2). The mental health score had a more modest treatment effect of 6.9 units (95% CI 2.1, 11.8) and the carry over score was not significant.

This study provides the first controlled evidence of physical rehabilitation for FMD. Specifically, this study suggests that multidisciplinary treatment with a physical focus, combined with a symptom explanation and positive reinforcement can provide improvement lasting at least 4 weeks in functional gait disorders of less than 5 years duration, in patients without significant psychopathology. The cross over design limits the control-intervention comparison to 4 week follow up only. This invites criticism over the necessity of treatment, in that both groups may have improved spontaneously over the follow up period without intervention. The counterpoint to this argument is the large body of evidence that patients with FMD tend to remain symptomatic without treatment,⁷⁰ while in this study benefit from treatment was maintained at one year. Another limitation is that a third of participants were lost to follow up (23% at one month and 33% by one year). The authors conducted a complete case analysis, meaning that subjects with missing data were excluded from analysis, potentially biasing the results. The authors justify this approach with a comparison to an observed data analysis, which suggested the dropouts did not result in a significant selection bias. An additional limitation was that the assessor was not blinded to patient group.

3.2.2 Cohort Studies of Physical Rehabilitation n>10

Czarnecki et al 2012

Czarnecki and colleagues⁷⁵ retrospectively report the outcome of 60 consecutive patients following an outpatient 5-day physical rehabilitation programme, consisting mostly of physiotherapy and occupational therapy, for functional movement disorders. To be considered for treatment, patients had to have a clinically established diagnosis and to have completed diagnostic testing. The functional movement disorders included gait disturbance, tremor, hyperkinetic movements and paresis. An important part of the treatment was the initial counselling of the patient. The important elements of this were: expressing confidence in the likelihood of success of treatment, a symptom explanation that was described as “a disconnect between brain and body” and an explanation of therapy as re-establishing this connection. The treatment consisted of 5 consecutive days of twice daily physiotherapy and occupational therapy. Treatment was described as motor reprogramming by establishing elementary movements and building on those. This included distracting motor tasks such as tapping to extinguish abnormal movements. Repetition was described as important to reinforce gains and a behavioural component included positive reinforcement of desired behaviour and ignoring abnormal movements. A psychological or psychiatric evaluation was completed prior to treatment. The aim of the evaluation was to identify possible contributory factors and psychological issues that might interfere with the programme and recovery, but this did not form part of the intervention.

Outcome was assessed using a patient rated 5-point ordinal scale at the end of treatment and by mail or telephone at follow up of median 25 months. The scale range was: no improvement or worse ($\leq 25\%$ improvement), mild improvement (26-50% improvement), moderate improvement (51-75% improvement), marked improvement (76-95% improvement), almost completely normal/in remission ($>95\%$ improvement). Based on these ratings, a good outcome was defined as a marked improvement or better. At the end of treatment, 69% of patients rated themselves as having had a good outcome. Eighty percent of patients were followed up at a median of 25 months, of these 60% continued to have a good outcome and 25% were considered treatment failures, being no better than mildly improved. It was reported that 87.5% of patients continued to have some degree of abnormal movements. The cohort were mostly chronically affected, the median symptom duration was 17 months (range 1-276) and 30% were “work disabled”. Forty-three percent had a history of depression and 41%

had a psychiatric diagnosis such as anxiety or personality disorder. It was reported that 21 cases were excluded from this analysis due to departure from the treatment protocol, no further information was given. The authors compared this treatment group to a historical control group who did not undergo the treatment for reasons such as lack of insurance, logistical reasons such as work, travel or family issues, the treatment had not yet commenced or non-acceptance of the diagnosis. The treatment group had significantly better outcomes. Arguably the control group is not a comparable group as it is likely to be affected by selection bias towards a worse outcome, especially due to the inclusion of subjects not accepting the diagnosis. Disregarding the historical control group and allowing for the limitations of this study, it provides uncontrolled evidence that physical rehabilitation can provide improvement lasting up to one year in selected patients.

Nielsen et al 2015

This paper presents the outcomes of our (Mark Edwards and Glenn Nielsen) 5-day physiotherapy programme for FMD in a prospective cohort of 47 consecutive patients.¹ The patients were admitted to the day hospital and those who did not live a commutable distance from the hospital stayed in a nearby hotel. The treatment was based on a symptom explanatory model for FMD highlighting self-focussed attention and expectations about movements as key aetiological mechanisms. Education using the symptom model formed the basis of the treatment and physical retraining aimed to normalise movement by progressive task retraining with redirected attention. The cohort was comparable to those in other studies in that 66% were women, the mean age was 44 and the mean symptom duration was 5.5 years (SD 6.7, range 2 months-40 years).

Outcome was assessed using a number of assessments. We used a 7-point clinical global impression scale to assess the patient's perception of change. The scale ranged from *very much worse* to *very much improved*. Based on this scale, 65% of patients were judged to have had a good outcome with treatment (a self rating of much improved or very much improved). This was reduced to 55% at 3-month follow up. In the SF36 there was a statistically significant improvement in the physical function and

physical role domains only (physical function: 26.1 SD 25.6 baseline, 35.0 SD 27.1 follow-up, $p=0.001$; physical role: 12.8 SD 26.5 baseline, 27.7 SD 31.4 follow-up, $p=0.001$). There was also a corresponding change in the (observer rated) physical assessments the Berg Balance Scale and 10 metre timed walk. This study provides further uncontrolled evidence of the effectiveness of physical rehabilitation, and uncontrolled evidence for treatment delivered by a physiotherapist. In addition it quantifies a treatment effect in the form of improved scores on the SF36 physical domains, Berg Balance Scale and 10 metre timed walk.

Moene et al 2002

Moene et al 2002⁸³ report the results of a randomised controlled trial of the addition of hypnosis to a 12 week inpatient multidisciplinary treatment for motor conversion disorder (which included paresis, gait disturbance, tremor and jerks). No additional effect was found for hypnosis and for the purposes of this review, the study is being counted as uncontrolled evidence for multidisciplinary treatment. The rehabilitation team consisted of a nurse, group therapist, creative therapist, sports therapist and physiotherapist. The group therapy consisted of psychological therapies (such as group psychotherapy and social skills training), creative therapy and sports. The physiotherapy was individual to the patient and described as step by step training of the lost function. When symptoms affected gait, patients were restricted to a wheelchair to prevent reinforcement of abnormal movement. Physiotherapy lasted one hour, 3 times a week during the first 6 weeks and 2 hours a week during the last 6 weeks. Patients were given homework and practiced their exercises 5 times a week.

The patients were 77% female, with a mean symptom duration of 47 months (SD 4.5 months, range 2 months to 22 years). The primary outcome measure was a video rating scale designed for the study, the Video Rating Scale for Motor Conversion Symptoms (VRMC). The scale rates symptoms during standardised movement on a 7-point Likert scale ranging from 1: unchanged to 7: very much improved. Based on this scale, 65% were judged to be “substantially to very much improved” at the end of treatment. This number increased to 84% at 6 month follow up.

Dalocchio et al 2010

The study by Dalocchio et al 2010¹¹⁵ stands out as one of only a handful conducted in an outpatient setting. The intervention was low to medium intensity progressive exercise in the form of outdoor walking in groups, conducted 3 times a week over 12 weeks. Participants (n=16) were required to be habitually sedentary at baseline and have functional movement disorders of mild to moderate severity. The primary outcome measure was the Psychogenic Movement Disorders Rating Scale (PMDRS), a standardised assessment of symptom severity scored blindly by video. There was a marked improvement in the PMDRS in 10 patients (62%), 2 of these patients experienced complete symptom resolution. In addition there was a statistically significant improvement in self-reported measures of anxiety and depression, improved VO2 max (maximal oxygen uptake) and body mass index. There was no follow up. As with the other studies described here, these results are interpreted with caution due to the relatively small sample size and lack of control. The cohort had characteristics that may be associated with a good prognosis, such as relatively short symptom duration (mean 15.5 months) and mild to moderate symptom severity. Taking into account these limitations, this study suggests that simple interventions such as nonspecific exercise and/or exercising in groups may be helpful for patients with mild to moderate symptoms who are habitually sedentary.

Shapiro & Teasell 2004

Shapiro & Teasell 2004¹¹¹ retrospectively reported outcomes of 39 consecutive patients who completed a rehabilitation protocol for “non-organic motor disorder”. Patients were initially entered into a standard rehabilitation programme where they were told that regardless of the origin of their disorder, their current symptoms were maintained by abnormal muscle patterns that had developed over time and that full recovery was possible with intensive inpatient rehabilitation. Treatment consisted of daily physiotherapy combined with positive reinforcement. If after 4 weeks there was no progress with this treatment, a *strategic protocol* was implemented, which involved telling patients that full recovery constituted proof of a physical aetiology and failure to recover constituted evidence of a psychiatric aetiology. It was explained to the

patient that their slower than expected progress was due to either the presence of a psychiatric problem called conversion disorder, or there was an aspect of their treatment programme that required modification, and once made, progress would be rapid and recovery complete. This change to treatment was “a minor and inconsequential change in physiotherapy”. There were 9 patients with acute symptoms (less than 2 months duration) and 30 patients with chronic symptoms (greater than 6 months). The standard protocol was effective for 8 of the 9 acute patients and only 1 of 28 chronic patients. Twenty-two patients went on to the strategic protocol (one acute, 21 chronic). The strategic protocol was reported as effective for 17 of 24 patients (71%) (some patients started in the strategic protocol). Of the 21 chronic patients who did not improve with the standard protocol and went on to the strategic protocol, 13 (62%) were completely or almost completely symptom free at discharge. This study did not use any standard assessment measures, outcome was subjectively judged by the authors as complete/near complete improvement, significant improvement, or minimal/no improvement and there was no follow up. It is therefore difficult to draw any conclusions or lessons from this study. The strategic protocol could be considered as deceptive, in that the clinicians were not upfront with the patients.

Matthews et al 2016

Matthews et al 2016¹¹² present a prospective case series of 35 consecutive patients with functional gait disorder and functional weakness who were referred to physiotherapy in an acute neurology ward over a 23 month period. On the whole, the cohort had a relatively acute presentation, with a median symptom duration of 2 weeks (range 1 day to 10 years) and 60% had had symptoms for less than one month. The mean number of physiotherapy sessions was 11.2 (range 2-43) and the mean length of stay was 18 days (range 2-62). Treatment included an explanation of the diagnosis, demonstration of reversibility of symptoms and movement retraining with distraction. The Modified Rivermead Mobility Index (MRMI) was recorded at baseline and discharge, there was no follow up. Half the patients also received intervention from a neuropsychologist or liaison psychiatrist, it was not stated how it was determined who received this additional treatment. The mean MRMI increased from

20 to 37 points, out of a possible 40. A ceiling effect was evident with this measure. Those with symptom durations greater than one month made less gains on average than the patients with more acute symptoms, but both groups made a statistically significant improvement. The addition of psychological intervention in this cohort did not have a statistically significant effect on outcome, as measured by the MRMI. This study demonstrates a large improvement in mobility score and differs from most of those reported here in that treatment was conducted on an acute hospital ward. This setting allows early treatment which may be advantageous given that long symptom duration has been associated with poor prognosis.⁷⁰ Follow up and control data from further studies is needed in order to be able to comment on the benefits of early intervention.

Heruti et al 2002

Heruti et al 2002¹¹³ report the results of 34 consecutive patients admitted to one of two rehabilitation centres in Israel, over a 27 year period, with suspected spinal cord injury, but later found to have no organic basis for their symptoms. The diagnosis of conversion disorder was made after rehabilitation had commenced and four patients were re-diagnosed as malingering. The spinal rehabilitation setting and perhaps other social and political factors may account for the higher percentage of male patients (74%). Treatment was provided by a multidisciplinary team consisting of physiatrists (rehabilitation physician), nurses, physiotherapists, occupational therapists, social workers and psychologists, with consultation from psychiatry. Outcomes were reported in terms of complete recovery, partial recovery and unchanged. Standardised outcome measures were not reported. Excluding the malingerers, 8 patients had complete recovery (27%), 8 had partial recovery (27%) and 14 patients did not improve (46%). There was no additional detail on treatment approach nor length of stay.

Specialist Multidisciplinary Treatment Programmes

Outcomes from two NHS specialist treatment programmes have been reported. These programmes comprise treatment from a multidisciplinary team, usually involving psychiatry, psychology (or psychological therapy from specialist cognitive behavioural

therapists), occupational therapy and physiotherapy. Some have additional input from neurology and speech and language therapy. These specialist programmes tend to accept patients with more significant comorbidity, such as higher physical disability and psychopathology that are excluded from some of the studies described above.

Demartini et al 2014⁷¹ report results from the 4 week inpatient treatment programme at Queen Square, London, in a prospective cohort of 66 consecutive patients. At one year follow up, there was a significant improvement with large effect size in the Health of the Nation Score (HoNOS), a clinician rated scale of outcome used in mental illness. In a patient reported clinical global impression scale at 12 months, 67% rated their general health as better or much better (points 5 and 4 on a 5-point Likert scale) and 64% rated their main symptoms as better or much better. There was also improvement in other self-report assessments. Only 55% of the 66 patients were followed up. In a separate cohort from the same treatment programme, a long term follow up assessment (median 7 years, IQR 4.5-8.5) was conducted via questionnaire.²⁶ Fifty-eight percent reported that the programme had been helpful or very. There was no change in employment status.

McCormack et al 2013⁷² report the results from the inpatient rehabilitation programme for conversion disorder at the Lishman Neuropsychiatry Unit, The Maudsley Hospital, London. In this retrospective review, 33 consecutive patients, were assessed pre and post-treatment using the Modified Rankin Scale, a 6-point scale ranging from 0=no symptoms to 5=severe disability, bedridden, incontinent and requiring constant nursing care and attention. There was a statistically significant improvement in mean scores from 3.64 (SD 0.86, range 2-5) to 2.83 (SD 0.85, range 2-5). There were also improvements in mobility and independence with activities of daily living. There was no follow up. This programme did not have a predetermined duration of treatment, the median length of stay was 101 days (IQR 84-130).

3.2.3 Patient Characteristics

Gender, Age and Symptom Duration

The published research on physical interventions for FMD includes 564 subjects, across 35 studies, of which 79% were women. The mean age and symptom duration at start of treatment was calculated where data was available. The mean age from 429 pooled subjects was 36 and the mean symptom duration from 336 pooled subjects was 36.5 months.

Disability

Level of disability or symptom severity at baseline may impact on prognosis and potential benefit gained from rehabilitation. This is difficult to quantify due to varying biological, psychological and social influences on disability amongst individuals. It is therefore often poorly described in studies. Baseline SF12/36 physical function scores may be a useful indicator of disability. This was reported in Jordbru et al (2014)⁷³, giving a value of 26.9 and Nielsen et al (2015)¹ with a value of 26.1. To provide some context to these values, normative data for a similar age range (35-44 years) is 89.4 (SD 16.1) for women and 91.9 (SD 14.5) for men.¹³⁸ Mobility aid use is reported in some studies as a measure of disability, usage rates ranged from 63%⁷³ to 85%.⁷² Employment status can be used as a surrogate measure of physical disability and mental health, though this will also reflect other factors such as comorbidity, chronicity, age and the social and welfare climate in the host country. A number of studies reported the percentage of patients not working due to ill health. These numbers are Jordbru et al (2014)⁷³ 20%, Czarnecki et al (2012)⁷⁵ 30% and Nielsen et al (2015) 64%. This data is presented in Table 3.3.

Table 3.3. Comparison of measures of disability at baseline

	SF 12/36 Physical Function Score	Wheelchair dependency	Mobility aid use including wheelchairs	Not working due to ill health
Jordbru et al 2014 ⁷³	26.9	25%	63%	20%
Nielsen et al 2015 ¹	26.1	-	-	64%
Czarnecki et al 2012 ⁷⁵	-	-	-	30%
Moene et al 2002 ⁸³	-	-	82%	-
McCormack et al 2013 ⁷²	-	61%	85%	88%*

* This figure did not differentiate unemployment due to ill-health from alternative reasons.

3.2.4 Symptom Phenomenology

In the literature, motor symptom phenomenology was usually broken down into the following categories: functional gait disorder, tremor, weakness/paralysis, dystonia or mixed movement disorder. The RCT by Jordbru et al (2014)⁷³ exclusively looked at functional gait disorders. The remainder of the studies with subject numbers greater than 10 had cohorts of mixed symptom phenomenology, except for Heruti et al (2002)¹¹³, which included symptoms due to suspected spinal injury only (and later found to have no organic basis). There is insufficient data to comment on whether one type of symptom responds more positively to physical rehabilitation or whether phenomenology may affect prognosis. There is however better evidence for the physical treatment of functional gait disorders based on the controlled study design by Jordbru et al (2014).⁷³

Classification of FMD phenomenology is not standardised and there is some cross over between categories. For example, a functional gait disorder may be comprised of functional tremor, weakness and/or dystonia. Classification may be unimportant if symptoms have a similar pathophysiological basis and respond similarly to treatment. However, there are reasons to suggest that this may not be the case, in particular for the specific symptom of fixed-functional dystonia. This symptom is often considered separately from other FMD for a number of reasons. Firstly, there is some disagreement over whether it fits more with an organic or functional aetiology. Also because it commonly crosses over with the diagnosis of complex regional pain syndrome.⁶⁵ No rehabilitation study has specifically looked at fixed-functional dystonia and it is unclear if studies have included patients with fixed-functional dystonia but classified them under labels such as *dystonia* or *mixed movement disorder*.

3.2.5 Treatment Parameters: Setting, Duration & Frequency

The majority of the 35 studies were conducted in an inpatient setting, with only 5 accounts of outpatient treatment,^{75,114–116,119} although some older studies described outpatient follow up after inpatient treatment.^{83,122} There were 2 studies of intensive treatment set over 5 consecutive days in either an outpatient⁷⁵ or day hospital¹

setting, with patients staying in a nearby hotel. This may be considered an intermediate step between inpatient and outpatient treatment. It is unclear if setting influences outcome, but an inpatient setting may enable higher intensity treatment regimens.

There were 6 purpose designed rehabilitation programmes for FMD, in all but one, length of stay was dictated by protocol. These were 12 weeks,⁸³ 4 weeks,⁷¹ 3 weeks,⁷³ and 5 days in length.^{1,75} One multidisciplinary inpatient programme had an open ended admission, the median length of stay was 101 days (IQR 84-130 days).⁷² Treatment provided outside of specialist programmes reported length of stays ranging from 4 days¹¹⁷ to 8 months.¹³⁵ It is therefore not possible to accurately define and compare the intensity of treatments based on the available data.

Outpatient treatments included; 5 consecutive days of physiotherapy and occupational therapy,⁷⁵ a group walking programme, which took place 3 times a week over 12 weeks,¹¹⁵ a tremor retraining with biofeedback study in which subjects attended 1 to 3 days of training equating to 2-6 hours¹¹⁶ and a study of transcutaneous electrical nerve stimulation (TENS), in which patients were set up with a device and instructed to use it daily for 30 minutes over 4 months.¹¹⁴

3.2.6 Composition of treatment

Explaining the Diagnosis and Education

The method of discussing the diagnosis with patients was considered an important part of treatment in several studies. In a previous literature review¹¹⁰ we categorised this approach into 3 groups. The first and most common was a direct approach that seeks to explain symptoms. Some explanations emphasised the importance of psychological factors, while others were skewed towards biological factors, such as “a disconnect between body and mind”⁷⁵ or the role of self-focussed attention in disrupting movement.¹ Most studies used a direct approach. The second approach was called a deceptive approach. In this, a diagnosis of FMD/conversion disorder was made, but deliberately withheld from the patient. Instead patients were presented

with a double bind, where they were told that if they recovered with rehabilitation it constituted evidence of a physical aetiology. Conversely, non-recovery was evidence of a psychological cause for their symptoms, requiring long term psychiatric treatment. Only 2 studies (from the same authors) were clearly deceptive.^{103,111} The third approach was described as a constrained approach, where no attempt was made to explain the origin of their symptoms and a structural disease process was not ruled out. Six studies followed this approach.^{113,120,124,133,135,137} These tended to be older studies and they were not part of treatment programmes set up specifically for FMD. Two studies described symptom education as being central to the intervention.^{1,112}

Overarching Principles of Treatment

Most studies described the importance of following broad principles that underpinned the treatment. See Table 3.4 for a list of commonly described treatment principles. One of the most prominent principles described in the literature is the behavioural approach to treatment, where positive behaviours and asymptomatic movement are praised and reinforced with attention, whilst undesirable behaviours and symptomatic movement are ignored.^{75,83,103,111,117,120–122,124–130,132,133,135,136} Shapiro and Teasell (2004) described how in their initial treatment protocol, *deep rest* was prescribed for patients who failed to meet their goals¹¹¹. This was considered an operant intervention, where all reinforcement for failure to progress was withdrawn by confining the patient to their bed without stimulation such as television, visitors or telephone contact. After a trial with 3 patients, they concluded that deep rest was unnecessary and removed it from their protocol. In some studies, behaviour modification was a dominant part of the intervention,¹²⁷ while in other it was supplemental to the other components of treatment.^{73,75}

The influential early paper by Trieschmann et al (1970), devised a treatment protocol for functional gait disorder which involved confining patients to a wheelchair outside of therapy sessions while their gait remained symptomatic.¹²⁸ This was designed to prevent reinforcement of symptomatic movement. As the patient progressed they were rewarded with walking privileges. This approach was instituted in several other studies.^{83,117,122,127,132,135,136}

Table 3.4. Commonly described overarching treatment principles

Principles of Treatment
Provide an explanation for symptoms ^{1,73,75,83,115,117,121}
Follow a behavioural approach with positive reinforcement ^{75,83,103,111,117,120–122,124–130}
Limit reinforcement of maladaptive movement patterns & behaviours ^{83,117,122,127,128,132,135,136}
Project confidence in the treatment ^{1,75,120}
Clear communication with patient and treating team ^{1,75,113,127,129,130}
Goal focussed rehabilitation ^{83,103,120,122,126–128,130,132,135,137}
Graded progression of exercise difficulty to reshape movement patterns ^{75,128,135,136}
Use of outcome measures to quantify and motivate ^{1,121,126,135,136}
Agree on a treatment contract/agreement at the beginning ^{83,113,120,133,136}
Involve the patient’s family in rehabilitation ^{83,111,124,127,128,130,136}
Allow for face saving ^{83,111,113,126,127}

Physical Treatment Strategies

Physical rehabilitation most commonly followed a motor relearning approach.^{1,75,83,111,112,117,122,127,128,130,132,136} This usually involved introducing elementary symptom free movements and gradually progressing the movements in stages, so that each stage progressively approximates normal movement. In their influential study, Trieschmann et al (1970) delivered this type of progressive motor learning approach in a very structured format.¹²⁸ Several stages of movement were prescribed in advance, the final stage being normal walking. The patient worked their way through these stages, but was not allowed to progress to the next stage until the previous stages had been mastered and they continued to be symptom free.

Some studies describe following treatment protocols of analogous organic conditions, for example stroke or spinal cord injury pathways,^{111,117,127,129} while others are structured specifically towards the diagnosis of FMD.^{1,71,73,75,83} Some studies described using distracting techniques to assist movement retraining, this included tapping or bouncing a balloon.^{1,75,126} Another treatment strategy described was positioning the patient on an unstable surface (such as a therapy ball) to elicit automatic postural responses.^{120,126,139}

There are few studies describing treatment strategies for functional tremor. Espay et al (2014), tested entrainment as a treatment approach using visual biofeedback in a small proof of concept study.¹¹⁶ The aim was to help the patient to develop volitional

control over the movement and bring it to a stop. This small study of 10 patients demonstrated improvements in tremor after three, 2-hour retraining sessions. Six patients reported lasting improvement at 6 months. Nielsen et al (2015) described retraining tremulous movement by imposing competing movements on top of the tremor and readjusting postures to change the frequency of the tremor.¹ A mirror was used for visual feedback.

Electrotherapies have a long history in the treatment of FMD and were initially used as aversive therapy.¹⁷ More recent descriptions of electrotherapies include TENS for sensory stimulus,¹¹⁴ electrical stimulation for muscle activation and function^{1,131,134} and EMG biofeedback to retrain movement.¹⁴⁰ Electrotherapy was described as a discrete intervention in only one study.¹¹⁴ In this study, a cohort of 19 patients with FMD were given a trial of TENS to elicit a sensory stimulation but not a muscle twitch. Fifteen patients continued to use the TENS, prescribed for 30 minutes daily for 4 months. One-third of these demonstrated a significant improvement in their symptoms (greater than 50% on a symptom scale) after a single trial. At 7 months follow up there was a statistically significant improvement in the PMDRS, and a quarter of patients had complete symptom resolution. This appears to be an unusually high success rate when compared to other studies with a similar cohort.

Nonspecific exercise is described as an important part of some of the interventions. It was central to the treatment for patients with mild to moderate FMD described by Dalocchio et al (2010), in the form of a group walking programme.¹¹⁵ In this study 62% of 16 patients were reported to have had a marked improvement in their symptoms immediately after treatment. Two studies included specialist instructors in physical activity,^{73,83} one of these included activities such as indoor rock climbing and canoeing as part of the intervention.⁷³ Some studies describe using specific strengthening exercises.^{111,127,132} Different therapeutic equipment described in the literature included a treadmill,¹ mirror,^{1,132} hydrotherapy,^{120,135} tilt table,¹²⁰ parallel bars,^{128,130,132} throwing and catching a ball or balloon,^{75,130} and a therapy ball.¹²⁰

Concurrent psychological treatment

Successful treatment has been described both with concurrent psychological therapy^{71,72,83,130} and without.^{1,75,112,115} Studies that do not include concurrent psychological therapy tend to exclude patients judged to have significant psychopathology, although it is difficult to determine exactly how the cohorts may differ in this regard. This is due to inconsistent use of standard psychological assessments and their inherent limitations. One possible comparison is the Hospital Anxiety and Depression Scale (HADS) scores on admission for a multidisciplinary treatment 15.8 (SD 8.5)⁷¹ and a physiotherapy delivered intervention 13.1 (SD 7.3).¹

3.2.7 Use of Outcome Measures

A variety of different outcome measures have been used in the literature, these are listed in Table 3-5 below. Many studies did not use any standardised assessment measures and described outcome subjectively in terms of recovery, partial recovery or non-recovery, rated by the clinician.^{111,113}

Table 3.5: Outcome measures used in the literature

Physical Outcome Measures	
Functional Independence Measure (FIM)	73,117,125,127,141
Berg Balance Scale	1,130
10 metre Walk Time	1,130
Functional Mobility Scale	73
Modified Rivermead Mobility Index	112
Psychogenic Movement Disorders Rating Scale	114,115
Video Rating Scale for Motor Conversion Symptoms	83
Gait Abnormality Rating Scale	132
Measures of physical fitness (BMI, HR, VO2max)	115
Dynamometry	122,134,140
Modified Rankin Scale	72
Patient Reported Outcome Measures (excluding explicit psychological measures)	
Clinical Global Impression Scale	1,71,75
Short Form 36, Short Form 12	1,73
Work and Social Adjustment Scale	1
EQ-5D-5L	1
International Classification of Impairment Disability & Handicap	83
Canadian Occupational Performance Measure (COPM)	71
Patient Health Questionnaire (PHQ-15)	71
Multidimensional Health Locus of Control Form C (MHLC)	114
Symptom Checklist 90	83
Revised Illness Perception Questionnaire (IPQ)	71
The Common Neurological Symptoms Questionnaire (CNSQ)	71

Measures of Anxiety, Depression and Other Psychological Assessments	
Hospital Anxiety and Depression Scale	1,71
Beck Anxiety Inventory	114,115
Beck Depression Scale	114
Hamilton Depression Scale	115
Health of the Nation Outcome Scale (HoNOS)	71
Dissociative Experiences Scale	114
Posttraumatic Stress Disorder Checklist (PCL)	114
Dissociative Experiences Scale	114
Fear Questionnaire	71

3.2.8 Outcome of Treatment

All the included studies (n=35) reported an overall positive effect of treatment in patients selected for rehabilitation. Some studies reported the proportion of patients that were judged to have had a good outcome with treatment. This was generally determined by a clinical global impression scale or a threshold score on a particular assessment. This data is presented in Table 3.6 below.

Table 3.6. Proportion of patients judged to have had a “good outcome” with treatment.

Study	How Outcome was Assessed	End of Treatment	Follow up
Czarnecki et al 2012 ⁷⁵	Patient rated scale	69%	60% 12 months
Demartini et al 2014 ⁷¹	Patient rated 5 point Likert scale	72.2%	66.6% 12 months
Moene et al 2002 ⁸³	VRMC by blind video rating	65%	83.7% 6 months
Nielsen et al 2015 ¹	Patient rated 5 point Likert scale	64%	55% 3 months
Dalocchio et al 2010 ¹¹⁵	PMDRS by blind video rating	62%	-
Heruti et al 2002 ¹¹³	Subjective clinician rating of partial or full recovery	55%	-

Key: VRMC=Video Rating Scale for Motor Conversion; PMDRS=Psychogenic Movement Disorders Rating Scale.

In the randomised controlled study of 3-weeks inpatient treatment versus waiting list control (Jordbru et al 2014),⁷³ there was a statistically significant improvement in the assessments of support required for activities of daily living (FIM) and physical function

(physical domains of the SF12) in the intervention group.⁷³ The question remains as to whether these were clinically important changes. The minimum clinically important difference (MCID) for the SF12 or SF36 has not been well established. A number of studies have reported values but these tend to vary considerably across populations.¹⁴² The change reported by Jordbru et al in the SF12 physical domain (11.7, CI 7.2-16.1) was considerably greater than the MCID calculated in a study of total knee replacement surgery (4.3, CI 3.8-4.8).¹⁴³ The MCID for the FIM has only been studied in a stroke population, this value was 22 FIM points.¹⁴⁴ The difference with treatment in Jordbru et al (2014)⁷³ was considerably less than this MCID at 8.4.

A number of prospective studies reported follow-up after the end of treatment; time scales ranged from 3 months¹ to 2 years.⁷⁵ In general on-going benefit from the treatment was reported. Jordbru et al (2014) found that treatment effects were maintained at 1-year follow up, though the cross over design of this study does not allow comparison with a control group. Some studies reported a reduction in gains made with treatment over time,^{1,71,75} but in general the majority of patients sustained at least some longer-term benefit. One study reported further improvement at follow up,⁸³ this being an MDT inpatient treatment delivered over 12 weeks. Saifee et al (2012) reported 7 year follow up from a 4 week inpatient multidisciplinary rehabilitation and found that 58% of patients reported continued benefit compared to their recall of symptom severity prior to admission.⁷⁴

Three studies included assessments of participants' mental health before and after treatment.^{1,71,73} Demartini et al (2014)⁷¹, evaluating a multidisciplinary treatment including psychiatry and cognitive behavioural therapy, reported significant improvements in the HADS and HoNOS. However, the physically based interventions studied by Jordbru et al (2014)⁷³ and Nielsen et al (2015)¹ reported no significant change in the mental health domains of the SF12 or SF36 respectively nor change in the HADS. These differences may reflect the absence of psychological therapy or other factors such as differences in mental health at baseline.

Only one study has considered the cost effectiveness of rehabilitation. Nielsen et al (2015)¹ converted the EQ-5D-5L change scores into quality adjusted life years (QALY)

and found a gain of 0.1 over 3 months¹. If this gain was maintained at 12 months, the comparatively low cost intervention would most likely be considered cost effective by the National Institute of Clinical Excellence. This calculation was considered a precursory estimate of cost effectiveness only.

3.3 Discussion

In this systematic review, 35 studies describing physical rehabilitation for FMD were identified. The overall quality of the evidence is low. There was only one randomised controlled trial and 10 cohort studies with subject numbers greater than 10, of these four were reported retrospectively. The relatively small number of studies seems to be at odds with the reported size of the problem in terms of cost and prognosis, as well as the high level of utilisation of physiotherapy/physical rehabilitation for FMD.^{16,145}

Despite low numbers and methodological limitations, the results from treatment reported in the literature are encouraging. There is controlled evidence from one study that physical based rehabilitation can provide at least short term improvement for patients with functional gait disorder without significant psychopathology and a symptom duration of less than 5 years.⁷³ There is also uncontrolled evidence from 6 relatively large (n>10) cohort studies that treatment for mixed FMD involving physical rehabilitation is beneficial for the majority (55-72%) of patients.^{1,71,75,83,113,115} This is supported by positive results from an additional 28 smaller cohort and single case studies. The effect size of physical rehabilitation on patient reported measures of quality of life tends to be small to medium.^{1,73} It appears that the majority of patients remain to some extent symptomatic after treatment⁷⁵ and that very few patients with chronic symptoms return to work.⁷⁴ There were no reports of significant adverse effects of physical rehabilitation for FMD, although this may reflect the fact that many studies were set within specialist services with experienced staff.

At least some of the improvements made with physical rehabilitation appear to be maintained at follow up of 1 to 2 years^{71,73,75} and patients have continued to report some benefit at 7 years following multidisciplinary treatment.⁷⁴ The evidence to support sustained improvement at follow up is uncontrolled.

The most appropriate outcome measures to use in physical rehabilitation of FMD remains unknown. The modest effect size seen with treatment may be related to a lack of sensitivity of standardised measures such as the SF36 in this population. Problems may also be encountered with ceiling effects, as was seen with the Modified Rivermead Index.¹¹² Snapshot measures of physical performance and symptom severity, such as the Berg Balance Scale, arguably have validity and reliability issues due to fluctuations in symptom severity inherent within the diagnosis. Clinical global impression scales of perceived change have been used most regularly in the more recent studies. These have the advantage of being valid, quick, free to use and sensitive to change. However these scales are unable to quantify the impact of treatment, for example change to disability, and are vulnerable to confounding factors. There are no formal studies of the cost effectiveness of rehabilitation for FMD, but there is some suggestion that this may be favourable.¹

The current evidence for the benefit of physical rehabilitation may not be generalisable to the FMD population as a whole. Cohorts tend to be highly selected and patients with non-acceptance of the diagnosis, significant psychopathology, pain, fatigue and other comorbidities (which are commonly seen in this population) are often excluded from studies.^{1,73,75,115} It is sometimes stated that certain patients are not suited to physical rehabilitation for these reasons^{1,75} and that it is difficult to determine suitability for rehabilitation.⁷¹ It is also not clear if different symptom presentations respond differently to treatment.

There is insufficient data to recommend one treatment approach over another, however what is done during the treatment does seem to matter. Two studies report improvement with specialised treatment following failed attempts at more generic physical rehabilitation.^{1,75} The interventions described in the literature are multifaceted, making it impossible to determine which are the most important ingredients in a treatment programme. There are however many useful ideas from which to design and empirically test a treatment. Explanation of the diagnosis and education about the disorder is likely to be important, and it forms a common part of the better-designed studies.^{1,73,75} It appears that physical rehabilitation that aims to retrain movement patterns in graded steps is useful and arguably more appropriate

than following treatment protocols for analogous neurological conditions. In addition, it is likely that there are important overarching principles that can support treatment, such as positive reinforcement, goal setting, family involvement and clear communication. The evidence is insufficient to determine the most appropriate treatment parameters in terms of setting, duration and intensity. The dominance of inpatient treatment studies may simply reflect research-resource distribution. Alternatively, there may be therapeutic benefits to inpatient treatment, such as a higher intensity with treatment delivered over consecutive days, as was considered important in some studies,^{1,75} but this requires further evaluation.

It is acknowledged that there are historical accounts of treatment of FMD in the literature that predate database indexing and the emergence of physiotherapy as a defined profession. Researching historical treatment was considered outside the scope of this work, but its omission is a limitation of this literature review. It is also acknowledged that there may be mechanisms of change following physical rehabilitation not discussed in this paper; including placebo, face-saving opportunities to “relinquish” symptoms and the psychological impact of physical rehabilitation.

3.4 Conclusions

This systematic review found 34 cohort studies of physical rehabilitation and 1 randomised (delayed start) controlled trial. This limited evidence base supports the use of physical rehabilitation for FMD. The best evidence is for inpatient rehabilitation for functional gait disorders and treatment within a specialist programme (as opposed to a generic treatment service). In view of the promising results reported in the literature more research is warranted. The many unanswered questions that future research should address include: what are the important therapeutic elements of treatment; what is the likely effect size of treatment; which are the most appropriate measures to assess outcome; who is most suitable for physically based treatment; how might symptom phenomenology and comorbidity affect treatment; what are the optimal treatment parameters in terms of setting, duration and intensity; can symptom relapse be prevented or minimised; and finally what is the economic benefit of physical rehabilitation of patients with FMD.

Chapter 4 Development of the Study Intervention

4.1 Introduction

Physiotherapy is generally considered an important part of treatment for patients with functional motor disorder (FMD).¹⁶ Physiotherapists specialised in neurology report seeing these patients frequently,¹⁴⁵ and physiotherapy forms a key part of specialist multidisciplinary inpatient rehabilitation, which is usually considered the gold standard treatment.^{71,72} High utilisation of physiotherapy for FMD has continued despite limited evidence for effectiveness and an absence of guidelines and formal descriptions of what physiotherapist should actually do to help such patients.¹¹⁰

Lack of progress in the development of physiotherapy treatment over the years may in part be explained by the uncertain territory occupied by FMD. It is diagnosed by neurologists, but treatment has been considered the responsibility of psychiatrists and psychologists. This is due to the dominance of aetiological theories that consider FMD as a manifestation of psychopathology triggered by adverse life events. In this situation the role of physiotherapy is unclear, there are no obvious symptom-mechanisms to address and physiotherapy may be considered as a placebo treatment or a “face saving” reason for recovery, allowing the stigma of mental ill-health to be side-stepped.¹¹¹ Additionally, in the past, some have suggested that physiotherapy treatment could be harmful when physical symptoms are a manifestation of mental illness.¹⁴⁶ Arguably these factors have steered the physical rehabilitation community away from interest in and ownership of patients with FMD.

It is increasingly recognised that pure psychological explanations for FMD do not apply to a sizeable proportion of patients with FMD and that broader biopsychosocial aetiological models are more relevant.²⁸ This change in perspective is reflected in the most recent version of the Diagnostic Statistical Manual for Mental Disorders (DSM-5),⁸ where the requirement for the presence of a psychological stressor preceding symptom onset has been downgraded from an essential to a supportive criterion. However, progress towards a unified biopsychosocial understanding of FMD has been limited by the “black-box” of the biological sphere, which has only recently become a

topic of interest in the scientific literature. The past 10 years has seen important developments in understanding how symptoms are produced and experienced as involuntary.³⁸ These “neurobiological” mechanisms provide a rationale for physiotherapy treatment, paving the way for a specific physically-based intervention targeting symptom mechanisms.

4.2 Developing and Evaluating a Complex Intervention

The MRC guidelines for developing and evaluating complex interventions informed the development of the physiotherapy intervention in this study.² A complex intervention in this context is one where there are several interacting components. For example, and in relation to physiotherapy for FMD, the intervention depends on behaviours required by those delivering and receiving the intervention; the intervention has several targets (i.e. biological, psychological and social domains); the intervention requires flexibility in how it is administered; and the intervention may have a variety of different outcomes measured across different domains (e.g. physical disability, social functioning and mental health). The MRC state that development of a complex intervention should start with identifying an appropriate theory that underpins the treatment approach.

4.3 A Theoretical Aetiological Model for Functional Motor Disorder

The theoretical underpinning of the study intervention is described in this section. The theory is based on a novel neurobiological explanatory model for FMD that has recently been described and supported by clinical evidence and laboratory experiments.^{38,39} The model emphasises the role of attention and expectation as the key mechanisms that *drive* functional symptoms. These “neurobiological” mechanisms are considered alongside other evidence-based aetiological factors, including physical precipitating events, psychological factors, social factors, neuroplasticity, pain and fatigue, to produce a physically based aetiological model for FMD that is amenable to physiotherapy treatment.

4.3.1 The Role of Attention in Functional Motor Symptoms

FMDs require attention in order to manifest. When the patient's attention is directed away from their abnormal movement, the symptom resolves or reduces. This is the basis for many clinical signs used to make a positive diagnosis of FMD.¹¹ For example, distractibility is a positive sign used to diagnose functional tremor and can be demonstrated by finger tapping tasks in the contralateral limb,¹⁴⁷ see Figure 4.1. The tapping frequency is directed by the clinician, the patient is required to copy this and in order to do so they must shift their attention away from their tremor and focus on the tapping task. This shift in attention leads to resolution of the tremor or entrainment of the tremor to the frequency of the tapping task. If the patient is unable to shift attention from the tremor they will be unable to perform the tapping task (poor task performance). Tremor entrainment, tremor resolution and poor task performance are positive signs for functional tremor.¹¹

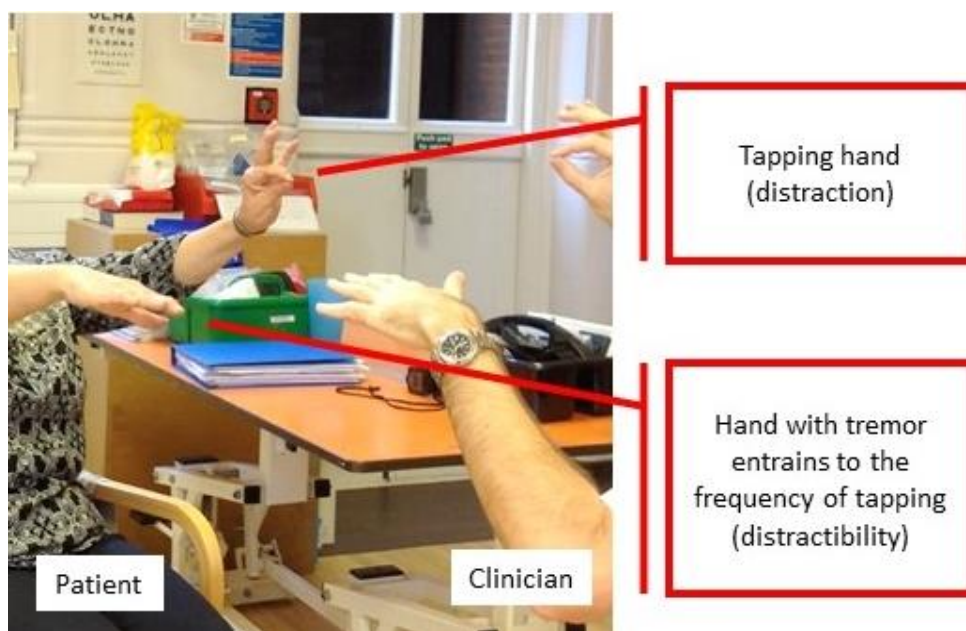


Figure 4.1 Tremor distraction and entrainment

The role of attention has been explored in laboratory experiments. One experimental paradigm showed that when movement is highly predictable, patients with functional tremor perform worse than normal controls. Conversely when movement is

unpredictable, patients perform normally.¹⁴⁸ The implication is that when movement is predictable, there is opportunity for attention to be focused on movement, which disrupts the movement. Conversely, when movement is unpredictable, there is no opportunity for self-focused attention to disrupt movement.

A potential clinical implication for physiotherapy for FMD is that unpredictable movements may be useful to elicit symptom free movement.

The disruptive effect of self-focused attention on movement is a normal phenomenon. It has been described and studied in sports science, colloquially termed “choking under pressure” in competitive situations. It is thought that the pressure to do well heightens self-focus (self-consciousness), resulting in conscious attention to the step-by-step process of movement, with the end result of impaired task performance.¹⁴⁹ Research aimed at improving the resistance of sports performance to such ‘choking under pressure’ has provided encouraging results for promoting implicit learning over explicit learning.¹⁵⁰ Implicit learning refers to learning by exposure or repetition and does not necessarily involve awareness of how the task is performed. In contrast, explicit learning refers to accumulation of knowledge or rules of how to perform a task, which can be verbally expressed. Implicit learning may be more appropriate in the rehabilitation of people with FMD, as it requires less awareness of (and attention towards) the processes of movement. Implicit motor learning paradigms have been applied to neurorehabilitation in subjects with Parkinson’s disease and stroke.^{151,152} Though they should be interpreted with caution due to small numbers (n=27 and 22 respectively), these studies suggest that skills acquired through implicit learning are more resistant to interference from dual tasking than explicit learning paradigms. Paradigms for implicit learning attempt to create learning without errors in order to reduce the accumulation of explicit rules of how to perform the task, whereas explicit learning paradigms encourage trial and error learning and the generation of rules for performing the task.

In relation to the rehabilitation of FMD, an implicit learning approach could limit errorful movement, avoid accumulation of verbal rules and discourage awareness of and attention to step-by-step components of a movement.

4.3.2 *The Role of Expectation and Belief in Functional Motor Symptoms*

Functional motor symptoms are associated with an expectation or belief that movement will be abnormal. Perhaps the clearest illustration of the role of expectation in FMD is the potential for curative responses to placebo treatment.¹¹ For example, botulinum toxin takes at least 72 hours to exert a neuromuscular blocking action, but when administered to patients with fixed functional dystonia together with the suggestion that the effect will be immediate, *some* patients have a dramatic instant (placebo) response.⁴⁰ Here expectation can be considered a mechanism driving the symptom and symptom-resolution.

Theoretical models of how the brain functions in a predictive manner have been used to explain the role of expectation in driving FMD.³⁸ In the model, expectation represents more than just a consciously reportable thought. Expectation also represents neurally encoded predictions of how the body will interact with the environment, based on an internal model of the world. It is proposed that, in a hierarchical brain system, expectations are conveyed via top down processing to influence sensory perception and motor output at a preconscious level.³⁸ An example of how an expectation can cause aberrant movement is the experience of lifting an object that is expected to be heavy, but in actuality is very light. The lifting action of the upper limb and the anticipatory postural adjustments are based on an expectation that is wrong (i.e. that the object is heavy), the selected motor actions are therefore inappropriate and overshoot what is necessary. The movement goes wrong in a way that feels involuntary and outside the control of the individual.

Belief and expectation appear to be important in the prognosis of patients with FMD. In a study of 716 neurology outpatients whose symptoms were “unexplained by disease”, poor outcome at one year (based on a patient reported clinical global impression scale) was predicted by patients’ expectation of non-recovery and patients’ belief that their symptoms were not attributable to psychological factors.¹⁵³

In a study comparing 107 patients with functional weakness (FMD) to 46 patients with organic neurological causes for weakness, many of those with FMD reported ongoing

beliefs about the presence of an underlying disease.³ The subjects with FMD were more likely to endorse the statements, “my illness is a mystery” and “my illness is permanent”. The authors note that it is hardly surprising that patients with FMD were mystified, given that many doctors also report poor understanding of the diagnosis. This mystery however may be implicated in the detrimental beliefs and expectations that form part of the symptom mechanism.

A clinical implication for FMD is that interventions that seek to help the patient understand their symptoms and positively influence their beliefs about the potential for recovery and the influence of psychological factors may improve prognosis.

Expectations of abnormal movement can be directly addressed in physiotherapy by demonstrating to the patient that their movement can be normal, by using distraction and clinical signs that demonstrate symptom reversibility.¹⁵⁴ Presenting the patient with evidence of normal movement may reduce the certainty with which they hold problematic illness beliefs and expectations. This demonstration may take the form of experiencing normal symptom-free movement during therapy sessions and watching their movement in a mirror or on video. Unhelpful expectations and beliefs may also be addressed by education.

Physiotherapy may address detrimental illness beliefs by demonstrating to the patient that their movement can be normal and through education on the role of attention and expectation in driving FMD.

4.3.3 Symptom Precipitating Events, Attention and Expectation

Functional motor disorders are commonly preceded by physical events that appear to trigger the onset of symptoms.^{29,30} Typical events include minor injury, surgical procedures, adverse reactions to medication, or the somatic consequences of panic (i.e. tremor, palpitations, hyperventilation, etc.).

Triggering events, together with self-focussed attention and expectation can be formulated together into a simplified aetiological model for FMD.³⁸ The triggering

event, for example a fall from a height, leads to an erroneous illness belief, such as, “I can’t move my leg, I must have a spinal injury”, and an expectation of paralysis or abnormal movement. The leg becomes the source of focussed attention, which interferes with the execution of normal movement. Somatic sensations are amplified under the attentional focus and interpreted as symptoms of disease or injury. Beliefs may be reinforced by social interactions, such as admission to hospital and being subjected to multiple medical investigations. Certain personality traits, such as health anxiety and obsessionality, may reinforce this process and make an individual more vulnerable to developing FMD.

In summary, the precipitating factor leads to high levels of attention directed towards the body and an expectation of abnormal movement, resulting in *involuntary* abnormal movement.

4.3.4 Psychological Factors

The above explanation for FMD plays down the role of psychological factors. They are clearly relevant, in that patients with FMD tend to report higher rates of anxiety and depression than the general population and neurological disease controls.^{6,27} In addition, panic attacks are common at the initial onset of FMD, particularly for those with rapid progression of their condition.¹⁵⁵ However, the evidence on adverse life events is inconclusive; a proportion of individuals exposed to early life stressors do not develop FMD and not all patients with FMD have a history of adverse life events.²⁷ This supports the view that psychological factors are not on their own explanatory, but important risk factors and that a biopsychosocial framework is necessary to understand FMD.

4.3.5 Social Factors

Social and societal factors are a recognised component of the illness experience in all health conditions. The “sick role” is described in sociology theory as a medically sanctioned societal role where illness exempts a person from his or her normal social responsibilities for a period of time. Sociologist Talcott Parsons described how the sick

role comes with potential secondary gains, and therefore patients can be unconsciously motivated to secure this role.¹⁵⁶ The importance of acquiring access to the sick role may be behind the commonly reported need of patients with FMD for a legitimising diagnostic label.¹⁵⁶ Legitimation may not be found in diagnostic explanations that emphasise psychological factors or that are perceived to trivialise symptoms. Arguably, accessing the sick role may be a necessary, transient first step in rehabilitation and recovery. If this is the case, patients who feel they have been denied a legitimate diagnosis may be denied access to the sick role and therefore the path to recovery.

A potential clinical implication is that the patient with FMD may need a diagnosis that legitimises their illness experience in order to engage with rehabilitation. Stone and Edwards (2012)¹⁵⁴ describe a method of delivering the diagnosis of FMD to patients, where it is emphasised that “symptoms are genuine and not ‘made up’ or ‘crazy’.”

4.3.6 Habitual Movement, Neuroplasticity and Secondary Changes

Functional motor symptoms are associated with chronic long term disability and worsening symptoms over time.⁷⁰ Chronic disability and deteriorating symptoms can, at least to some extent, be explained by abnormal movement patterns becoming habitual. Symptomatic habitual movement may be associated with neuroplastic changes in the central nervous system. Neuroimaging and neurophysiological studies have suggested that FMD is associated with abnormal neural circuitry, changes in cortical thickness, and reduced volumes of specific structures, although these findings are based on studies with small numbers and should be interpreted with caution.¹⁵⁷ Additionally, it is possible that some of these findings may be pre-existing conditions that make individuals more prone to developing FMD.

In addition to the role of problematic self-focused attention, abnormal movement may be an adaptive behaviour that serves some purpose, for example reducing the weight through a painful ankle or “unloading” (taking weight off) a leg that is at risk (or perceived risk) of giving-way and causing a fall. The end result is an altered gait

pattern. Illness behaviours may also be important for demonstrating distress to others, for example with analgic movement patterns.

Maladaptive behaviours may occur secondary to FMD and contribute to chronic disability. Examples include sedentary behaviour due to functional weakness (e.g. wheelchair use), leading to physical deconditioning; avoidant behaviour leading to sensitisation of pain; avoidant behaviour leading to heightened anxiety on exposure; and boom-bust activity patterns exacerbating chronic pain and fatigue.

4.3.7 Chronic Pain and Fatigue

Chronic pain and fatigue are common in patients with FMD. In our previous cohort study of 47 patients with FMD selected for physiotherapy treatment, 77% reported persistent pain and 60% regular fatigue.¹ Pain and fatigue may act as risk factors for developing FMD, or as symptom precipitating factors and/or symptom maintaining factors. Chronic pain and fatigue is sometimes considered part of the same syndrome as FMD.¹⁵⁵ Whichever perspective one takes, when present, management of pain and fatigue is probably a necessary part of treatment of FMD.

4.3.8 Physical Biased Symptom Explanatory Model

Bringing these factors together we can produce a symptom explanatory aetiological model for FMD that highlights mechanisms and issues that can be addressed in physiotherapy treatment.¹ This physically based model is presented in Figure 4.2 below. It is acknowledged that this model is a simplistic and incomplete representation of FMD, the role of psychological factors and other comorbidity may be underrepresented and it may not be generalisable to every patient.

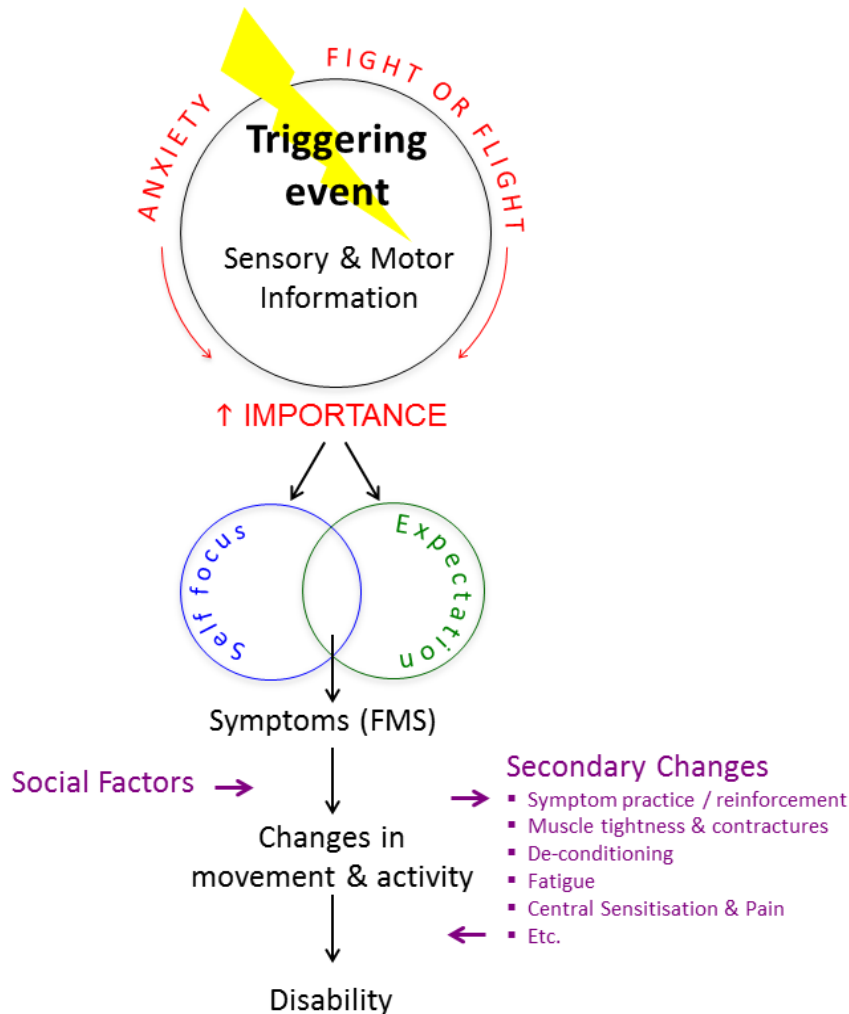


Figure 4.2. Symptom explanatory model

This model depicts that FMD are (commonly) triggered by an event, typically a physical event such as a fall, within the context of a stressful situation that for some patients may result in panic (or a panic attack). Panic may be more easily understood and acceptable to patients when described more biologically as a “fight or flight response”. The precipitating event (e.g. a fall) results in sensory and motor information (e.g. pain with give-way weakness), which due to the context of the triggering event is interpreted as a significant threat. This leads to increased attention directed towards the body (self-focus) and an expectation of abnormal movement or an illness belief (e.g. the expectation is “I am unable to walk”, the illness belief is “I have damaged my spine”). This illness belief is reinforced by the “evidence” of altered movement. Self-

focused attention and expectation drive functional motor symptoms and lead to secondary changes which in turn result in increasing disability. The model also acknowledges the role of social factors, which might include employment issues, family dynamics and societal pressures.

4.3.9 Rationale for Physiotherapy

Physiotherapy can address the mechanisms and other specific issues highlighted in this symptom model. Movement can be retrained by redirecting the focus of motor attention away from the body. Expectation and illness belief can be addressed through education, by demonstrating to the patient that they are able to move normally and teaching them how to initiate normal movement. Addressing secondary changes such as physical deconditioning, fatigue and central sensitisation of pain are accepted and well described roles of physiotherapy.^{158,159} Physiotherapy may also address social factors. This may be directly, for example as part of vocational rehabilitation, or teaching family members how to support rehabilitation, or indirectly via other rehabilitation goals.

This intervention treatment model can be used to help explain the diagnosis of FMD and the rationale of physiotherapy treatment to patients. This was described in previously published work, with a cohort of 47 patients.¹ The model was used as a framework to formulate an individualised explanation to account for how each person had developed FMD.

In summary, a rationale for physiotherapy is that FMD can be conceived of as learnt patterns of movement that are outside the patient's control. Physiotherapy seeks to retrain movement with redirection of the focus of motor attention and to change unhelpful illness beliefs.

4.4 The Physiotherapy Treatment Protocol

The theoretical aetiological model for FMD described above was used to inform the design of the 5-day specialist physiotherapy treatment protocol. The protocol was influenced by my previous clinical experience in the rehabilitation of patients with

FMD, as well as experience in neurorehabilitation and chronic pain management. The overall approach of education and activity is based on established practices of physiotherapy for chronic back pain.^{160,161}

The treatment protocol can be broken down into four main components: Assessment, Education, Movement Retraining, and Self-Management. The treatment protocol is presented in Appendix 2 (page 218) , and has been described in recent publications.^{1,162}

4.5 Testing the Intervention: A Prospective Cohort Study

The 5-day physiotherapy intervention was previously tested in a prospective cohort of 47 consecutive patients with FMD, during the period from August 2012 to March 2014 at the National Hospital for Neurology and Neurosurgery.¹ Participants were admitted to the day hospital for five consecutive days and received eight physiotherapy sessions lasting up to 90 minutes each. Prior to treatment, the patient had attended a consultation with the neurologist, where the diagnosis was made and explained in detail to them, according to a structured format.¹⁵⁴ All patients were followed up at three months.

Immediately after treatment, 96% of patients rated their symptoms as improved on a 7-point Likert scale, this reduced to 85% at 3 months. We defined a good outcome as a self-rating of “very much improved” or “much improved” (points 7 and 6 on the scale); 64% had a good outcome after treatment and 55% at three months. This corresponded to a significant improvement with moderate effect size in more objective outcomes at three months. These included the SF36-Physical Function, SF36-Physical Role, Berg Balance Scale, 10 metre timed walk, and Work and Social Adjustment Scale. Measures that were not significantly different at follow-up were the remaining 6 domains of the SF36 and the Hospital Anxiety and Depression Scale. Quality adjusted life years (QALY) were calculated from the EQ-5D-5L, a generic health questionnaire commonly used for this purpose. A projected mean QALY gain of 0.1 per patient at 1 year was calculated for an approximate cost of £2000 per patient. The resulting incremental cost

effectiveness ratio of £20,000 per QALY suggests a high probability that the intervention is cost effective.

4.6 The next stage in the MRC Guidelines for Developing Complex Interventions

In this chapter I have presented a theoretical basis for the use of physiotherapy for FMD. The theory is derived from empirical clinical and laboratory evidence on the nature of functional motor symptoms, focusing on the importance of attention and expectation as mechanisms driving FMD. A specific physiotherapy treatment approach is described which is designed to address these theoretical symptom mechanisms. The treatment has been tested in a cohort of 47 patients with FMD. The results were promising, providing preliminary evidence for efficacy. The next stage of research, as described in the MRC guidelines was to gather information and evidence of feasibility, in order to inform the design of a pragmatic randomised controlled trial. This is the topic of the following chapters.

Chapter 5 Feasibility Study Methods

The work presented in Chapters 5 to 7 has been published in the form of a research paper: Nielsen G, Buszewicz M, Stevenson F, Hunter R, Holt K, Dudzic M, Ricciardi L, Marsden J, Joyce E, Edwards MJ. Randomised Feasibility Study of Physiotherapy for Patients with Functional Motor Symptoms. J Neurol Neurosurg Psychiatry. Published Online First: 30 September 2016. doi: 10.1136/jnnp-2016-314408

5.1 Introduction

It has been established that FMD is a common cause of disability and distress, with limited evidence for the few treatment options that are available. Physiotherapy has shown promise as a potentially effective treatment, although controlled evidence is lacking. A randomised feasibility study was conducted to determine the feasibility of conducting a pragmatic RCT of specialist physiotherapy for FMD and to obtain the necessary data to inform the design of such a trial.

In this chapter, I report the methods of: **A randomised controlled feasibility study of specialist physiotherapy for functional motor disorder.**

5.2 Study Design and Setting

The study design was a two parallel arm, randomised feasibility study of a specialist physiotherapy-led intervention conducted over five consecutive days versus a treatment-as-usual control, for patients with FMD. This study took place at the National Hospital for Neurology and Neurosurgery, London, UK (NHNN).

5.3 Ethics Approval

Approval was obtained from the National Research Ethics Service Committee London – City Road & Hampstead (14/LO/0572). A trial steering committee oversaw the conduct of the trial. The trial was registered at ClinicalTrials.gov (NCT02275000).

5.4 Participants

Sixty participants were recruited from new patients attending an outpatient neurology clinic specialising in movement disorders and FMD.

Inclusion criteria were:

1. A clinically established diagnosis of FMD according to Fahn-Williams criteria⁴⁵
2. Age 18 years or older
3. Completed diagnostic investigations
4. Acceptance of the diagnosis on the balance of probability (i.e., we did not exclude patients who continued to express some doubt over the diagnosis).
5. FMD duration of at least six months
6. Symptoms severe enough to cause distress (subjectively described by the patient) or impairment in social or occupational functioning

Exclusion criteria were:

1. Unable to understand English
2. Pain or fatigue that were judged to be the primary cause of the patient's disability
3. Prominent dissociative seizures which the patient required assistance to manage
4. Clinically evident anxiety or depression that we felt required assessment before starting physiotherapy treatment
5. High level of disability that prevented participation in an outpatient/day hospital environment
6. Unable to attend five consecutive days of treatment

Prior to enrolment, all participants attended a consultation with the study neurologist (Mark Edwards) where the diagnosis of FMD was made. Each patient received a standard comprehensive explanation of the diagnosis. The components of the diagnostic explanation have been described in detail elsewhere.¹⁵⁴ In brief, this involved showing patients their positive signs of FMD and emphasising the role of self directed attention in driving symptoms. The patient was also referred to online

sources of information (www.neurosymptoms.org and www.FNDHope.org). Patients meeting the selection criteria were provided with written information about the trial (see Appendix 3: Patient Information Sheet, page 227) and invited to return for consent and baseline assessment. All participants gave written informed consent (See Appendix 4: Consent Form, page 230), which was obtained by the study lead physiotherapist Glenn Nielsen or independent research physiotherapist Magdalena Dudzic. Baseline assessment was conducted within 4 weeks of the initial neurology consultation.

5.5 Randomisation and masking

Eligible consenting participants were randomly allocated with a 1:1 ratio to the intervention or control group using a secure online randomisation application (Sealed Envelope, London, UK). The randomisation procedure was completed after baseline assessment by Glenn Nielsen or Magdalena Dudzic. Participants were immediately informed of their treatment allocation. Both participants and clinicians were unmasked to treatment allocation.

5.6 Intervention Group

The intervention was a protocolised physiotherapy-based programme, delivered over five consecutive days by a neurophysiotherapist (Kate Holt), who had undertaken additional specific training (from Glenn Nielsen). An expanded description of the study intervention can be found in Appendix 2 (page 218). Participants were admitted to the day hospital at the NHNN for five consecutive days within four weeks of baseline assessment. As part of the day hospital admission, patients who did not live within a commutable distance of the hospital (defined as outside the M25 motorway) were provided with hotel accommodation.

The first treatment session was a joint consultation with the study neurologist and physiotherapist where information from the initial neurology consultation was reviewed and the aims of the programme discussed. The aims were explained as retraining movement and learning how to manage symptoms in the longer term. The

initial consultation aimed to give an expectation of improvement as a result of taking part in the intervention while being realistic about prognosis. The programme consisted of eight sessions of physiotherapy conducted over five consecutive days, each lasting between 45-90 minutes. Each session included a combination of education, movement retraining, and development of a management plan. Education was centred on a physical biased aetiological model for FMD, see Figure 5.1. The physiotherapist and participant collaboratively devised a formulation to describe how the patient had developed the movement problem. This took into account triggering events, comorbidity, psychological factors (such as panic at onset), self-focussed attention disrupting normal movement, and unhelpful reinforcement of symptomatic movement patterns. Movement retraining aimed to restore normal movement during problematic activities (such as standing from a chair, transferring, walking, and upper limb tasks) by redirecting the focus of motor attention. Specific physiotherapy treatment strategies are listed in Appendix 2 (page 218), and are described in a recent publication.⁴¹ Over the five days, the participant and physiotherapist made notes in a workbook, documenting the individualised symptom formulation, information about FMD, specific symptom management strategies, daily reflections, a personal self-management plan and what to do in case of symptom exacerbation or relapse. Family members were encouraged to attend the initial consultation and some (but not all) of the treatment sessions.

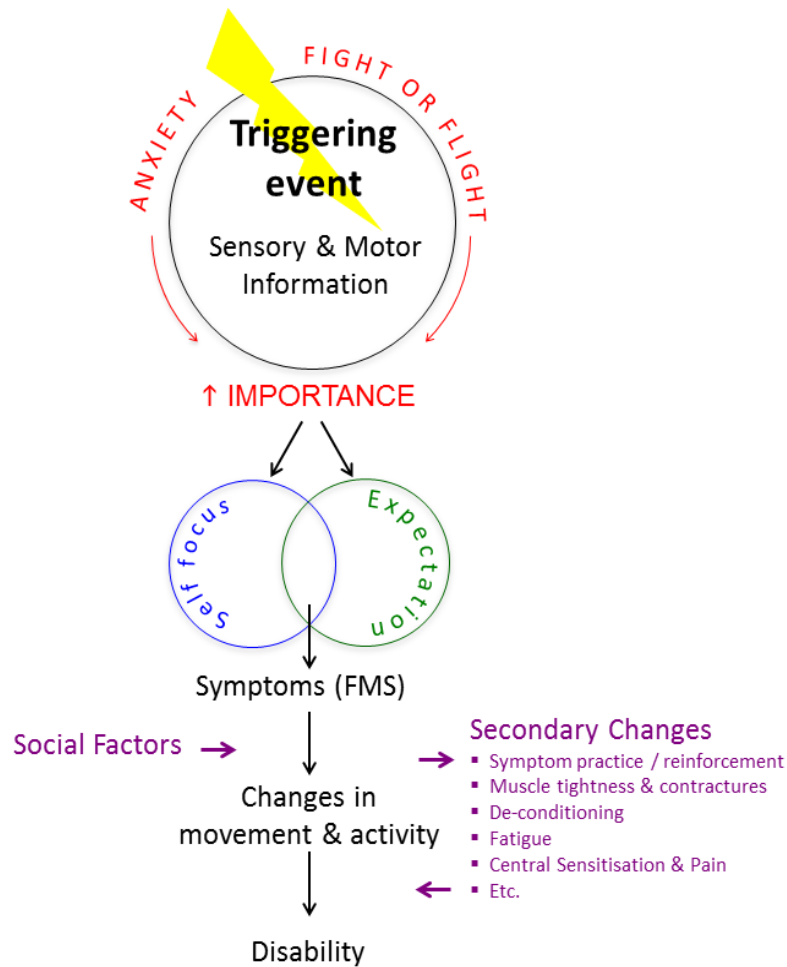


Figure 5.1 Symptom explanatory model used in the study intervention

5.7 Control Group

The control condition was treatment as usual. Following randomisation, a referral was made to the control participant’s local neurophysiotherapy service. The referral letter contained information about the diagnosis, specific treatment goals, and welcomed contact being made by the relevant local team to the specialist centre for further information regarding the diagnosis or treatment advice. No attempt was made to standardise treatment as usual. Input received was recorded, based on patient report.

5.8 Assessment

All participants were assessed at baseline (prior to randomisation), four weeks (at the end of treatment in the intervention arm), and six months. Baseline assessments were completed by the study lead physiotherapist or the independent research physiotherapist. Four week and six months assessments were completed by the independent research physiotherapist only. For the intervention group, the four-week assessment coincided with the final day of treatment. Patients' travel costs associated with assessment were reimbursed.

5.9 Outcome Measures

We collected measures of feasibility and clinical outcome. Measures of feasibility were: recruitment rate, retention, intervention fidelity and acceptability of the intervention. Fidelity of the intervention was assessed by attendance, completion of intervention workbook and participant feedback questionnaire. Acceptability was assessed by participant feedback questionnaire. Safety was assessed by recording participant reported adverse events related to receiving the intervention or control conditions.

We did not specify a primary clinical outcome measure as the primary aim of this study related to feasibility. Clinical outcome measures collected are listed in Table 5-1. These included patient reported questionnaires relating to quality of life, anxiety, depression, and physical function; administered physical assessments; and assessments of the economic impact of symptoms. Participants in the intervention group were also asked to complete a short feedback form to assess acceptability of the intervention.

Table 5.1. Clinical outcome measures. Measures were collected at baseline, four week assessment and six month follow up (unless otherwise stated).

Outcome Measure	Measurment Domain	Description
Patient reported questionnaires		
Short Form 36 (SF36) ¹⁶³	Health-related quality of life 36 items 10 minutes or less	A commonly used generic health-related quality of life measure, producing 8 separate domain scores, a physical and mental health composite score and a total score. It has been demonstrated to be both valid and reliable. ¹⁶⁴ The SF36 was administered at baseline and six month follow up only (as the questionnaire has a four week recall period).
Work and Social Adjustment Scale (WSAS) ¹⁶⁵	Work and social function 5 items 2 minutes or less	A measure of psychosocial function and ability to work. Has acceptable reliability and validity.
Hospital Anxiety and Depression Scale (HADS) ¹⁶⁶	Anxiety and depression 14 items 5 minutes or less	A psychological screening tool that has shown to be a valid and reliable measure of anxiety and depression in and out of the hospital environment. ¹⁶⁶
Brief Illness Perception Questionnaire (B-IPQ) ¹⁶⁷	Illness beliefs 9 items 5 minutes or less	A quantitative measure of illness beliefs, shown to be valid and reliable. Produces 8 dimensions that can be analysed separately or as a total score. ¹⁶⁷
Clinical Global Impression Scale (CGI) ^{77,168}	Patient perception of change 1 item 1 minute or less	Patient rated perception of improvement on a 5 point Likert scale. It has been used in previous treatment studies of FMD. ^{77,80} Administered at 4 week assessment and 6 month follow up only.
Disabilities of the Arm Shoulder and Hand (DASH) ¹⁶⁹	Upper limb disability 30 items 5 minutes or less	A measure of upper limb function that is not disease specific. It has good reliability and validity and is responsive to change. ¹⁷⁰ Participants without upper limb symptoms did not complete this assessment.
Functional Mobility Scale ¹⁷¹	Mobility related disability 3 items 1 minute or less	Very brief scale that quantifies functional mobility by determining assistance required when walking 5, 50 and 500 metres. The scale has been shown to have good validity and reliability in children with cerebral palsy. ¹⁷¹ It was used in a previous study of rehabilitation for FMD and therefore

		useful for comparison. ⁷³ Participants whose symptoms did not affect their gait did not complete this assessment.
Administered physical assessments		
10 metre timed walk ¹⁷²	Walking speed 1 activity 5 minutes	A quick and reliable test of gait speed and step length over 10 metres. ¹⁷² Participants whose symptoms did not affect gait or balance did not complete this assessment.
Berg Balance Scale ¹⁷³	Balance 14 activities 10-15 minutes	A widely used measure of balance shown to have good reliability and validity. ¹⁷⁴ Participants whose symptoms did not affect gait or balance did not complete this measure.
The Simplified Functional Movement Disorders Rating Scale (S-FMDRS) ¹⁷⁵	Movement impairment 12 activities 10 minutes or less	A standardised rating scale to assess and score FMD via video. It was developed by the thesis' author (GN), based on an existing scale (Psychogenic Movement Disorders Rating Scale). ⁹⁸ The new scale was developed to address problems with the original scale, which are described in detail elsewhere. ¹⁷⁵ We tested the inter-rater reliability, validity and sensitivity of the S-FMDRS and found the scale performed satisfactorily and was comparable to the original scale. Participants were videoed completing a standardised set of movements and postures. The videos were scored using the S-FMDRS by a blind assessor (neurologist, Lucia Ricciardi).
Health Economic Analysis		
EQ-5D-5L ¹⁷⁶	Health-related quality of life 6 items 5 minutes or less	A simple quality of life measure that is the preferred instrument of the National Institute for Health and Clinical Excellence (NICE) for generating quality adjusted life years (QALYs). ^{176,177}
Client Services Receipt Inventory (CSRI) ¹⁷⁸	Health service utilisation and related costs 10 items 20 minutes	A standardised, yet adaptable assessment of health service utilisation (such as GP or hospital attendance), informal care, lost work time, and social benefits. ¹⁷⁸ The CSRI is commonly used in health economic analysis. A version of the CSRI was developed for the study (see Appendix 5, page 231) and administered at baseline and six months follow up only. This questionnaire is completed by the participant and asks about service receipt retrospectively over the previous three months. The data collected will be used to inform the design of a version of the CSRI suitable for a large trial.

5.10 Statistical Analysis

A power calculation was not performed as the primary aim of this study was to assess feasibility. The sample size of 60 was predetermined and considered sufficient to meet the objectives of collecting data on outcome measure variation, recruitment and retention.

All data were initially entered into a Microsoft Excel spread sheet (version 2013), by Magdalena Dudzic. Data were double entered (by Magdalena Dudzic and Glenn Nielsen) to detect and correct data entry errors. Statistical analysis was conducted using SPSS version 22. Summary statistics were calculated for each clinical outcome measure. For continuous measures, the difference between groups was assessed using a linear regression model, adjusting for the baseline scores of the measure. Treatment effect was calculated using Cohen's *d* to allow comparisons between outcome measures.¹⁷⁹ Incomplete cases due to drop out were excluded from analysis. The Clinical Global Impression scale (CGI) was collapsed into two groups for subgroup analysis: good outcome (ratings of much improved and improved) and poor outcome (ratings of no change, worse or much worse). For the B-IPQ, a total composite score was calculated by combining the sub scores, reversing scores for items 3, 4 and 7.

Health Economist, Rachael Hunter, completed all health economic analyses, using STATA version 12. The EQ-5D-5L utility scores¹⁸⁰ were converted to Quality Adjusted Life Years (QALYs) by calculating the area under the curve adjusting for baseline differences¹⁸¹. Physiotherapist and neurologist salaries and overheads were obtained from the Personal Social Services Research Unit,¹⁸² and multiplied by the average contact time per patient. Other costs were obtained from trial costings documentation. The estimated mean cost per patient of the intervention minus the mean cost of the control neurophysiotherapy was divided by the difference in QALYs gained between groups to calculate the incremental cost-effectiveness ratio (ICER).

Data from the CSRI was collated using software Microsoft Excel (by Glenn Nielsen) and analysed using software STATA version 12 (by Rachael Hunter). Service use was reported with descriptive statistics. Service costs were estimated using data obtained

from the Personal Social Services Research Unit.¹⁸² Costs were categorised as community care costs and acute care costs. Community care costs were services provided in the community; this included seeing the GP, practice nurse, community physiotherapist and community occupational therapist. Acute care costs were services provided within a hospital setting; this included hospital outpatient appointments, day procedures, elective and non-elective admissions and seeing physiotherapists etc. while in hospital. Further statistical analyses were not reported due to the low participant numbers in this feasibility study.

The number of prescribed medications was reported with descriptive statistics. Work days lost through sickness were reported for participants who were in paid employment. Questions with missing data and areas of high service utilisation or high costs were noted to inform a future version of the CSRI.

5.11 Exploration of the Data

In an exploratory analysis, the relationship between the presence of anxiety or depression at baseline and intervention outcome was explored. This was an exercise in hypothesis generation, in recognition that the data was not specifically powered for subgroup analysis. Outcome (Good vs Poor) was based on collapsed CGI scores. Cases of anxiety and depression were determined by a cut off score of 10 and above in the HADS anxiety and depression subscales. This cut off score is thought to exclude false positive cases¹⁶⁶. This data was presented in a 2x2 contingency table and analysed with a Chi-square test.

Chapter 6 Feasibility Study Results

Measures relating to feasibility are presented, followed by clinical outcomes.

6.1 Recruitment

The trial profile and flow of participant through the study is shown in Figure 6.1. Recruitment began 8 September 2014 and was completed by 4 June 2015. During these nine months, 210 patients were screened for inclusion and 143 were excluded. The most common reasons for exclusion were dominant pain (n=57, 27% of screened patients), clinically evident anxiety or depression requiring assessment (n=50, 24% of screened patients) and dominant fatigue (n=22, 10% of screened patients). Seven patients declined to participate and the remaining 60 were recruited and randomly assigned to the intervention (n=30) and control (n=30) groups. The percentage of screened patients meeting the selection criteria was 31.9% (95% CI 25.6, 38.2), 90% of eligible patients consented to participate.

6.2 Retention and Dropouts

The number assessed at the primary endpoint was 29 for the intervention group (1 dropout) and 28 for the control group (2 dropouts). The overall dropout rate was 5% (95% CI 0.0, 10.5%). The participant that dropped out from the intervention group did so prior to commencing treatment. Only one participant gave a reason for dropping out, which was a new, separate health condition that was under investigation. We suspect the other two participants dropped out due to anxiety (which we underestimated on initial assessment) and distance required to travel to the hospital.

One participant from the intervention group was unable to attend the final assessment, they completed questionnaires by post and their final physical assessment measures (Berg Balance Scale, 10 metre walk time and S-FMDRS) were missing from the final analysis. Three participants from the control group did not attend the interim four-week assessment, but did attend the final assessment. The

most common reason for nonattendance was the distance or difficulty with travel to the hospital.

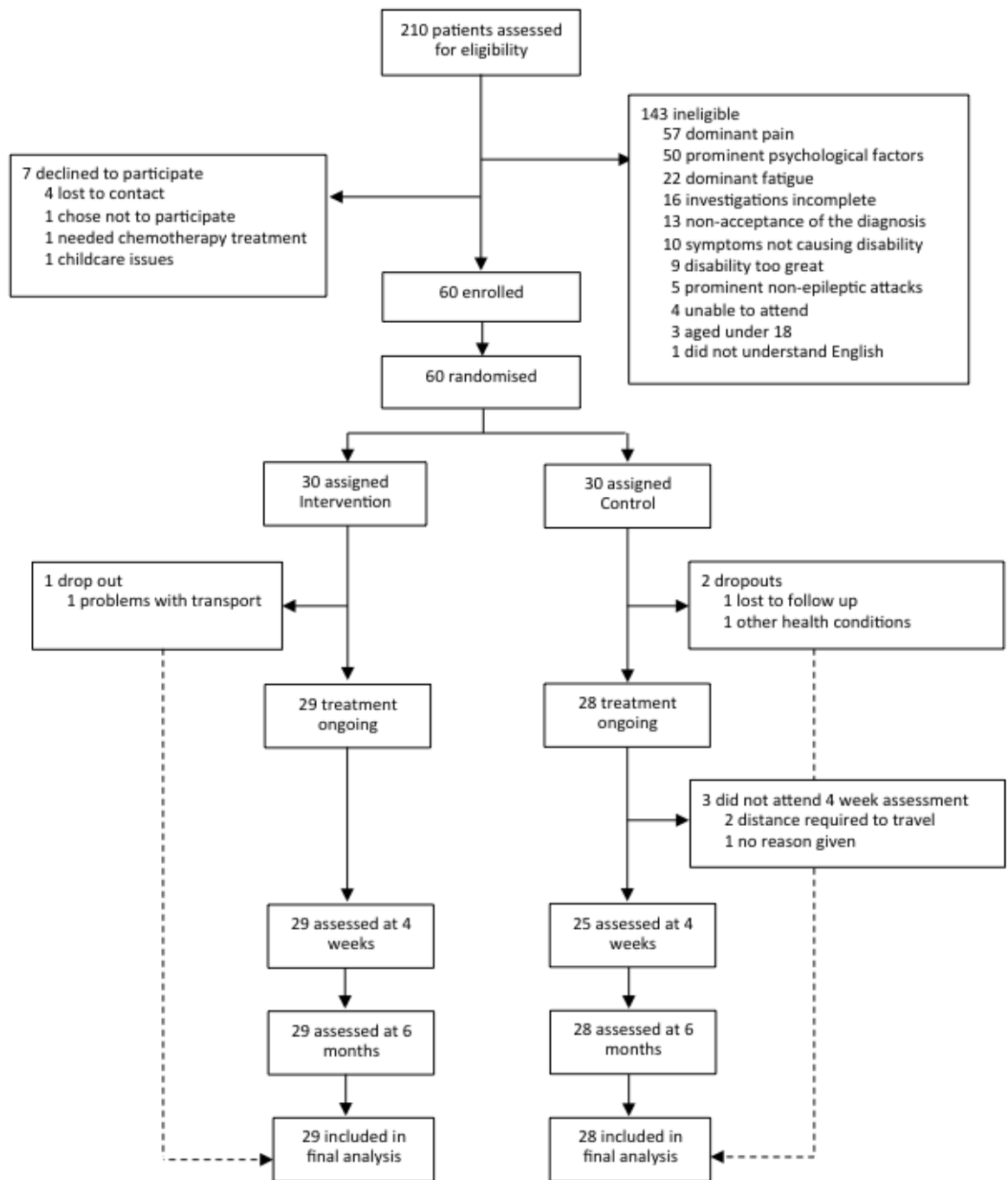


Figure 6.1. Trial Profile

6.3 Fidelity

Participants in the intervention group attended all five days of treatment, except for one participant who missed two days due to sickness. All components of the workbook were covered for each participant and the majority of participants endorsed the following statements in post-treatment feedback (strongly agree on a 5-point Likert scale): The programme helped me understand my symptoms (93%); The programme helped me gain more control over my symptoms (83%); The programme has helped me to create a plan to improve my symptoms (90%); and The programme included information about thoughts, feelings and psychological influences on my symptoms (86%). The participant feedback questionnaire and results are presented in Appendix 6 (page 236).

6.4 Acceptability of the Intervention

In the post-treatment feedback questionnaire, all participants in the intervention group reported they were either completely satisfied (86%) or satisfied (14%) with their treatment and they would be extremely likely (93%) or likely (7%) to recommend the programme to family and friends if they required similar treatment. The intensity of treatment was considered about right (38%) or very intense but manageable (48%). Two participants (7%) found the treatment too intense, and two (7%) felt the treatment was not intense enough. See Appendix 6 (page 236) for feedback questionnaire and results.

6.5 Follow Up Assessment Timeframes

The average time from recruitment into the trial to four-week assessment was 5.4 (SD 2.4) weeks for the intervention group and 5.4 (SD 1.8) weeks for the control group. The average time from recruitment to six-month follow up was 6.5 (SD 0.8) months for the intervention group and 6.4 (SD 0.8) months for the control group.

6.6 Safety

No serious adverse incidents were reported during the study period. Some participants from the intervention group reported exacerbation of chronic pain or fatigue during and the week following the intervention. This resolved without the need for new intervention and was usually attributed to being more physically active around sessions, such as walking to and from the physiotherapy gym, rather than the treatment itself. One participant from the intervention group reported having had a fall in the months after treatment, sustaining a wrist fracture, which required surgery. This participant had reported falling regularly at baseline.

6.7 Treatment Received by Control Group

Self-report data regarding details of physiotherapy treatment was available from 28 of 30 control participants. Only one participant did not see a physiotherapist in the period from baseline to six month follow up. A referral was made but they had not been contacted for an appointment. Three participants opted for private physiotherapy treatment (one had private treatment in addition to NHS treatment) and they received between 6 and 9 sessions (6, 6, 9). Twenty-five participants saw an NHS physiotherapist. The number of sessions ranged from 1 to 17, the median number was 5 (IQR 3 – 7.5).

The content of physiotherapy sessions included gait retraining, stair practice, balance work, general cardiovascular exercise, specific strengthening exercises, and stretching. Four participants were provided with a walking aid or splint. One participant had fatigue management education and one participant was given strategies to practice to control a functional tremor.

Contact from physiotherapists providing treatment as usual (control) physiotherapy for advice was received for 5 participants. In each case a published paper describing recommendations for physiotherapy for patients with FMD⁴¹ was emailed to the physiotherapist.

6.8 Baseline Characteristics of Participants

Baseline demographic and clinical characteristics of all the participants are shown in Table 6.1 and Table 6.2. The mean age was 43 years, 72% were women and 48% were not working due to ill health. The mean symptom duration was 5.8 years (SD 7.3) and the mean age of symptom onset was 37 (SD 12.0). The most common predominant symptom at presentation was a mixed movement disorder (40%), followed by gait disturbance (27%), weakness (12%) and upper limb tremor (10%). The category of mixed movement disorder represented patients with symptoms that included dystonic-type movement, tremor and weakness. Sensory symptoms were common, these included pins and needles (63%), numbness (55%), dizziness (48%) and visual disturbance (38%). Other common comorbid symptoms were general weakness (72%), headache (60%), speech disturbance (48%) and bladder problems (33%). Half the participants rated their daily levels of pain and fatigue as severe to extreme at baseline and only a small minority did not experience regular pain (12%) or fatigue (5%). Forty-eight per cent of participants reported having falls since their symptoms started. Most had previously seen a physiotherapist (77%), and 32% had had psychological therapy. The control and intervention groups were evenly matched for baseline demographic and clinical characteristics.

Table 6.1 Baseline demographic characteristics

Demographic Data	Intervention group n=30	Control group n=30	Combined N=60
Age, mean (SD)	44 (13.1)	41 (13.1)	43 (13.1)
Female Gender	22 (73%)	21 (70%)	43 (72%)
Marital status			
Married/partner	19 (63%)	18 (60%)	37 (62%)
Single	9 (30%)	11 (37%)	20 (33%)
Widowed	0	0	0
Divorced/separated	2 (7%)	1 (3%)	3 (5%)
Employment status			
Full time work	7 (23%)	7 (23%)	14 (23%)
Part time work	6 (20%)	5 (17%)	11 (18%)
Retired / not working	2 (7%)	3 (10%)	5 (8%)
Volunteer work	0	0	0
Current sickness	2 (7%)	0	2 (3%)
Not working due to health	11 (37%)	15 (50%)	26 (43%)
Medically retired	1 (3%)	0	1 (2%)
Student	1 (3%)	0	1 (2%)
Employment status summary			
In paid work or full time study	14 (48%)	12 (40%)	26 (43%)
Not working due to ill health	14 (45%)	15 (50%)	29 (48%)
Other	2 (7%)	3 (10%)	5 (8%)
In receipt of state benefits	20 (67%)	17 (57%)	37 (62%)
Education level			
Less than 16 years	2 (7%)	1 (3%)	3 (5%)
Up to 16 years	8 (27%)	13 (43%)	21 (35%)
Up to 18 years	4 (13%)	6 (20%)	10 (17%)
Degree level qualification	13 (43%)	9 (30%)	22 (37%)
Post graduate qualification	3 (10%)	1 (3%)	4 (7%)
Current litigation	0	0	0

Table 6.2. Baseline clinical characteristics

Clinical data		Intervention group n=30	Control group n=30	Combined n=60
Symptom duration, years, mean (SD)		5.9 (8.3)	5.6 (6.2)	5.8 (7.3)
Age at symptom onset, mean (SD)		38 (12.9)	36 (11.2)	37 (12)
Primary Symptom, frequency:				
	Weakness	3 (10%)	4 (13%)	7 (12%)
	Gait disturbance	7 (23%)	7 (23%)	14 (23%)
	Upper limb tremor	3 (10%)	3 (10%)	6 (10%)
	Lower limb tremor	2 (7%)	0	2 (3%)
	Head tremor	2 (7%)	1 (3%)	3 (5%)
	Fixed dystonia	0	1 (3%)	1 (2%)
	Jerks	2 (7%)	1 (3%)	3 (5%)
	Mixed movement disorder	11 (37%)	13 (43%)	24 (40%)
Sensory Symptoms, frequency				
	Visual disturbance	11 (37%)	12 (40%)	23 (38%)
	Hearing difficulties	8 (27%)	6 (20%)	14 (24%)
	Pins and needles	15 (50%)	23 (77%)	38 (63%)
	Numbness	14 (47%)	20 (67%)	34 (55%)
	Dizziness	15 (50%)	14 (47%)	29 (48%)
Other Symptoms / Complaints:				
	Weakness	20 (67%)	23 (77%)	43 (72%)
	Headache	14 (47%)	22 (73%)	36 (60%)
	Sleep disturbance	18 (62%)	20 (67%)	38 (63%)
	Gastrointestinal complaints	9 (30%)	6 (20%)	15 (25%)
	Bladder problems	9 (31%)	11 (37%)	20 (33%)
	Speech disturbance	13 (43%)	16 (53%)	29 (48%)
	Concentration or attention problems	23 (77%)	22 (73%)	45 (75%)
	Dissociative attacks/seizures	6 (20%)	3 (10%)	9 (15%)
Pain, self-rating:	None	6 (20%)	1 (3%)	7 (12%)
	Slight to moderate	10 (34%)	14 (47%)	24 (40%)
	Severe to extreme	13 (45%)	15 (50%)	28 (47%)
Fatigue, self-rating:	None	2 (7%)	1 (3%)	3 (5%)
	Slight to moderate	13 (43%)	14 (47%)	27 (45%)
	Severe to extreme	15 (50%)	15 (50%)	30 (50%)
Patients who report falling		10 (33%)	19 (63%)	29 (48%)
Previous physiotherapy		23 (79%)	23 (77%)	46 (77%)
Previous psychology		9 (31%)	10 (33%)	19 (32%)
Botulinum toxin injections		6 (20%)	4 (13%)	10 (17%)

6.9 Continuous Clinical Outcomes

Clinical outcomes are presented in Table 6.3, Table 6.4 and Figures 6.2 to 6.10. Inspection of baseline data suggests that the control group had generally worse scores than the intervention group, which were accounted for in the analysis.

The assumptions of the regression model were met. After adjusting for baseline scores, at six months the intervention group had superior scores (representing better health) in three domains of the SF36 (Physical Function, Physical Role, and Social Function, see Table 6.3); the Berg Balance Scale, the 10 metre walk time, the Functional Mobility Scale, the DASH, and the composite B-IPQ score (Table 6.4). Two outliers skewed the results of the 10 metre walk time, inflating the treatment effect. After removing these outliers, the mean difference remained significant. Effect sizes were medium to large, ranging from $d=0.46$ to 0.79, with the greatest effect seen in the Functional Mobility Scale and Simplified-PMDRS. Outcomes that were not significantly different between groups were the remaining five domains of the SF36 (Bodily Pain, General Health, Vitality, Role Emotional, and Mental Health), the HADS anxiety and depression scores, and the WSAS.

We excluded cases with incomplete data due to dropout from the analysis. When data were analysed by carrying forward baseline scores of incomplete cases in a sensitivity analysis, there was overall little difference. However the difference between groups in SF36 General Health and WSAS scores became significant at the 0.05 level, which suggests our approach of excluding missing cases was the more conservative option.

Table 6.3. Short Form 36 outcomes at baseline and six months

SF36 Domains	Intervention Group mean (SD)		Control Group mean (SD)		Regression coefficient for group, baseline as covariate (95% CI)		Cohen's <i>d</i>
	BASELINE	FOLLOW UP	BASELINE	FOLLOW UP			
Physical function	34.8 (23.7)	51.9 (27.2)	23.7 (19.0)	23.2 (21.3)	19.8	(10.2, 29.5)	0.70
Physical Role	31.7 (28.9)	47.0 (30.3)	19.4 (21.7)	26.8 (22.5)	13.0	(0.8, 25.2)	0.46
Bodily Pain	45.6 (33.5)	47.4 (33.1)	32.1 (25.3)	33.9 (27.4)	3.6	(-8.0, 15.3)	0.12
General Health	47.3 (23.9)	54.1 (28.3)	40.7 (23.4)	39.6 (22.6)	9.0	(-0.1, 18.2)	0.34
Vitality	32.3 (21.4)	39.2 (27.3)	26.6 (17.6)	28.3 (20.2)	6.2	(-3.6, 15.9)	0.25
Social Function	39.7 (33.2)	56.9 (30.2)	34.4 (29.8)	37.0 (25.1)	17.1	(5.0, 29.2)	0.58
Role Emotional	70.1 (29.5)	68.7 (34.5)	61.0 (32.6)	62.5 (35.4)	0.1	(-15.1, 15.4)	0.00
Mental Health	65.5 (21.1)	67.9 (23.8)	58.4 (23.8)	59.3 (25.2)	3.4	(-6.4, 13.2)	0.14
Physical Summary score	33.1 (11.1)	38.7 (10.8)	28.7 (7.9)	29.5 (9.2)	5.9	(2.1, 9.7)	0.54
Mental Summary score	45.2 (13.0)	45.9 (13.6)	42.6 (13.3)	43.3 (14.2)	0.9	(-4.9, 6.8)	0.06

Higher scores represent better health. Abbreviation: SF36=Short Form 36. Regression coefficients in bold reached statistical significance at 0.05 level.

Table 6.4. Continuous outcome measures at baseline and six months

	Intervention Group mean (SD)		Control Group mean (SD)		Regression coefficient for group, baseline as covariate (95% CI)		Cohen's <i>d</i>
	BASELINE	FOLLOW UP	BASELINE	FOLLOW UP			
HADS Anxiety	6.5 (3.8)	6.9 (4.8)	7.7 (4.9)	7.9 (5.6)	-0.1	(-2.1, 2.0)	-0.02
HADS Depression	5.4 (4.0)	5.2 (3.9)	8.0 (4.5)	8.4 (5.0)	-1.4	(-3.2, 0.5)	-0.30
WSAS	24.7 (7.9)	20.2 (10.5)	27.6 (7.5)	26.9 (10.2)	-4.2	(-8.4, 0.1)	-0.39
Berg Balance Scale	39.0 (13.8)	47.7 (13.8)	35.7 (13.2)	37.0 (14.7)	8.0	(2.9, 13.1)	0.53
10 metre walk time **	16.8 (10.0)	9.6 (3.8)	24.6 (17.3)	19.0 (10.6)	-6.7	(-10.7, -2.8)	-0.72
Functional Mobility Scale	11.7 (4.1)	14.5 (3.5)	10.0 (3.6)	10.0 (3.9)	3.4	(1.9, 5.0)	0.79
DASH	51.8 (19.6)	39.6 (25.6)	51.2 (15.0)	48.1 (21.4)	-9.1	(-17.4, -0.8)	-0.38
S-FMDRS	17.6 (9.0)	10.6 (9.1)	15.6 (7.8)	16.6 (8.6)	-7.4	(-11.0, -3.8)	-0.79
B-IPQ total score	50.0 (10.8)	39.4 (16.1)	54.6 (10.6)	51.0 (13.0)	-8.0	(-14.4, -1.6)	0.51

** 2 outliers removed from the intervention group (baseline times of 197 and 182 seconds). Removing these outliers decreased the treatment effect by 1.4 seconds. Higher scores represent better health in the Berg Balance and Functional Mobility Scale. Higher scores represent worse health for HADS, Work and Social Adjustment, 10 metre timed walk, DASH and Simplified-PMDRS. Abbreviations: HADS=Hospital Anxiety and Depression Scale; WSAS=Work and Social Adjustment Scale; DASH=Disabilities of Arm Shoulder and Hand. Regression coefficients in bold reached statistical significance at 0.05 level.

Itemised results for the B-IPQ are reported in Appendix 7 (page 240). Figures 6.2 to 6.10 present mean scores and 95% confidence intervals of continuous clinical outcome measures for the intervention group (green line) and control group (blue line), at baseline, four weeks and six months.

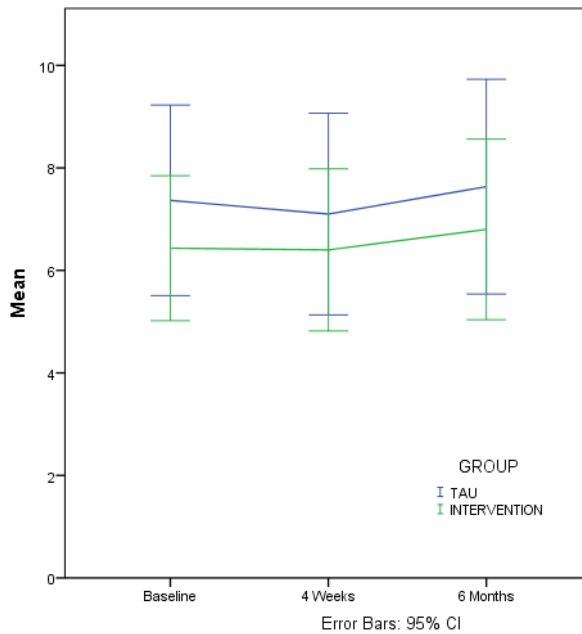


Figure 6.2. Hospital Anxiety and Depression Scale, Anxiety Score. Adjusted difference between groups at six months -0.1 (95% CI -2.1, 2.0).

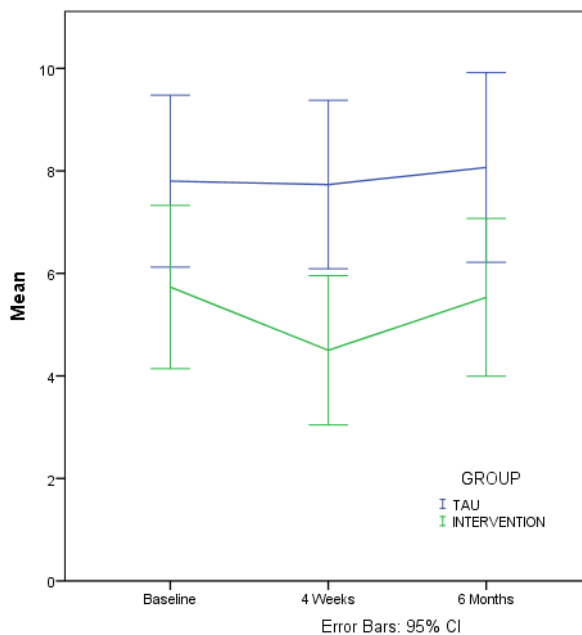


Figure 6.3. Hospital Anxiety and Depression Scale, Depression Scores. Adjusted difference between groups at six months -1.4 (95% CI -3.2, 0.5).

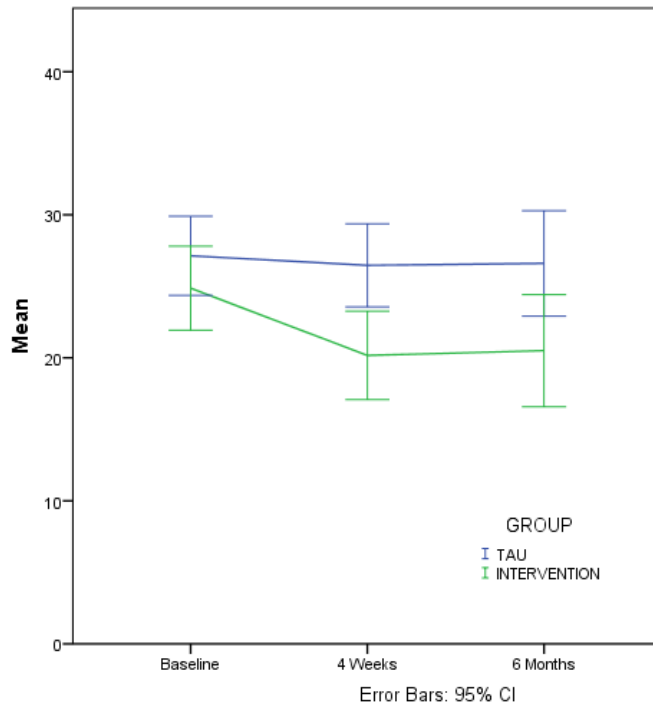


Figure 6.4. Work and Social Adjustment Scale. Adjusted difference between groups at six months -4.2 (95% CI -8.4, 0.1).

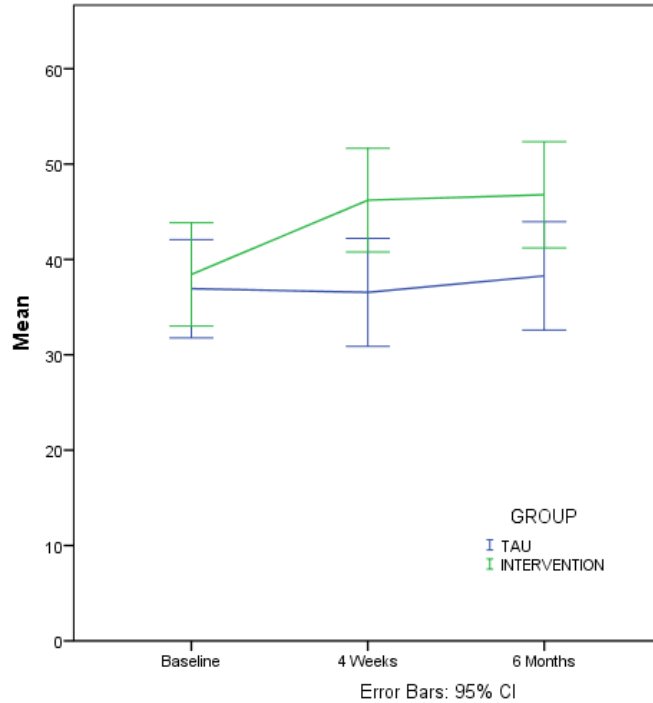


Figure 6.5. Berg Balance Scale. Adjusted difference between groups at six months 8.0 (95% CI 2.9, 13.1), $d=0.53$.

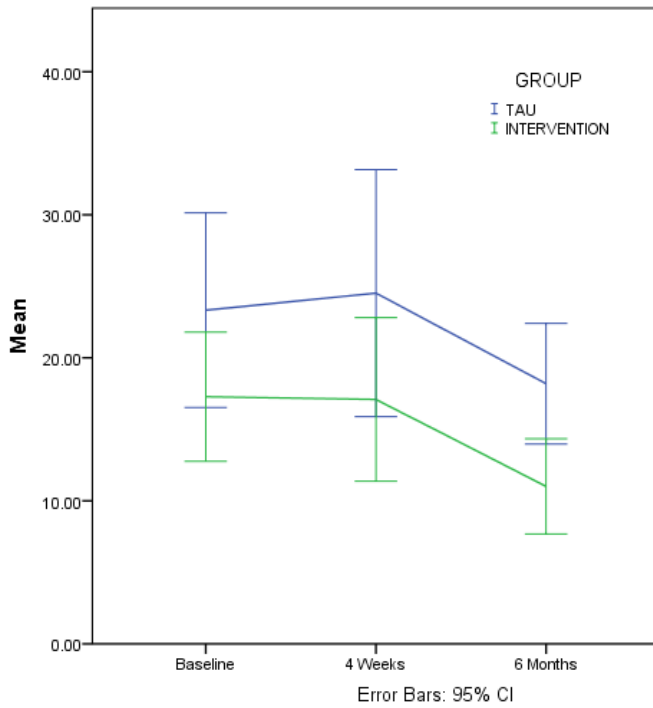


Figure 6.6. 10-Metre Timed Walk. Adjusted difference between groups at six months -6.7 (95% CI -10.7, -2.8), $d=0.72$.

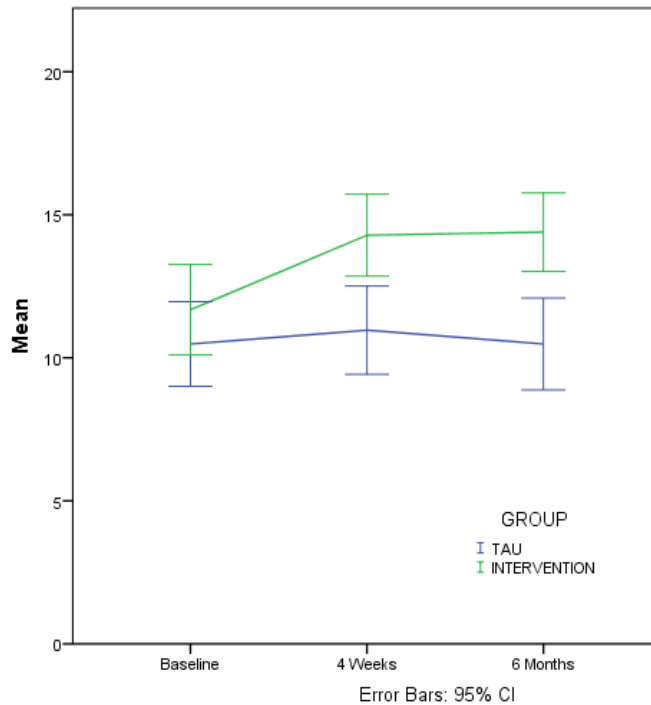


Figure 6.7. Functional Mobility Scale. Adjusted difference between groups at six months 3.4 (95% CI 1.9, 5.0), $d=0.79$.

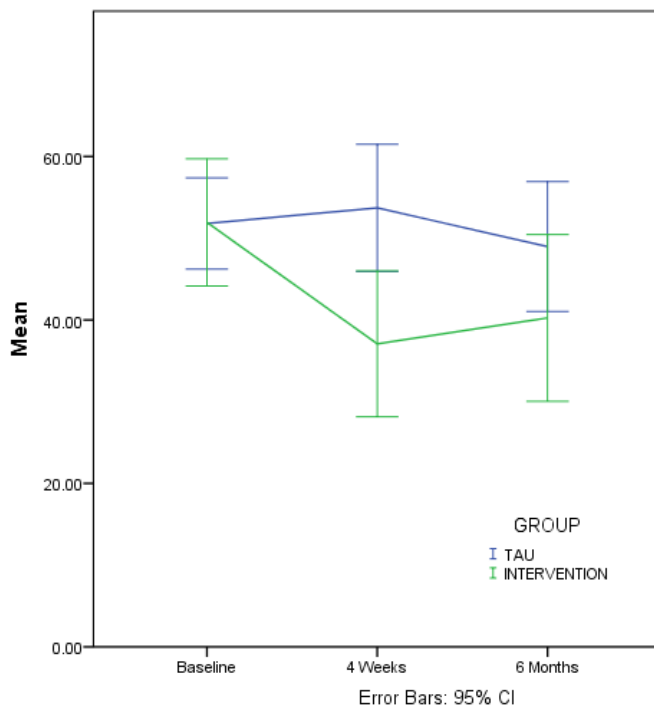


Figure 6.8. Disabilities of the Arm Hand and Shoulder (DASH). Adjusted difference between groups at six months -9.1 (95% CI -17.4, -0.8), $d=0.38$.

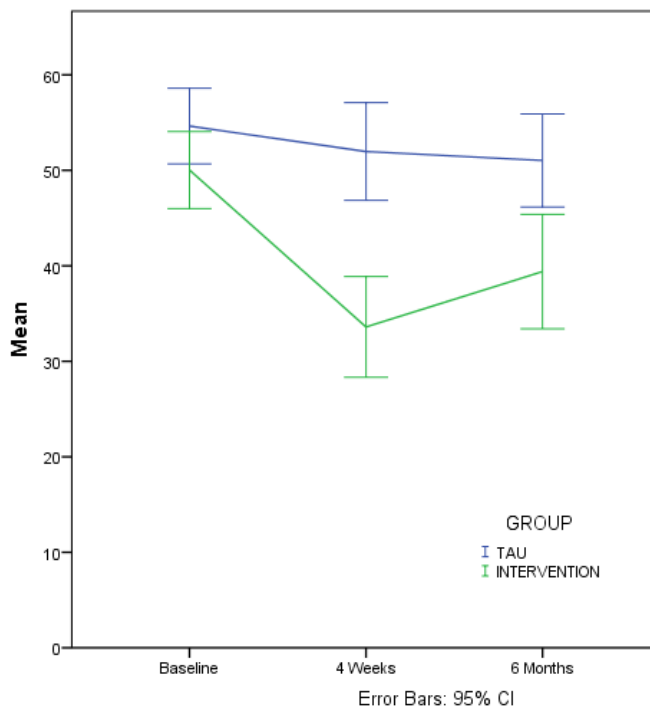


Figure 6.9. Brief Illness Perception Scale, Total Score. Adjusted difference between groups at six months -8.0 (95% CI -14.4, -1.6), $d=0.51$.

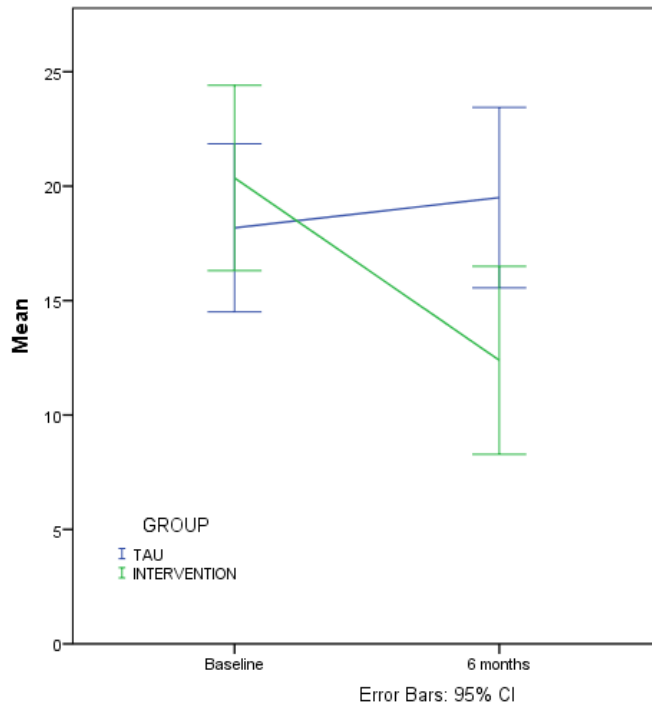


Figure 6.10. Simplified Psychogenic Movement Disorders Rating Scale at baseline and six months follow up. Adjusted difference between groups at six months -8.6 (95% CI $-12.8, -4.4$), $d=0.79$.

6.10 Categorical Clinical Outcomes

The CGI data are presented in Table 6.5. At six months, 72% of the intervention group reported a good outcome, compared to 18% in the control group. Thirty-two per cent of the control group felt that their symptoms had got worse from baseline to six month follow up, compared to 3% in the intervention group. There appeared to be a perceived loss of treatment effect with time, in the intervention group the proportion with a good outcome at four weeks (93%), reduced at six month follow up (72%).

Table 6.5. Clinical Global Impression Scale at four weeks and six months.

CGI Rating	4 Weeks				6 Months			
	Intervention Group		Control Group		Intervention Group		Control Group	
Much worse	0		1	(4%)	0		3	(11%)
Worse	0		7	(28%)	1	(3%)	6	(21%)
No change	2	(7%)	14	(56%)	7	(24%)	14	(50%)
Improved	12	(41%)	2	(8%)	11	(38%)	5	(18%)
Much improved	15	(52%)	1	(4%)	10	(35%)	0	
Collapsed Scores								
Good Outcome	27	(93%)	3	(12%)	21	(72%)	5	(18%)
Poor Outcome	2	(7%)	22	(88%)	8	(28%)	23	(82%)

Table 6.6 presents outcome at six months by treatment group in a 2x2 contingency table. The assumptions of the Chi Squared test were met. Allocation to the intervention group was significantly associated with a good outcome ($\chi^2(1) = 17.09$, $p < 0.001$), with an odds ratio of 12.08 (95% CI 3.41, 42.75).

Table 6.6. Contingency table of outcome by intervention at 6 month assessment

Outcome	Intervention		Control		Total
Good outcome	21	(72%)	5	(18%)	31
Poor outcome	8	(28%)	23	(82%)	26
TOTAL	29 (100%)		28 (100%)		57

There were more cases of anxiety and depression (HADS subscale score ≥ 10) in the control group compared to the intervention group at baseline (anxiety: 4 cases in the intervention group, 9 in the control group; depression: 6 cases in the intervention group, 11 in the control group), see Table 6.7 below. Average HADS scores did not

change, see Table 6.4, but there was an increase in the number of cases of anxiety in the intervention group only. Cases of depression did not change.

Table 6.7. Cases of anxiety and depression, as defined by Hospital Anxiety and Depression Scale sub scores of 10 or greater

HADS Caseness	Intervention Group			Control Group		
	Baseline	4 Weeks	6 Months	Baseline	4 Weeks	6 Months
Anxiety	4	6	10	9	8	9
Depression	6	3	6	11	9	11

Table 6.8 and Table 6.9 present six month outcome by cases of anxiety and depression for the intervention group. The assumptions of the Chi Squared test were not met (cell counts less than 5), therefore Fisher's exact test was performed. Anxiety or depression 'caseness' was not statistically associated with a particular outcome.

Table 6.8. Contingency table of intervention group outcome at 6 months by anxiety caseness at baseline

Outcome	Anxiety case		Anxiety non-case		Total
Poor outcome	1	(25%)	7	(28%)	8
Good outcome	3	(75%)	18	(72%)	21
TOTAL	4 (100%)		25 (100%)		29

$X^2 (1) = 0.016$, $p=1.00$, Fisher's Exact Test = 1.00

Table 6.9. Contingency table of intervention group outcome at 6-months by depression caseness at baseline

Outcome	Depression case		Depression non-case		Total
Poor outcome	2	(40%)	6	(25%)	8
Good outcome	3	(60%)	18	(75%)	21
TOTAL	5 (100%)		24 (100%)		29

$X^2 (1) = 0.466$, $p=0.597$, Fisher's Exact Test, $p=0.597$

Further comparisons of baseline characteristics of the intervention group by good versus poor outcome is presented in Table 6.10. This comparison is limited by the small number of participants with a poor outcome. Differences are not statistically significant, except for 'pain self-rating of severe to extreme', (Fisher's exact test $p=0.0142$).

Table 6.10. Baseline characteristics of the intervention group by outcome

Baseline Characteristic / Score	Good Outcome n=21	Poor Outcome n=8	Intervention Group (all) n=30
% Female	81%	63%	73%
Age, mean (SD)	41.5 (14.2)	48.4 (10.3)	44 (13.1)
Graduate or post graduate qualification	57%	50%	53%
Symptom duration, years, mean (SD)	5.8 (8.8)	6.9 (8.1)	5.9 (8.3)
Not in work due to ill health	43%	50%	45%
Number PMH issues listed, mean (SD)	2.4 (1.98)	3.2 (2.8)	2.6 (2.2)
Pain self rating severe to extreme, n (%)	7 (33.3%)*	7 (87.5%)*	13 (45%)
Fatigue self rating severe to extreme, n (%)	10 (47.6%)	4 (50.0%)	15 (50%)
Primary Symptom, n (%)			
Mixed movement disorder	8 (38.1%)	3 (37.5%)	11 (37%)
Gait disturbance	4 (19.0%)	2 (25.0%)	7 (23%)
Tremor	4 (19.0%)	3 (37.5%)	7 (24%)
Weakness	3 (14.3%)	0	3 (10%)
Jerks	2 (9.5%)	0	2 (7%)
Baseline Clinical Outcome Measure Scores			
HADS Anxiety Score, mean (SD)	6.2 (3.7)	7.2 (4.4)	6.5 (3.8)
HADS Depression Score, mean (SD)	5.2 (4.2)	6.0 (3.8)	5.4 (4.0)
BIPQ Threat Score, mean (SD)	48.5 (10.2)	52.9 (12.6)	50.0 (10.8)
WSAS, mean (SD)	24.9 (7.0)	24.0 (10.6)	24.7 (7.9)
Berg Balance Scale, mean (SD)	37 (14.5)	37.5 (12.5)	39.0 (13.8)
10-metre Walk Time	33.9 (40.9)	17.3 (6.9)	16.8 (10.0)
DASH	48.9 (17.8)	59.3 (23.5)	51.8 (19.6)
Simplified-PMDRS	19.9 (9.5)	19.3 (14.3)	19.6 (10.6)
Short Form 36			
Physical Function	33.3 (21.8)	38.8 (29.5)	34.8 (23.7)
Physical Role	31.8 (28.9)	31.3 (31.0)	31.7 (28.9)
Bodily Pain	51.1 (32.1)	31.1 (34.9)	45.6 (33.5)
General Health	51.0 (20.1)	37.4 (31.3)	47.3 (23.9)
Vitality	33.6 (21.3)	28.9 (22.6)	32.3 (21.4)
Social Function	39.9 (31.8)	39.1 (39.2)	39.7 (33.2)
Role Emotional	74.2 (30.1)	59.4 (26.5)	70.1 (29.5)
Mental Health	67.6 (21.2)	60.0 (21.4)	65.5 (21.1)
Physical Summary Score	33.5 (10.9)	32.1 (12.3)	33.1 (11.1)
Mental Summary Score	46.5 (13.3)	41.7 (12.5)	45.2 (13.0)

* Statistically significant difference (Fisher's exact test $p=0.0142$)

6.11 Health Economic Analysis: Cost-Utility Analysis

The mean EQ-5D-5L utility scores at baseline, four weeks, and six months are presented in Figure 6.11. Using a linear regression model to adjust for baseline differences, the mean QALYs over six months for the intervention group was 0.34 (95% CI 0.31, 0.37) and 0.26 (95% CI 0.22, 0.30) for the control group with a mean gain in QALYs per patient of 0.08 (95% CI 0.03, 0.13). The cost of the intervention was estimated to be £1200 per patient. This took into account the time of the neurologist (£105 per hour)¹⁸² and physiotherapist NHS band 7 (£49 per hour),¹⁸² provision of equipment (usually downsizing walking aids and splints for some patients only), day hospital admission costs (including meals), and hotel accommodation (required for 85% of participants). The cost of the control was on average 4.8 sessions per patient multiplied by the cost of one hour of an NHS band 7 physiotherapist, or £233 per patient. Based on this data, the mean incremental cost per QALY gained was £12,087.

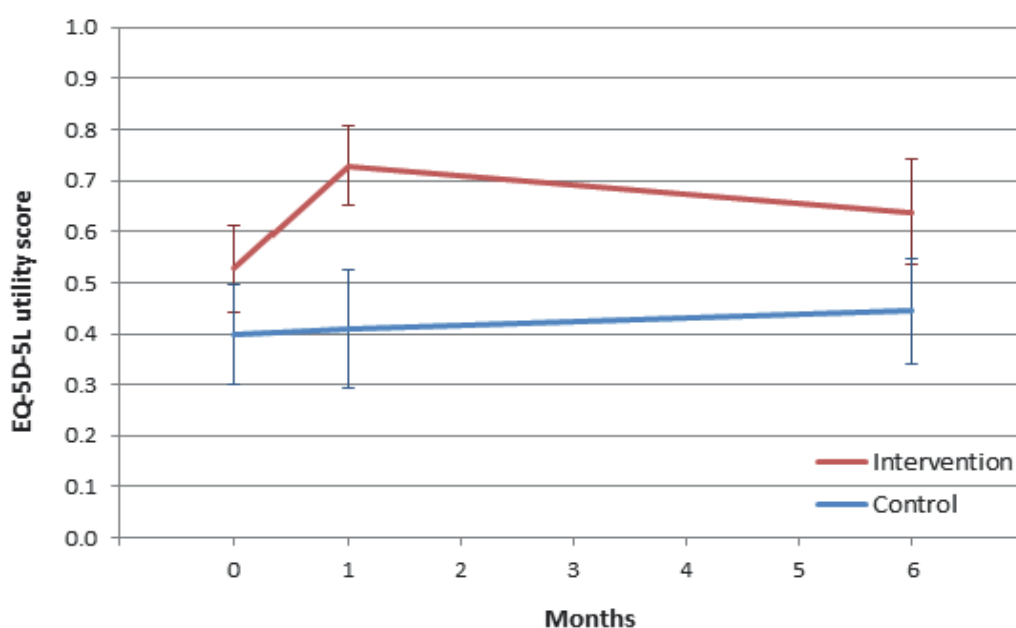


Figure 6.11. Mean EQ-5D-5L utility scores at baseline, 4-weeks and 6-months for the intervention and control groups. A utility score of 1.0 represents full health

6.12 Health Economic Analysis: Client Service Receipt Inventory

Table 6-11 displays services used and associated costs for both groups reported at baseline and six-month follow up. At baseline, the control group had higher service use costs than the intervention group (£719 compared to £579). There was a small reduction in total costs for the intervention group at six months compared to baseline (from £579 to £441) and an increase in total costs for the control group (from £719 to £1104).

Acute care costs (hospital based service) made up approximately 75% of total costs and community care 25% of total costs for both groups at both time points. Hospital outpatient appointments and day case procedures/tests were particularly common. The control group reported utilising these services more frequently than the intervention group. At follow up, 35% of the intervention group had attended a hospital outpatient appointment compared to 54% of the control group. Also at follow up, 10% of the intervention group had a day hospital admission compared to 36% of the control group. Acute care services came with a high cost and accounted for most of the difference in costs between the control and intervention groups.

The questionnaire did not allow for further details on hospital appointments, such as procedures performed or specialities under which participants were admitted, therefore assumptions based on mean costs were made which may have over- or under-estimated the true costs. The questionnaire also did not ask participants what they paid when they received private physiotherapy or occupational therapy. Private therapy costs were omitted from the total costs calculation as references for these costs are not well documented and in this case their impact would have been negligible.

Table 6.12 summarises the number of medications taken. At baseline, the mean number of medications taken regularly was 3.9 (SD 3.6) for the intervention group and 4.6 (SD 3.4) for the control group. Approximately 30% of participants took 5 or more

medications regularly. There was little change at follow up, although both groups had a slight reduction in numbers.

Days lost through sickness absence were calculated. The average number of sick days in the intervention group was 5.4 (SD 16.3) at baseline and 3.4 (SD 13.3) at follow up. Average sick days for the control group were 3.5 (SD 11.2) at baseline and 4.8 (SD 16.5) at follow up. The CSRI provided insufficient data to calculate sickness absence as a proportion of hours worked (a measure that accounts for differences due to part-time workers).

Overall, data completion for the CSRI questionnaire was satisfactory. Yes or No tick-box questions were rarely missed. When further qualification was required (i.e if yes was ticked for "Seen GP at the surgery", how many times did you see your GP?) data was missing in approximately 5-10% of cases. Only 16 participants wrote a response to the question "What is your gross income (i.e. before tax) per week?" This equates to 59% of those who were in paid employment. Most participants were able to remember their medications, though some did not know the correct spelling, which may have led to inaccurate data. Participants who brought a list of their current medication found this exercise easier, and this list was photocopied, anonymised and attached to the data collection sheet. In retrospect the headings for the table in question 3, in which participants were asked to list their medications were unclear and open to different interpretations. These headings were "Duration of use (days)" and "Dose/no. per day". The CSRI questionnaire is presented in Appendix 5 (page 231).

Table 6.11. Services used and costs, data from Client Service Receipt Inventory

Resources utilised		BASELINE		FOLLOW-UP	
		Intervention n=30	Control n=30	Intervention n=29	Control n=28
Table legend	Yes	Number of participants utilising the service (%)			
	Number (if yes)	Mean frequency of service utilisation amongst users (SD)			
	Cost (all)	Average cost of service use per participant (SD)			
	Missing (count)	Participants with missing data			
GP – practice	Yes	22 (73.33%)	22 (73.33%)	24 (83%)	20 (71 %)
	Number (if yes)	2.95 (2.23)	3.77 (2.81)	2.2 (3.1)	3.5 (1.8)
	Cost (all)	£70 (76)	£91 (97)	£59 (72)	£78 (72)
	Missing (count)	2	0	2	3
GP - home	Yes	1 (3.33%)	2 (6.67%)	3 (10.34%)	2 (7.14%)
	Number (if yes)	4	2 (1.4)	1 (0)	3
	Cost (all)	£7.26 (40)	£7.26 (31)	£3.89 (14)	£6.05 (31)
	Missing (count)	0	0	1	1
GP - phone	Yes	11 (36.67%)	11 (36.67%)	7 (24%)	9 (32%)
	Number (if yes)	1.63 (1.19)	3.27 (4.20)	1.3 (0.52)	5.3 (9.7)
	Cost (all)	£10 (20)	£24 (59)	£5.71 (12)	£26 (100)
	Missing (count)	3	0	1	3
Nurse - practice	Yes	9 (30%)	10 (33.3%)	11 (38%)	7 (25%)
	Number (if yes)	3.88 (6.56)	1.3 (0.48)	1.6 (0.8)	2.2 (1.2)
	Cost (all)	£13 (45)	£5 (8)	£11.79 (18)	£9.15 (20)
	Missing (count)	1	0	0	1
Home Help	Yes	0	2 (6.67%)	0	1 (3.57%)
	Number (if yes)	0	5.5 (2.1)	0	84
	Cost (all)	0	£11 (44)	0	£36 (190)
	Missing (count)	0	0	0	0
Social Work	Yes	0	2 (6.67%)	0	1 (3.57%)
	Number (if yes)	0	1.5 (0.7)	0	3
	Cost (all)	0	£6 (22)	0	£5.89 (31)
	Missing (count)	0	0	0	0
Physiotherapist - hospital	Yes	7 (23.33%)	13 (43%)	5 (17%)	16 (57%)
	Number (if yes)	3 (2.1)	4.9 (5.4)	2.8 (2.9)	4.3 (4.1)
	Cost (all)	£24 (54)	£64 (139)	£16 (53)	£82 (127)
	Missing (count)	0	1	0	1
Physiotherapist - home	Yes	0	2 (6.7%)	0	3 (10.71%)
	Number (if yes)	0	1.5 (0.7)	0	1
	Cost (all)	0	£6.7 (27)	0	£2.58 (13)
	Missing (count)	0	0	0	2
Physiotherapist - GP practice or clinic	Yes	2 (6.7%)	3 (10%)	1 (3.45%)	2 (7.14%)
	Number (if yes)	2.5 (2)	2.5 (2)	0	12
	Cost (all)	£6 (27)	£6.21 (27)	0	£16 (83)
	Missing (count)	0	1	1	1
Physiotherapist Private - home	Yes	1 (3.3%)	0	0	0
	Number (if yes)	5	0	0	0
	Cost (all)				
	Missing (count)	0	0	0	0
Physiotherapist Private	Yes	3 (10%)	2 (6.7%)	1 (3.45%)	5 (17.86%)
	Number (if yes)	1.7 (0.6)	11.5 (12)	2	5 (1.4)

Resources utilised		BASELINE		FOLLOW-UP	
		Intervention n=30	Control n=30	Intervention n=29	Control n=28
Outpatient	Cost (all)	*			
	Missing (count)	0	0	0	0
Physiotherapist Private Hospital	Yes	2 (6.7%)	0	0	0
	Number (if yes)	4 (0)	0	0	0
	Cost (all)	*			
	Missing (count)	0	0	0	0
Occupational Therapist Hospital (NHS)	Yes	3 (10%)	1 (3.6%)	0	3 (10.71%)
	Number (if yes)	1	1	0	2 (0)
	Cost (all)	£1 (5.2)	£1 (5.2)	0	£6 (18)
	Missing (count)	2	0	0	0
Occupational Therapist Home (NHS)	Yes	1 (3.3%)	3 (10%)	1 (3.45%)	2 (7.14%)
	Number (if yes)	1	1.5 (0.7)	0	1 (0)
	Cost (all)	£1.30 (7.1)	£4.17 (16)	0	£2.79 (10)
	Missing (count)	0	1	1	0
Occupational Therapist Hospital or Home (Private)	Yes	0	0	0	0
	Number (if yes)				
	Cost (all)				
	Missing (count)				
Accident & Emergency	Yes	3 (10%)	5 (17%)	3 (10.34%)	4 (14.29%)
	Number (if yes)	2 (0)	1.2 (0.4)	1.3 (0.6)	1.8 (1.0)
	Cost (all)	£25 (76)	£25 (60)	£17 (55)	£31 (87)
	Missing (count)	0	0	0	0
Elective Overnight hospital stay	Yes	1 (3.3%)	2 (6.7%)	0	0
	Bed days (if yes)	2	2		
	Cost (all)	£94 (513)	97 (522)		
	Missing (count)	0	1		
Non-Elective Overnight hospital stay	Yes	2 (6.7%)	3 (10%)	1 (3.45%)	1 (3.57%)
	Bed days (if yes)	5 (2.8)	5.5 (6.4)	9	1
	Cost (all)	£176 (724)	£200 (980)	£164 (881)	£19 (100)
	Missing (count)	0	1	0	0
Intensive care or High dependency overnight stay	Yes	0	1 (3.3%)	1 (3.45%)	1 (3.57%)
	Bed days (if yes)		4	4	5
	Cost (all)	0	£33 (181)	£34 (184)	£35 (187)
	Missing (count)	0	1	0	0
Outpatient hospital appointment	Yes	14 (46.7%)	22 (73.3%)	10 (35%)	15 (54%)
	Number (if yes)	1.7 (1.3)	2.7 (4.2)	1.4 (0.5)	2.6 (2.2)
	Cost (all)	£85 (136)	£216 (422)	£50 (82)	£149 (226)
	Missing (count)	1	1	0	1
Day case procedure or test	Yes	1 (3.3%)	6 (20%)	3 (10.34%)	10 (35.71%)
	Number (if yes)	4	1.8 (1.3)	1 (0)	2.1 (1.8)
	Cost (all)	£94 (514)	£258 (653)	£49 (182)	£495 (1011)
	Missing (count)	0	0	0	1

See next page for totals.

Resources utilised		BASELINE		FOLLOW UP	
		Intervention n=30	Control n=30	Intervention n=29	Control n=28
Community care cost	Average (SD) Missing no. (%)	£118 (146) 2 (6.7%)	£202 (367) 1 (3.3%)	£77 (67) 3 (10.34%)	£192 (243) 4 (14.29%)
Acute care cost	Average (SD) Missing no. (%)	£449 (1038) 2 (6.7%)	£517 (850) 2 (6.7%)	£330 (1105) 0	£840 (1227) 2 (7.14%)
Total cost	Average (SD) Missing no. (%)	£579 (1056) 3 (10%)	£719 (905) 2 (6.7%)	£441 (1167) 3 (10.34%)	£1104(1378) 6 (21%)

Community care costs are those that do not occur in a hospital setting (e.g. GP, practice nurse, community physiotherapy and social work). Acute care costs are costs incurred in a hospital setting (e.g. inpatient admissions and outpatient hospital appointments). The sum of each column does not exactly equal listed total costs due to adjustments for missing data.

* Costs were not calculated for these items as they are unknown, and were overall negligible to the total costs. Future questionnaire ask for private therapy costs.

Table 6.12. Number of medications taken

Number of Regular Medications	BASELINE		6 MONTHS	
	Intervention n=30	Control n=30	Intervention n=29	Control n=28
Mean (SD)	3.9 (3.6)	4.6 (3.4)	3.4 (3.7)	3.9 (3.7)
Median	3	4	2	3
Min	0	0	0	0
Max	13	14	13	14
Taking 5 or more medications	33%	47%	28%	29%

Chapter 7 Feasibility Study Discussion

7.1 Key Findings Related to Feasibility

A randomised feasibility study of specialist physiotherapy for FMD was conducted, with the primary aim of determining the feasibility of conducting a large scale RCT of such an intervention. Recruitment rate, enrolment and retention were high and clinical outcomes were promising, providing evidence that an appropriately powered RCT is feasible, timely and important.

7.1.1 Recruitment and Retention

Thirty-two per cent of new patients presenting with FMD to the recruiting neurology clinic met the selection criteria, with a 90% enrolment rate and only a 5% drop out rate subsequently from the study. Given the high prevalence of such patients in general neurology clinics it follows that there should be sufficient patients to run a larger version of this trial.^{4,183} Problems with travel to follow up assessment appointments were the most common reason for missing data. A future trial that minimises travel requirements should reduce loss.

7.1.2 Fidelity to the Intervention

Fidelity to the intervention as gauged by attendance, completion of the intervention workbook and participant feedback was high. Fidelity is defined as the degree to which the intervention was implemented as intended. Assessing fidelity is an important consideration in the design of a large trial, as it is required in order to make causal claims about the intervention and to reduce the chance of type I or II errors.¹⁸⁴ The treatment workbook provided a useful measure of fidelity as it logged the education, movement retraining and self-management components of the intervention. In addition, the daily reflections that participants were requested to write provided insight into their levels of engagement. In the feedback forms, participants were asked to respond to the following statements: The programme helped me understand my symptoms; The programme helped me gain more control over my symptoms; The

programme has helped me to create a plan to improve my symptoms; and The programme included information about thoughts, feelings and psychological influences on my symptoms. These statements relate to the key components of the intervention, which were: education, movement retraining, enabling self-management and considering symptoms within a biopsychosocial framework. These measures of fidelity proved to be practical and useful.

7.1.3 Acceptability of the Intervention

The high rates of recruitment, retention, and fidelity point to the intervention being acceptable. This is supported by the positive ratings in the post intervention feedback questionnaire.

7.1.4 Safety

There were no reported serious adverse events associated with the intervention in either group. Temporary exacerbation of chronic pain and fatigue was not considered to be an adverse event. On the contrary, exposing such patients, who may otherwise exhibit avoidant behaviour, to activity may have therapeutic value. It is likely that the relatively high intensity and short duration is an important therapeutic element of the intervention, but this may make it unsuitable for some patients. With this in mind, patients were excluded if chronic pain or fatigue was the dominant problem; however half the enrolled participants still rated their pain and fatigue as severe to extreme at baseline.

No mental health related adverse events associated with the physiotherapy treatment were found. Patients with clinically evident anxiety and depression warranting assessment were excluded from the study and referred to more appropriate treatment. On average, measures of mental health (HADS and SF36 Mental Health domain) did not change in either group.

7.1.5 Determining a Primary Outcome for a Future Trial

An important aim of this research was to test the utility of a range of outcome measures and determine which is the most suitable to use in a power calculation for a future clinical trial. Measuring outcome in FMD is complicated by the variable nature of symptom severity inherent to the diagnosis. For this reason, snapshot measures of disability are likely to have problems with test-retest reliability, limiting their usefulness. Gait and balance outcome measures are restrictive as they are not applicable to patients with upper limb symptoms only. The SF36 Physical Function domain was the most promising potential primary outcome. It had a medium to large effect size ($d=0.70$), it was applicable to all participants, and it is not as vulnerable to symptom fluctuation as answers are given based on the respondent's perception of the average experience within the set recall period of four weeks.

7.2 Clinical Outcomes

To my knowledge, this is the first reported randomised study of physical rehabilitation for FMD, with a control period greater than four weeks. With the caveat that this research was primarily designed to assess feasibility, the study found a moderate to large treatment effect size across a range of measures of physical function, compared to a treatment as usual control. In addition, a larger proportion of the intervention group rated their symptoms as improved (72%) compared to the control group (18%) on the Clinical Global Impression scale (CGI). Also of note, 32% of the control group felt their symptoms had worsened over the follow up period, compared to only 3% in the intervention group.

The agreement between the CGI (patient impression of change) and other outcome measures in the intervention group, suggests the reported treatment effect most likely represents a clinically significant difference. Minimum clinically important difference (MCID) values differ between conditions and vary widely between studies. The MCID for the SF36 has not been well established. A review of MCID in hip and knee orthopaedic surgery found 3 studies reporting MCID for the SF36 Physical Function domain; these were 3.2 (95% CI 2.8,3.9), 11.6 (95% CI 6.5-16.7) and 20.4 (95% CI 14.4-

26.4)¹⁴². The difference between groups in the current study for this outcome was 19.8 (95% CI 10.2, 29.5).

The improvements reported occurred in a sample of patients with characteristics commonly associated with a poor prognosis. The average symptom duration was 5.8 years (SD 7.3), participants had multiple coexisting symptoms, and high rates of unemployment due to ill health. It is possible that if the intervention occurred earlier in the course of their disorder, it may have been more effective.

Subgroup analyses were performed as an exercise in hypothesis generation, recognising that there was insufficient power to detect a statistically significant association. In the intervention group, there was no association between the presence of anxiety or depression (as defined by a HADS anxiety or depression score of 10 or greater) and outcome (good outcome or poor outcome as defined by the CGI). When the baseline demographic and clinical variables of the intervention group were compared between those with good versus poor outcome, there was a significantly higher proportion of participants with high pain levels (self-rating of severe to extreme pain) in the poor outcome subgroup. This needs further investigation, however it may indicate that severe pain is associated with a worse prognosis. If this is the case, it may suggest that a different treatment approach is required when patients report severe pain; or that a future trial may wish to consider stratifying participants by pain level during randomisation. There is no good evidence that a future trial should exclude participants based on any of the variables measured in this trial, which suggests the study eligibility criteria were appropriate.

7.2.1 Comparison to Similar Studies

The effect size of the intervention across the outcome measures assessed is consistent with those in similar published studies in FMD. Direct comparison of study outcomes is limited by differences in study design, interventions, follow up time frames, and use of outcome measures.

Jordbru et al (2013)⁷³ reported a larger treatment effect at 12 months in the Functional Mobility Scale and Short Form 12 physical score (comparable to the Short Form 36 Physical Summary score¹⁸⁵) compared to the current study (Functional Mobility Scale: 6.9 units compared to 3.4; SF12: 11.7 units compared to 5.4). Some of this difference may be explained by the shorter symptom duration in their participants (9.5 months compared to 5.8 years) and lower scores at baseline. Other significant differences between studies include the nature of the intervention (three week multidisciplinary inpatient rehabilitation) and absence of a control comparison beyond 4 weeks, so regression toward the mean is not accounted for in their 12-month follow-up measures.

My previous cohort study of the same intervention, published in 2015,¹ reported similar outcomes at three months to the current study. The gain in the SF36 Physical function domain was greater in the current study (19.8 units compared to 8.9), but other outcomes showed a slightly greater treatment effect in the cohort study, including SF36 Physical Role, Berg Balance Scale and 10-metre timed walk. An interesting difference between these studies was the proportion of the different symptom phenotypes. The earlier cohort study had a larger proportion of subjects with fixed dystonia (25% compared to 3% in the current study), a condition that shares many characteristics with complex regional pain syndrome and is often associated with soft tissue contracture.⁶⁵ As such, fixed dystonia may have physiological limitations on recovery rate.

Czarnecki et al (2012)⁷⁵ reported a retrospective cohort of 60 patients with FMD who received an intensive 5-day physiotherapy and occupational therapy intervention following a mechanistic explanation for their symptoms (symptoms due to disconnect between brain and body). Outcome was reported as patient perception of change. Immediately after treatment, 69% of patients rated their symptoms as markedly improved or resolved and 60% at one year postal follow. These results are similar to the 72% with a good outcome at six months reported in the present study.

7.3 Health Economic Analysis

7.3.1 Cost Utility Analysis

The EQ-5D-5L is the preferred instrument for generating Quality Adjusted Life Years (QALYs) by the UK organisation the National Institute for Health and Care Excellence (NICE).¹⁸⁶ The average difference in QALYs between the groups adjusting for baseline differences was 0.08 QALYs and the resulting incremental cost effectiveness ratio of (ICER) £12,087 suggests the intervention is most likely to be cost effective. In general, an ICER below £20,000 is considered cost effective.¹⁷⁷ This is without accounting for a potential reduction in the costs of health and social care utilisation, reduction in disability benefits and return to paid employment. In our sample, we found little overall change in employment status over the six month period. In the intervention group, one participant was medically retired, another reduced to part time hours and one participant moved from unpaid voluntary work to part time paid work. In the control group one part time worker stopped work due to ill health.

ICER's are reported by a number of large therapy trials for chronic conditions. An appropriate comparison to the current study is the PACE trial, which compared adaptive pacing, cognitive behavioural therapy (CBT), graded exercise and specialist medical care for symptoms of chronic fatigue syndrome at 12 months.¹⁸⁷ Compared to specialist medical care, the ICER's were £18,374 for CBT, £23,615 for graded exercise and £55,235 for pacing. Studies of therapy for subacute to chronic low back pain (including group treatments) have reported more impressive ICER's ranging from £1,786 to £8,700.¹⁸⁸

7.3.2 Health Service Use: CSRI

The primary purpose for including the CSRI was to inform the design of a version of this questionnaire to be used in a future RCT. Overall the CSRI questionnaires had fair levels of completeness. Adjustments to the questionnaire may help to reduce missing data. Only 59% of eligible employees reported their annual income and participants may have been uncomfortable sharing this information. An alternative to consider in a

future iteration of the CSRI would be to estimate incomes using standard reference figures based on reported occupation and hours worked. There was insufficient information in the CSRI to determine sickness absence as a proportion of hours worked and the cost impact of improved engagement with employment. An additional questionnaire, such as the Work Productivity and Activity Impairment Questionnaire,¹⁸⁹ may be required for a more robust cost-impact analysis in a future trial. Encouraging participants to bring pharmacy medication lists to their appointment should help improve the accuracy of medication reporting.

Acute care costs made up 75% of the total costs and a future version of the CSRI should focus more attention on these costs. More information is required in order to reduce the number of assumptions made regarding the actual services received and the associated costs. Additional information should include the reason for any hospital admission, the medical discipline under which the admission was recorded (e.g. neurology, endocrinology, etc.), the procedure(s) or interventions performed, the length of appointment/admission, and the health professionals/disciplines involved. A future CSRI should also ask for costs paid for private services and to improve the quality of the data, the questionnaire could be administered rather than self-report.

Overall, the intervention group reported lower total costs than the control group at six months, £441 (SD 1167) compared to £1,104 (SD 1378). All service costs were higher for the control group, except for seeing a nurse at the GP practice and elective overnight hospital admissions. In addition, the intervention group reported a reduction in sickness absence. These results are promising; however, in order to consider if the intervention may have contributed to the cost difference found, a larger sample size and more information would be required. Given that some service costs for both groups reduced at six months (e.g. GP appointments, community physiotherapy, non-elective hospital admissions), regression to the mean may account for some changes.

7.4 Mechanism

The B-IPQ total score is thought to represent the threat value of an illness. The intervention was associated with a reduction in the B-IPQ total and there was little

change in the control group. It is hypothesised that the intervention helped participants to understand their symptoms and improve control over their movement, both of which resulted in diminished concerns. This may represent one mechanism by which the intervention affects change, although it is noted that in some health conditions, the total B-IPQ score as an outcome measure may have problems with internal consistency.¹⁹⁰

On average, measures of mental health (HADS anxiety and depression scores and the SF36 Mental Health domain) did not change; however an interesting finding was an increase in the number of participants reaching levels of anxiety 'caseness' in the intervention group. This may be a clinically insignificant chance occurrence. However if it represents a true change in these participants, it can be interpreted in a number of ways. A superficial interpretation is that the intervention made some participants more anxious. An alternative explanation is that the intervention helped some participants gain a better insight into their symptoms, leading them to rate their anxiety higher; and/or the trust and therapeutic rapport developed over the intervention period reduced the participant's concerns about reporting psychological symptoms. These interpretations would provide support for the intervention aim of helping the patient to understand their problem within a biopsychosocial framework.

7.5 Limitations

There are a number of limitations to this study. It was not specifically designed or powered to detect a treatment effect. However, given the absence of controlled trials in the literature it was considered appropriate to report the clinical outcomes. At baseline, the control group had scores that represented worse health than the intervention group. The analysis accounted for baseline differences and there was still a large treatment effect with the intervention and little or no change with the control condition. A larger, powered study is less likely to have such discrepancies between groups; however a future trial could also consider a randomisation procedure that involved minimisation to account for baseline severity.

Another limitation was that the participants and assessors were not masked to treatment allocation, which may have introduced bias. Most outcomes were subjective patient reported outcomes, which may be influenced by many factors, including the lack of blinding. However the S-FMDRS was scored blindly (by video) and showed the largest treatment effect size ($d=0.79$).

A potential criticism is that the criterion for excluding patients with anxiety and depression was not operationalised. Clinical judgements were made by clinical impression based on whether it was felt that levels of anxiety or depression would interfere with engagement with the treatment, as we were unable to find an assessment tool suitable for this purpose. Other limitations were the lack of standardisation of the control condition and that follow up was limited to six months. Additional reflections on the trial methodology can be found in Appendix 8 (page 242).

7.6 Generalisability

A strength of this study was that the sample was generally representative of the population suitable for physiotherapy, thereby increasing generalisability. We did not exclude participants on the basis of age, symptom duration, phenotype, and availability of medical insurance as other studies have done.^{73,75} Also, most eligible participants elected to take part. We did however exclude participants with a primary problem of pain, fatigue or dissociative seizures and those who had significant comorbid psychopathology, for which there are specific specialist treatments available on the NHS.

Being a single centre study, it is not known whether these results could be replicated outside the study setting; thus, a multicentre RCT is required. Together with the previous cohort study,¹ this study provides useful preliminary evidence that other physiotherapists can be taught to deliver the intervention.

7.7 Clinical Implications

This study adds to a growing body of evidence supporting specialist physiotherapy and physical rehabilitation for FMD. The intervention in this research differs from standard

physical rehabilitation in a number of key ways. Firstly, and similarly to some physiotherapy led interventions for chronic pain,¹⁶⁰ there was an emphasis on symptom education. Secondly, movement retraining aimed to distract participants' attention away from the body. Rehabilitation for neurological disease often encourages the patient to think about movement or attend to their body, which is known to exacerbate FMD and therefore such an approach can be counterproductive. The different approach required for treatment of FMD compared to other neurological symptoms suggests there may be a role for specialist clinicians and highlights a need for treatment to be delivered by experienced or well informed clinicians. Another potential difference from standard physiotherapy was the high intervention intensity and short duration.

7.8 Future Research

Aside from support for progressing to a powered RCT, this study highlights some directions for future research. Measuring outcome in FMD remains challenging and currently available outcome measures do not appear to reflect the true burden of the diagnosis. An assessment tool specifically designed for FMD may increase sensitivity, validity and reliability of outcome measurement. A specific tool should consider the burden from comorbidity (such as pain, fatigue, anxiety, bladder dysfunction, etc.); the variable nature of the motor symptoms, and the impact across biopsychosocial domains. The intervention could be further developed and refined based on research aiming to elucidate the mechanisms by which it is likely to work. There may be several factors mediating recovery, possible candidates are: a change in illness belief, reduced self-focused attention during movement, acceptance of the diagnosis, understanding the diagnosis, reduced threat value of symptoms, a sense of self efficacy, reduced avoidance behaviour, reduction in perceived pain level, and increased physical activity. Finally, there may be value in exploring common symptom phenotypes (e.g. weakness, tremor, dystonia) independently, which may lead to more effective treatments.

7.9 Conclusions

In summary, this study demonstrates the feasibility of performing a larger trial of specialist physiotherapy for FMD. A large treatment effect and evidence of cost benefit was found in a group of patients that are prevalent, have poor quality of life and have a poor prognosis with the current standard treatment. The study data strongly support the need for a multicentre randomised trial of this intervention.

7.10 Acknowledgements and Contributions

A report of this study was previously published in the Journal of Neurology, Neurosurgery and Psychaitry.¹⁹¹ The first draft of the published manuscript was written by Glenn Nielsen. The following people helped to revise the manuscript: Mark Edwards, Marta Buszewicz, Fiona Stevenson, Eileen Joyce and Rachael Hunter.

Glenn Nielsen and Mark Edwards devised the physiotherapy treatment programme. The following individuals contributed to the design of the study: Glenn Nielsen, Mark Edwards, Marta Buszewicz, Fiona Stevenson, Eileen Joyce, Rachael Hunter, Lucia Ricciardi, and Jonathan Marsden. Kate Holt delivered the trial intervention. Magdalena Dudzic completed data collection. Glenn Nielsen completed data analysis. Rachael Hunter completed health economic analysis.

The study was supported by the National Institute for Health Research (NIHR/HEE Clinical Doctoral Research Fellowship, GN, CDRF-2013-04-034). The study was also supported by the National Institute for Health Research Clinical Research Network (NIHR CRN), who provided funding for a research physiotherapist. PRIMENT Clinical Trials Unit and two patient members of the trial advisory committee provided advice on trial design.

Chapter 8 Qualitative Study – Introduction and Methods

A Longitudinal Qualitative Study of Patients with Functional Motor Disorder Selected for Specialist Physiotherapy

8.1 Introduction

In the previous chapters I have described the development and feasibility assessment of trialling a specialist physiotherapy programme for patients with functional motor disorder (FMD). Embedded in this research, was a longitudinal qualitative study of the subjective experiences of participants receiving the study intervention.

There is little qualitative data about patients with FMD. Studies of neurologists, neuroscience nurses and physiotherapists report that clinicians find these patients clinically challenging and often have negative attitudes towards them.^{145,192,193}

However, we know little about patients' perspectives, such as their perceptions about getting this diagnosis and interactions with health care professionals (HCP), how it feels to have FMD and priorities and preferences for treatment. Results from previous clinical trials suggest that physical rehabilitation for FMD leads to improved measures of disability and quality of life,^{1,73,75} but these quantitative values provide little insight into the real world impact of treatment. Qualitative research methods are an ideal way to explore these unknowns, which are important to consider in order to develop an intervention that is both effective and acceptable to patients.

Qualitative research embedded into a clinical trial can provide additional rich explanatory data. This includes information about the acceptability of the intervention, potential mechanisms involved in the intervention, explanations for why a particular outcome occurred, and outcomes that may not be captured with quantitative measures. At the feasibility stage, findings from qualitative research can be used to optimise the intervention and inform the design and conduct of a definitive clinical trial.¹⁹⁴

The current literature, while limited, provides some data on patient perspectives. Nettleton et al (2005)¹⁹⁵ explored perspectives of patients with “medically unexplained” neurological symptoms with a narrative analysis of in-depth interviews. This cohort of 18 patients recruited from neurology clinics was similar to those in the current feasibility study in that participants had motor symptoms (spasms and paralysis) and/or non-motor symptoms (blurred vision, non-epileptic seizures, pain, fatigue, etc.). A difference was that some of the patients in the Nettleton study only had non-motor symptoms. A key finding from that study was that participants commonly felt that clinicians, family and acquaintances saw them as a fraud, time waster, hypochondriac or malingerer and that such perceptions were associated with their symptoms being classified as psychological. Patients felt they had been marginalised from medicine; the authors used the phrase *Medical Orphans* to describe this. The patients’ narratives were classified as chaotic, without a clear beginning, middle and end. This was related to their difficulties in articulating their problem and entering a sick role that was legitimised by the system. The authors suggest that the practical implications from their findings include the importance of listening to patients’ narratives, and acknowledging their symptoms as genuine. They also suggest there may be value in the doctor providing explanations that may in part be consistent with the patient’s point of view.

Qualitative research methods have been used more extensively to study patients with the related condition of non-epileptic seizures. Non-epileptic seizures, also called functional seizures, are usually thought to have a shared aetiology with FMD and patients with non-epileptic seizures also commonly experience functional motor symptoms. Rawlings and Reuber (2016)¹⁹⁶ produced a narrative systematic synthesis of qualitative studies investigating patients’ accounts of living with non-epileptic seizures. From 21 studies including 220 patients they identified themes relating to the symptom (seizure event), diagnosis, treatment, emotional events and the impact of the symptoms. In regards to the diagnosis, they found that many patients felt confused and often exhibited resistance to psychological attributions being suggested. Patients felt isolated, they usually described an inability to work and often felt a sense of loss.

The illness beliefs of patients with FMD have been explored using questionnaires. It was found that a patient expectation of non-recovery and non-attribution of symptoms to psychological factors (believing that psychological factors are not relevant) are predictive of poor outcome.¹⁵³ This study supports the importance of considering patients' beliefs and expectations as part of their treatment, although the research was not designed to explore why participants held particular illness beliefs.

Thus, the existing small body of research shows that patients with FMD often have difficulty understanding the diagnosis and often show resistance to psychological explanations. There is however little data to suggest why patients may hold these particular points of view and how their beliefs and experiences influence their interaction with treatment. Confirming or disconfirming these findings and exploring other themes in a cohort with an established diagnosis of FMD is of high clinical relevance.

In this qualitative study, I aimed to develop an understanding of the participants' perspectives and experiences of their journey from developing FMD to completing the study intervention. In particular, I planned to explore how they interacted with the intervention and its impact on their life at six months follow up. To gain a greater understanding of the intervention, I planned to explore the data for evidence of how it may have helped participants, and the underlying mechanisms for this, or conversely why it may have failed to help.

8.2 Methods

8.2.1 Ethics Approval and Consent

Ethics approval for this study was obtained from the National Research Ethics Service Committee London – City Road & Hampstead (14/LO/0572). All participants gave written informed consent. See Appendix 9 and 10 (page 246 and 249) for patient information sheet and consent form.

8.2.2 Study Design

I employed longitudinal semi-structured interviews to explore participants' beliefs, perceptions and experiences. The longitudinal design enabled change associated with the intervention to be a key focus of the interviews and analysis, as well as investigating the possible causes and consequences of such changes and the usefulness or otherwise of the intervention over a longer period.¹⁹⁷

8.2.3 Purposive Sampling

I selected the qualitative study participants purposively from those allocated to the intervention condition of the feasibility study. Purposive sampling is designed to achieve symbolic representation of the diversity of the population being studied but not necessarily a proportional and statistically representative sample.¹⁹⁸ The purposive sampling criteria were designed to ensure representation of common symptom phenotypes, symptom duration, age and gender. See Table 8.1 for purposive sampling criteria. A minimum of eight participants were required to meet these criteria, but I planned to recruit a minimum of 10 participants, and up to 20 if I felt that I had not approached data saturation.

Table 8.1. Purposive sampling criteria (participants could be represented in more than one box)

Criteria		Aimed minimum number to recruit
Symptom	Tremor	1
	Gait Disturbance	1
	Mixed Movement Disorder	1
	Weakness	1
Symptom duration	Less than 18 months	2
	18 months to 5 years	2
	More than 5 years	2
Age bracket	18-29	2
	30-59	2
	60+	2
Gender	Male	3
	Female	5

I chose these particular purposive sampling criteria as they may influence the lived experience of having FMD or be associated with different prognoses. For example,

different symptom phenotypes may have greater or lesser association with common comorbidities such as pain, fatigue, anxiety or depression; however, research exploring subgroups of FMD is scarce. A systematic review of the prognosis of FMD found that a shorter duration of symptoms has consistently been found to predict a better outcome; whilst younger age predicted a better outcome in some studies but not others and no study has found gender to have an effect on outcome.⁷⁰ I am not aware of any studies comparing the outcomes of functional motor symptom phenotypes. As baseline interviews were conducted prior to commencing the intervention, it was not possible to purposively select participants based on intervention outcome; that is, to ensure a mixture of participants rating their symptoms as improved, no change and worse.

Symptom phenotypes were tremor, gait disturbance, weakness and mixed movement disorder. Mixed movement disorder represented a mixture of dystonic-type movement, tremor and weakness. Symptom duration brackets were chosen to ensure inclusion of participants in the 'relative' acute phase (less than 18 months); participants with chronic symptoms (18 months to 5 years); and those with a long term condition (5 years or greater). Age brackets were chosen to represent key phases of adulthood: 18-29 = 'Early' adulthood, capturing participants prior to having dependents/children and an established career; 30-59 = 'Mid' adulthood, capturing participants who may have dependents and are at working age; and 60+ = 'Mature' adulthood, capturing participants who may be in or approaching retirement. Nested criteria in the form of a sampling matrix (e.g. having a spread of ages across symptom type) was not feasible given the proposed sample size. The gender proportion was biased toward females, as this reflects epidemiological findings that FMD is more common in women by a factor of 2–3:1.²⁰

8.2.4 Data Collection

I interviewed participants in the qualitative study at (i) baseline (following initial consultation with the study neurologist, but prior to starting the intervention), (ii) post-treatment (after the final physiotherapy session), and (iii) at six months follow up. Interviews were conducted in outpatient clinic rooms at the National Hospital for

Neurology and Neurosurgery. Family, friends and partners were asked to step out of the room during interviews. The interviews were semi-structured, following a topic guide. The topic guide was prepared and piloted in a practice interview. The topic guide was updated throughout the data collection period in an iterative manner to explore emerging themes. I recorded Interviews using a digital voice recorder (Olympus VN-732PC) and later transcribed them verbatim, with identifying data removed to preserve anonymity.

Where possible, I transcribed interviews on the day they were recorded and aimed to complete interview transcription within one week. During transcription, interviews were played at reduced speed (50-100%) using Media Player for Windows version 7 and transcripts were typed into Word, Microsoft Office Professional Plus 2013.

8.2.5 Data Collection Reflexivity

I introduced myself to interview participants as a physiotherapist who was part of a team researching FMD. The study neurologist may have previously introduced me as a specialist physiotherapist in FMD in the clinic where patients were being recruited for the study. How the participants responded to the interview questions is likely to have been influenced by my perceived status and connection to the treating clinicians. For example, some participants may have felt concerned about appearing ignorant when discussing their understanding of their disorder or concerned about appearing ungrateful when discussing ongoing problems after the intervention. On a few occasions, some interviewees avoided answering questions by turning the question back on me. I responded by saying that I did not know the correct answer or that there were no right or wrong answers. I attempted to distance myself from the clinical team and environment by wearing casual clothes, choosing interview rooms with less of a clinical feel and providing tea and coffee.

From the outset of this study, I had a particular understanding or conceptual model for FMD that was influenced by my experiences working with this patient group over several years. This experience included working within a neuropsychiatry led multidisciplinary rehabilitation programme and setting up a specialist physiotherapy

service for FMD. I considered FMD to be a genuine problem that is distinct from malingering and to be a heterogeneous biopsychosocial condition that is commonly precipitated by physical events. I believed that psychological factors are relevant to FMD, but that the relative importance and severity varies between individuals. My assumption was that patients with less significant psychopathology were more likely to benefit from the study intervention. My particular beliefs may have impacted on my line of questioning and the focus of the interviews.

Prior to this study, I had no experience in qualitative research. As the study progressed, through a process of regular supervisory feedback and reflection, my interview skills developed leading to better quality data. Transcribing interviews as I went along allowed me to receive and act upon supervisory feedback. Personally transcribing each interview and by necessity listening and re-listening to each interview during transcription gave me the opportunity to reflect on my interview technique and get to know my data in depth.

An early limitation of my interview style was asking clinically orientated questions, such as asking for information regarding previous interventions and their outcome. I was mindful of avoiding 'clinical questions' in subsequent interviews and endeavoured to maintain a focus on the participants' real-life experiences and perceptions. Another limitation of my early interviews was that at times I would summarise what people had said, potentially imposing my interpretation and meaning on the conversation. Other early limitations and potential issues that I noted and was mindful to avoid in subsequent interactions were: filling 'awkward' silences rather than leaving space for the participant to talk; closing topics of conversation too early; leading questions (e.g. "Was it difficult when..."); double questions (why and how); and finally on a few occasions I prefaced questions with an apology that they were difficult (e.g. "I'm sorry this is a difficult question, but can you tell me..."). Improvements that can be seen in later interviews include leaving longer silences to give space for participants to speak; using probes more effectively to explore topics; and an improvement in the flow of interviews, with fewer leading questions.

The model with which I conceptualised patients with FMD changed subtly during data collection and analysis. Some preconceptions softened, which allowed me to let people express their own understanding with less influence from my own beliefs via leading questions and specifically focused questions. As an example, I initially made the assumption that all participants would be offended by psychological symptom explanations, which was not necessarily the case. With a softened stance, I allowed participants to tell their own story, rather than shifting the topic of conversation to what I perceived to be important. There were incidental consequences of the longitudinal research design. The three interviews over six months allowed development of rapport and trust. I found that some participants would reveal more intimate information in subsequent interviews, such as personal relationship issues, mental health history and difficult experiences from their past.

8.2.6 Analysis

Data were analysed using inductive thematic analysis. The approach was informed by the process described by Braun and Clarke (2006),¹⁹⁹ which identifies six phases of analysis: (i) familiarisation with the data, (ii) generating initial codes, (iii) searching for themes, (iv) reviewing themes, (v) defining and naming themes, and (vi) producing a report. Thematic analysis was chosen as the method of analysis for several reasons. First, it can be considered a foundation method of analysis, suitable for novice qualitative researchers.¹⁹⁹ Second, thematic analysis is flexible, in that it is free from theoretical commitments, unlike other specific methods of analysis.¹⁹⁹ Finally, I wanted to perform an inductive analysis, where themes are derived directly from the data, rather than from interview questions or my preconceptions.

Analysis was concurrent with data collection, allowing the interview topic guide to be updated iteratively in order to explore emerging themes. Familiarisation with the data started during transcription; this allowed time to think about the data and consider any changes in the patients' perspectives across the time points. Each interview transcript was then read, re-read, highlighted and annotated in a process of familiarisation with the data. Selected interviews were discussed in detail with the analysis team (Fiona Stevenson, Medical Sociologist and Marta Buszewicz, Academic

GP and Clinical Trialist), which met at regular intervals throughout the data collection and analysis period. Topics and themes from each interview were identified and discussed.

After I had transcribed, annotated and highlighted all the interviews, I collated a comprehensive list of topics from all the transcripts. This list was refined by consensus within the team and then used to create a coding framework (see Appendix 11 page 250, for the list of codes). Each transcript was then coded according to the coding framework with the aid of the computer software, NVIVO for Windows, version 10. Data for each code was retrieved and subjected to further analysis. The data was considered within cases, where the unit of analysis was the participant, and across cases, where the unit of analysis was interview time point. The research team agreed that the findings from an across case analysis was the better way to address the research questions. Themes and subthemes for each time point (baseline, post-treatment and follow up) were identified from the coded data. The original transcripts were then reviewed to ensure the themes were an authentic representation of the data and to seek disconfirming data. Un-coded text was examined for disconfirming evidence.

The first draft of the manuscript was then written, with each time point considered separately. The final list of themes at each time point was presented initially without quotations in order to develop arguments. This draft was then discussed and refined by the research team and quotations to support the arguments were then sought from the interview transcripts. This gave an additional check that the themes were grounded in the data.

8.2.7 Evolution of the Topic Guide

The topic guide was updated iteratively based on emerging themes. Some participants' accounts tended towards the factual, with little in the way of expression of feelings. To address this issue I made a note to ask for anecdotes, such as "Can you give me an example of..." or "Can you tell me about a time when...".

Participants' views about being given psychological explanations and attitudes towards mental health problems emerged as an important theme early in the data collection period. I started to explore these issues in more depth and returned to this topic in the follow up interviews to look for any changes in perceptions. I also added questions about close relationships, as it became clear that family and friends were an important part of the participants' narrative. Finally, in order to encourage constructive feedback I started to ask participants how could we have made the programme more helpful and asked what advice they would give others in similar situations. As I became more familiar with the topic guide I was less inclined to follow it in a linear fashion. See Appendix 1 (page 251) for interview topic guide.

8.3 Sample Description

From a pool of 29 participants who had been allocated to the intervention condition of the feasibility study, 11 fitting the purposive criteria were invited to participate in the qualitative study. All invited participants agreed to take part and there were no dropouts or missing data.

Clinical and demographic characteristics are presented in Table 8.2, where participants are identified by assigned pseudonyms. The sample is representative of the larger feasibility trial sample and similar to previous descriptions of this population.³ The average age was 44 (range 21-67), 82% were female and the average symptom duration was 6 years (range 1 to 30 years). There was a mixture of the most common symptom phenotypes.

Table 8.2. Participant clinical and demographic characteristics

Pseudonym	Sex	Age	Symptom Duration (years)	Symptom
Amy	F	43	2	Head & upper limb tremor
Michael	M	46	2	Mixed movement disorder
Julie	F	50	6	Upper limb tremor & gait
Lynn	F	56	4	Gait disturbance
James	M	36	1	Mixed movement disorder
Mary	F	67	10	Head tremor
Nicole	F	45	4	Gait disturbance
Deborah	F	58	5	Mixed movement disorder
Sarah	F	21	1	Left sided weakness
Megan	F	22	1	Weakness
Lisa	F	43	30	Weakness

Table 8.3 presents target versus actual recruitment for the purposive sampling criteria. The recruitment targets for each purposive sampling criterion were met, except for the number of males and the age bracket 60+. Two out of a planned three men were recruited and there was only one out of a planned two participants in the older age bracket of 60+. The failure to meet these sampling criteria related to the limited number of males and participants aged over 60 recruited and randomised to the intervention group.

Table 8.3. Purposive Sampling Criteria Targets (participants fulfil multiple criteria).

Criteria		Aimed minimum number to recruit	Actual number of subjects recruited with criterion
Symptom	Tremor	1	3
	Gait Disturbance	1	3
	Mixed Movement Disorder	1	4
	Weakness	1	1
Symptom duration	Less than 18 months	2	3
	18 months to 5 years	2	5
	More than 5 years	2	3
Age bracket	18-29	2	2
	30-59	2	8
	60+	2	1
Gender	Male	3	2
	Female	5	9

Thirty-three Interviews were conducted and analysed (three per participant), ranging in length from 36 to 71 minutes. The findings from each interview time point will be explored in three separate chapters, Baseline, Post-Treatment and Follow Up. A summary of central themes across all time points is presented at the end of Chapter 11, after the findings from interviews conducted at six month follow up. Quotations supporting the findings were found by reviewing the coded transcripts using the software NVivo. When it was relevant to the quotation, the interview question was included and prefaced with the label "I:". The participants' quotes are prefaced with the label "R:" for respondent. I aimed to provide a spread of quotations from all participants. Each quote ends with the participant's pseudonym and age in parentheses.

Chapter 9 Qualitative Findings: Prior to Treatment

9.1 Introduction

This chapter will explore themes from the baseline qualitative interviews, conducted prior to commencing the study intervention. Several weeks before the interview, the participants had attended an initial consultation with the study neurologist (Mark Edwards). During this consultation they were given the diagnosis of FMD which was explained according to the study protocol. The explanation emphasised the importance of self-focused attention as part of the mechanism driving symptoms and described psychological issues as part of the problem for some patients but not necessarily everyone. Many of the participants had been diagnosed with FMD prior to their initial consultation with the study neurologist, although often a different diagnostic label was used, such as Conversion Disorder.

The key themes that emerged from the baseline interviews stem from the participants' lack of understanding of their problem and their perception that health care professionals (HCPs) also did not understand their problem. The themes are explored under the following sections: Symptom Onset, Early Interactions with Health Care Professionals, Resistance to Psychological Explanations, Feeling Unconvinced by the Diagnosis, Conflict and Marginalisation, the Impact of their Symptoms, Relationships, Isolation, Symptoms Remain Mysterious and Powerlessness.

9.2 The onset of FMD

Seven (of the eleven) study participants associated the onset of their motor symptoms with a sudden injury, illness or biological event. This event was often perceived as a frightening life threatening emergency.

Respondent (R): I was just cooking some dinner. Making some scrambled egg and I went all sort of tingly and on my tongue and down the side of my face. And my hand had gone all like [short pause] tingly. And I sort of panicked because working in healthcare I was thinking I was having a stroke or something. And I was really panicked about it and my mum called 111, which is the out of hours and they sent a paramedic around. And they took me to the hospital. [Sarah, age 21]

The remaining four participants described a slower more insidious onset of their troubling physical symptoms. Consultations with doctors had led them to believe that their motor symptoms were associated with neurological disease; specifically multiple sclerosis (Nicole), Parkinson's disease (Julie), scarring seen on her brain MRI (Mary), and some as yet undiagnosed neurological problem (Michael).

R: So I was dragging my leg. And then from then, it's just been a series of tests, different consultants to get to the point where I was then diagnosed with possible MS. Through MRI scans, but the leg slowly got worse. [Nicole, age 45]

Physical or biological precipitating incidents or an initial causal attribution of symptoms to neurological disease were central to all the participants' illness narratives.

9.3 Early Interactions with Health Care Professionals

The onset of motor symptoms led the participants to a series of interactions with HCPs, often initially in the context of a perceived medical emergency. These contacts usually became drawn out over a long period of time, while waiting for investigations and waiting to be seen by specialist doctors.

9.3.1 Nobody knows what's wrong

The perception of most of the participants was that their doctors did not know what was wrong. This belief was reinforced by the commonly reported experience of undergoing multiple investigations that failed to identify the cause for their symptoms (negative test results). Rather than being reassuring, negative tests were often interpreted as the unhelpful and sometimes frightening news that the cause of their symptoms remained unknown.

R: Because I went for the DAT scan, then I went to see the consultant. And he showed me the brain results on his screen and told me what the normal levels should be. And said well you don't have Parkinson's disease, but I don't know what it is that's wrong... and it was, and then he said you don't look very happy. But it was plunging at the unknown then as I hadn't a clue what the diagnosis was. [Julie, age 50]

Other events that led participants to believe that nobody understood their problem were successive referrals to different medical specialists that often did not result in a conclusive diagnosis; conflicting information from different doctors; and prescribed treatments or advice that were considered in retrospect to be unhelpful or harmful.

9.3.2 Harmful treatments

Six out of the 11 participants reported that they had been given inappropriate treatment that they believed made their problem worse. These treatments were: treatment with anti-Parkinson's disease medication (Julie, see quote below); advice to use alcohol to control a tremor (Mary, see quote below); provision of splints without follow up (Lisa, see quote below); prescription of escalating doses of benzodiazepine medication (Michael); functional electrical stimulation (FES) to aid gait (Nicole); and anti-sickness medication that caused an acute dystonic reaction (Deborah).

R: I was given Parkinson's medication for two years... And then the medication was making the symptoms worse... I can only attribute it to the Parkinson's medication because it wasn't as bad as that. I had a tremor in my hand at the start of it all and by the end of taking Parkinson's medication, I've been left with weakness in my right side. [Julie, age 50]

R: Because I was taking, one of the consultants recommended that I took a glass of wine every evening. And it did relax me instantly. And even that if I took a glass of wine it would stop it, you know, for a while. But of course once you start you need more and more. Your body becomes reliant on it and I'm afraid I did become an alcoholic. [Mary, age 67]

R: So they felt that a [splint] would be more appropriate and I was advised not to walk without it. And I never questioned it and I think there was supposed to be a time where I didn't use that. But I was never. I never questioned it or pushed it and I was never advised not to wear it. I've always linked, if I need to walk, I need to wear my brace. Um and still, um 10 years down the line, that's how I still see things. So I never really thought about anything else. [Lisa, age 43]

9.4 Resistance to Psychological Explanations

Eventually all participants were given a psychological explanation for their symptoms, which left them feeling dismissed and disbelieved and some felt ashamed. There

appeared to be several issues associated with the general resistance to or rejection of psychological explanations.

9.4.1 Resistance to psychological explanations was associated with...

Psychological explanations were at odds with the participants' physical experiences. This included perceived physical precipitating events, associations with neurological disease (some had been given diagnoses of multiple sclerosis or Parkinson's disease before being told their problem was functional), and the physical impact of their symptoms.

R: And one doctor sort of just said to me, like, "Oh, it's all in your head". And I was a bit confused because, well, it's swollen [my arm], it's not in my head. I am seeing that. I was sort of like, are you seeing that or not? [Sarah, age 21]

Psychological explanations were often interpreted as meaning that there was nothing wrong with them, with the implication that the patient was imagining the problem, exaggerating their symptoms or faking it.

R: Because that's what it feels like, psychological feels like it should mean, it's literally you are making it up. It's all in your head, there's nothing wrong with you at all. [Megan, age 22]

Participants understanding of psychological problems did not fit with their personal illness narrative or perception of self. It was common for the participants to associate psychological problems with pejorative negative stereotypes. For example: psychological problems are trivial and the person would get better if they really wanted to; psychological problems are a character flaw and the fault lies with the patient; or psychological problems were either trivial or signs of severe mental illness. The participants' accounts often distanced themselves from negative stereotypes.

R: They said it might be psychological. I thought, well, surely I would have beaten it, that, because, you know, I'm determined to get better, you know. [Mary, age 67]

R: It makes you feel um that you are a mass murderer. Do you know? When someone says psych you think Norman Bates, do you know what I mean? Something bad. You're a bad person because you've got a psychiatric problem. ...

You automatically think they're a nut-job, they must have bumped their wife off.
[James, age 36]

R: Um, well, before all this happened I was like, I'd been on holiday with my friends. I'd been to Ibiza, which was really good and... I don't think there was anything really that could of caused any stress really... And I've never sort of been one to like, I don't know, stress about things. [Sarah, age 21]

Some participants implied that their resistance to psychological explanations related to the danger and undesirability of the corresponding psychological treatments. One participant described a frightening experience of being admitted to a locked psychiatry ward. Another described how he was denied active treatment and prescribed escalating doses of benzodiazepines.

R: So then I had investigations in, I self-admitted, well I agreed to an admission into a psychiatric unit for 3 days, which scared the living daylights out of me... I was called in front of a panel of psychiatrists and after 3 days there were 7 people in the room, I was absolutely terrified. I was 17 and I was, err, in with some quite, quite dangerous patients, I was with some paranoid schizophrenics, yeah it was quite a secure wing. [Lisa, age 43]

R: Because they have absolutely no experience of my condition at all. All it was, was keep on taking medication [diazepam] and we'll see you in 6 months. That's great, increase my misery for 6 months and do nothing about it. Hopeless.
[Michael, age 46]

Finally, there appeared to be resistance to psychological explanations that was associated with a perceived negative attitude of HCPs towards problems associated with psychological factors.

R: Yeah, he said you haven't got a, you haven't got a brain tumour and you haven't got cancer, I've got other patients. Like, he said like, because I didn't have cancer he didn't want to help me. And he, he literally like rolled me around like a bit of meat when he... put the pin up your foot... [Sarah, age 21]

R: So I was always led to feel almost, ah, I don't want to, I don't know embarrassed but, quite shamed, in that that was the reason. That's how I always felt, in that I was contributing or a contributory to my condition. Um, without anyone actually coming out and saying that, that was kind of how I was always left to feel. [Lisa, age 43]

9.4.2 Psychological problems were often considered as separate or secondary

There was a subgroup of participants, who were resistant to psychological explanations for their motor symptoms, but also acknowledged a past medical history of psychological problems. These participants generally considered that their psychological problems were separate and unrelated, or that problems such as anxiety or depression were secondary to the physical impact of FMD.

R: I've seen a psychologist for the pain, because that's part of the pain management. And there are issues, underlying issues, you know from getting over my dad's death and different things like that. But I think that's separate. I think this is something different. [Julie, age 50]

Often these individuals felt that the doctor had dismissed their physical symptoms due to their past medical history of psychological problems, preventing them from performing a thorough medical assessment. This left them with little faith in the diagnosis.

R: And I saw another consultant after that who more or less told me it was psychological. Um that because I had a history of, a history, I'd had stress and depression in the past. And he sort of honed in on that and because of that, it's just psychological your symptoms and we don't think that there's anything wrong. [Julie, age 50]

9.5 Feeling Unconvinced by the Diagnosis

Most of the participants had been given the diagnosis of FMD (some under a different name, such as Conversion Disorder) or a probable diagnosis of FMD before being referred to the study neurologist. They generally described feeling confused and unconvinced when they were first given the diagnosis. This was related to psychological explanations as described above, but biological explanations were also often met with confusion.

R: She said it's like software problem, she said and it's basically, say you are trying to tell that leg to do that, your other leg might go, you're focusing and it's not going where you need it to. And that was the easiest way she could describe it. But to me it sounded alien. It seemed like how could that be me? That can't be right. There's either something wrong or not. [James, age 36]

Sometimes feeling unconvinced about the diagnosis was related to a perception that the diagnosis was reached because no other problem could be found (based on negative tests or a diagnosis of exclusion).

R: Ahh I, I took it as bullshit really. I just thought, you're putting me in a, you can't find anything specifically wrong with me. My brain MRI is clear. There's no lesions showing on my brain. My spinal MRI is clear. All the other tests, the nerve conduction studies are clear. The EMG's are clear. So it has to be a functional neurological disorder. Because we can't find anything else wrong with you. So it has to be this. That was my interpretation. [Michael, age 46]

9.6 Conflict and Marginalisation

Anecdotes describing conflict with HCPs and perceived poor treatment featured prominently in many participant's narratives. These experiences had a powerful and lasting impact on participants, leaving many with a sense of indignity. It was common for participants to describe being left feeling that their problem was not worthy of help.

R: Part of me would love to put a complaint in and say, here just to let you know about the treatment I received under Dr so and so, I wouldn't like this to happen to anybody else. [Michael, age 46]

Participants often felt that they had been abandoned by doctors and let down by the health care system. Nettleton et al (2005)¹⁹⁵ used the term "medical orphans" to describe a similar situation in patients with unexplained neurological symptoms who felt they had been marginalised from medicine.

R: All it was, was keep on taking medication and we'll see you in 6 months. That's great, increase my misery for 6 months and do nothing about it. Hopeless. ...I tried chasing him for 3 days. And each time he ignored me. He ignored my phone call for help. [Michael, age 46]

R: So getting that diagnosis was a plus, but then it was also a negative because I didn't know what I was dealing with. Does that make sense? Because there's not enough, like some, if someone had a heart attack for instance, there's a set of actions to do, to help that person. But there didn't seem anything to sort of help me with my situation. [James, age 36]

In contrast, one participant described an overall positive experience of interacting with HCPs. She reported that everyone had tried their hardest to understand and help her. Most participants had encountered at least one clinician who they considered praiseworthy. The characteristics commonly associated with these clinicians were listening, believing, open mindedness, and being supportive.

R: I've got a very good doctor and he's been looking it up. And he's been very supportive. [Amy, age 43]

9.7 Impact of Symptoms

The negative impact of FMD on participants' everyday life was significant. A few described how they had adapted to minimise the limitations on their lives. More commonly however participants described how their symptoms limited their ability to work, fulfil their role as a parent, or perform activities of daily living such as washing and dressing. The impact described by each participant is summarised in a representative quote in Table 9.1.

Table 9.1. Participant's descriptions of the impact of FMD

Participant	Participant's description of the impact of FMD
Amy age 43	"Well it is quite debilitating. Um, it has a complete impact on, on your life. You have to, reassess what's important to you and what's not. And you know, to the point of, um... I've had to let people look after my children when I am usually primary carer. I'm a very hands on mum, and I've had to let people take over. So it's been quite hard to ask people to help." [Head & upper limb tremor]
Michael Age 46	"I've been off [work] for the last 13 months. I've been registered disabled. I have, it has drastically affected my life over the last, that was January so over the last 13-14 months. I've lost virtually a year of my life, because of my condition." [Mixed movement disorder with jerks]
Julie Age 50	"Because of the pain, I'm up most of the night with the pain, at most I have 2 hours, 2 and half hours of sleep a night. And I wake up and I'm really tired and mornings I just feel... I don't know, poorly is the only way I can describe it. As if I could, I could just stay in bed and just sleep. I don't because I push myself to, but I just, I just feel really under the weather, it's as if I can't function sometimes." [Tremor and functional gait disorder]

Lynn age 56	“It’s just that I’m an active person and it’s so frustrating sitting there. I want to do it. My brain wants to do it but my body doesn’t want to do it. That’s it really, it’s just so frustrating. Just sitting there. It’s gone on for a long time now. It’s slowly getting to the stage where I’m accepting that I can’t do it.” [Functional gait disorder]
James age 36	“I just feel like I’m 36 and I’ve got the needs of a 5 year old. Like having to have help to do stuff, having to have someone to get me something. Not being able to drive. And, and things like that, it just feels like I’m cut off and I can’t do nothing about it.” [Mixed movement disorder]
Mary age 67	“Well, it’s constant. And people say, oh, it’s hardly noticeable. But to me, it’s day and night, you know. Even if I wake up in the night it still shakes. And um, well, my favourite hobby, reading. And I loved my books and especially autobiographies, my favourite author, believe it or not is Charles Dickens. Oh yes. And the best book is Barnaby Rudge. Of the lot. And, of course I can’t read now, because everything flickers, you know.” [Head tremor]
Nicole Age 45	“And that’s it and I just ended up, you know, you accept it and get on and just focus on the positives. But it is hard. Because, you know, we’re [my children and I] at the park and I want to be running around and getting involved with them, because I’m quite an active person. I want to play rounders with them, or kick a ball. It is hard.” [Functional gait disorder]
Deborah age 58	“I’ve always been terribly independent and I want to remain that way and I want my independence back. And I feel like this is starting to take it away from me really. Because I don’t want to be pushed around in a wheelchair and have somebody take me on the Ambi-lift and help me here there and everywhere else. I just want to get on and do it myself.” [Mixed movement disorder]
Sarah age 21	“I do get annoyed but. I just get frustrated because I, I get angry because like, you see other people doing things. Like a simple thing like going in to town and having a coffee. There’s so much you have to think about when I go into town and I have to make sure I’ve got my crutches, make sure I’ve got a bag that I can carry with my crutches. Make sure I haven’t parked too far away.” [Functional weakness]
Megan age 22	“[It affects me] quite a lot and it’s pretty horrible. It stops me doing a lot of things. And it does get me down quite a lot, not being able to work.” [Functional weakness]
Lisa age 43	“I think I’ve lived with it for such a long time that I don’t really, I mean, I’ve lived with this condition um longer than I’ve not. Um, and you know, I was 13 when I first started getting it. So I’ve accepted it as part of who I am. I don’t really know any different... And I worked quite hard to, not be as limited as much as possible. So I don’t feel I’m limited in any. I have a social life. A working life that I achieve, in spite of that. So it has limited in lots of ways but, yeah, I don’t believe it defines me.” [Functional weakness]

The experience of FMD was most commonly described as frustrating. Frustration was expressed in relation to an inability to control their movements and the inconvenience of having a disability. Other sources of frustration were being misunderstood, lack of support from the health service and the lack of answers. Frustration often developed into anger and distress.

R: It's just that I'm an active person and it's so frustrating sitting there. I want to do it. My brain wants to do it but my body doesn't want to do it. That's it really, it's just so frustrating. [Lynn, age 56]

R: I do get really down about it and I get very frustrated and very angry. And I try not to continually to go back to the past. So I try not to, to like say, you know to go back to why has this happened. [Julie, age 50]

In general, the experience of having FMD appeared to have a negative effect on the participants' sense of self-worth.

R: I just feel a bit useless. I feel sometimes, I just feel like I just want to get up and do stuff. And it's frustrating. It's frustrating to not be able to do anything about it. [Megan, age 22]

R: And it's just things like I'm back to being a kid again, do you know what I mean? And, that's how I feel, I feel like I'm a kid. [James, age 36]

9.7.1 Relationships

The support of family and friends was considered important in order to cope, although many disliked feeling dependent on others and some were concerned about becoming a burden.

R: I worry, well it doesn't worry me, I feel bad because I'm always asking people to do things. Because I can try and do what I can, but sometimes I just can't do it and I don't like to put loads of pressure on to people. [Sarah, age 21]

Some reported that their health problems had become a source of tension in their relationships.

R: It's, it's made a severe impact on the quality of the relationship I have with my wife. She gets very, very impatient at times. She gets, ahh. It's more annoyed and distressed I would say, more than anything else. My 15 year old at times would

like me to be able to do more things. ...a lot of the times I have to say I'm sorry girls I'm just not well enough. I'm just not well enough today, I have to go to bed. [Michael, age 46]

R: Um, yeah I can get really snappy. I never used to be. So I can get really grumpy. Which is not very nice. I'm grumpy at my children quite a bit. They will tell you. [Amy, age 43]

Participants often felt that family and friends were unable to understand or relate to their experience. They perceived that others questioned whether their problem was genuine or that they underestimated the severity and impact of their problem. Some related the lack of understanding from others to the invisibility of the pain and fatigue, which most experienced in addition to their motor symptoms.

R: And this is really, really frustrating. Because people, because I look perfectly ok and people comment, "Oh you look well" and yeah. I can't explain to people. I can't do it. And I think they think that I'm telling little porkies. [Lynn, age 56]

9.7.2 Isolation

Most participants described feeling socially isolated. This was especially a problem for those who had left work or education due to ill health. Isolation was associated with difficulty accessing some environments, withdrawing from social activity due to pain or fatigue, and avoiding going out in public due to the embarrassment caused by their symptoms attracting unwanted attention. Some participants described feeling lonely, as they experienced a gradual loss of contact with their friendship network.

R: Friends, as I've said, I've cut a lot of them off. Not in a nasty way. I've just said when I'm feeling better, when I'm feeling better. So as time goes by, out of sight, out of mind. So it's like they think, oh he'll get in touch when he's ready, so you hear less and less and less from people. Which makes it worse, because you normally get a random text and that, and now you get nothing. [James, age 36]

9.8 Symptoms Remain Mysterious and the Uncertainty is Frightening

Despite an average FMD duration of five years in this cohort and multiple consultations with HCPs, participants generally reported that their symptoms were mysterious and the cause unknown. The uncertainty of the diagnosis and prognosis was a common source of concern.

R: It's not knowing of whether you're going to get better or not. It's, it's not in the back of your mind it's in the front of your mind. [Michael, age 46]

Waiting while not knowing what's wrong was often distressing, leaving many with a heightened sense of powerlessness.

R: [The podiatrist said] A second opinion is what you need. And then it's back to the doctors and back to the doctors and then you're waiting and waiting and waiting and I'm just sitting at home.

I: What effect did the waiting have?

R: Oh dear, very, very, I'm going to get upset now,.. Very depressing [voice breaks] it really, really was depressing. Because I can't do anything. [Lynn, age 56]

9.9 Powerlessness

It was common for participants to describe feeling stuck because they did not know what was wrong and therefore did not know how to help themselves. This, together with feeling abandoned by the health care system, appeared to leave many feeling powerless and unable to move forward in their lives.

I: So what does it feel like to have a bad day?

R: I just can't control my head or my hands or my legs at all. I'm trying to tell them to stop. They won't stop. Um, it gives me headaches. I get a lot of pain, I end up going to bed early. [Amy, age 43]

9.10 Summary

This chapter explored themes arising from an inductive analysis of interviews with patients diagnosed with FMD, prior to receiving the study intervention. Participants commonly associated the onset of their symptoms with a preceding injury or illness. Their problem led them to a series of encounters with medical professionals which were usually perceived as having been unproductive. There was a common perception that HCPs did not know what was wrong. Virtually all the participants had been given psychological explanations for their symptoms which left them feeling disbelieved and dismissed. Reports of conflict with HCPs were common. FMD had a significant impact on the participants' lives, causing disability and distress and many felt isolated and

abandoned. A perceived lack of understanding from both professionals and their social circle left them feeling powerless to help themselves.

Chapter 10 Qualitative Findings: Post-treatment

10.1 Introduction

The previous chapter explored participants' experiences up until their engagement with the study intervention. In this chapter I will explore themes identified in the interviews conducted immediately after completing the five day intervention.

The intervention participants completed 8 sessions of physiotherapy over five consecutive days. The first session of the intervention was a joint consultation with the study neurologist (Mark Edwards) and treating physiotherapist (Kate Holt). The diagnostic explanation previously given during the initial neurology consultation was reiterated. The study intervention was introduced to the participant as a specific treatment for FMD. Optimism was expressed that the patient would see an improvement in their symptoms by the end of the week, but it was stated that the main objective was to learn how to manage their symptoms in the long term. All subsequent sessions were run by the physiotherapist. The second session elaborated on the explanation for FMD. Using the physical biased aetiological model described in the previous chapters, the aim was for the participant and physiotherapist to come to a shared understanding of the participant's problem. Education continued during the subsequent sessions but the focus shifted towards movement retraining using strategies to divert the focus of motor attention. The intervention concluded with writing a long-term symptom management plan and completion of the final outcome measures.

Themes from post-treatment interviews are explored under the sections: Physical Changes, Ingredients of the Intervention, I Could See It Working, Understanding, Less Resistance to Considering Psychological Factors, Work in Progress, Hope, A Positive Experience and Outlier.

10.2 Physical Changes

All but one participant felt the intervention had helped their motor symptoms. There was considerable variation in the perceived extent of change; some described only modest improvements (e.g. James, see quote below) while others described significant changes such as no longer requiring walking aids (e.g. Sarah, see quote below) and one person reported complete resolution of their symptoms (Michael, see quote below).

R: I still got the shakes but the leg movement is a lot more advanced, as we've seen, it's a lot more advanced as to where it was at the beginning of the week. [James, age 36]

R: My walking is so much better now and I don't drag my foot along. I'm almost walking normal [laugh]. I think that will come with time. I can actually walk up the stairs now without lifting, like I had to lift it up with my hand before, whereas I can just lift it up now. [Sarah, age 21]

R: If I'd been told Monday that in the next couple of days, you're not going to have any symptoms, I would have found it hard to believe. But here we are, come Friday, I can sit, I can talk. There is normal muscle control. And it's fabulous. [Michael, age 46]

10.3 Ingredients of the Intervention

The intervention was highly praised by the participants. There were several elements that were commonly considered important ingredients in making the treatment effective.

10.3.1 *The Role of the clinician*

The clinicians were perceived as specialist and eminent. This contrasted with the participants' previous experiences, where it was commonly reported that HCPs had not been specialised enough and did not know what was wrong or how to help. Most participants' reported that the study clinicians had taken their concerns seriously and understood their problem. Some participants commented on the personal attributes of the physiotherapist that helped them develop rapport and trust. This included being calm, kind, non-judgemental and interested.

R: I actually feel that [the treatment team] might not be going through it but they actually understand what [I'm] going through because they've done the research, they've done, they've had the clinical trials and stuff. [James, age 36]

R: [The study physiotherapist] was really good to have that relationship [with]. Um and she's such a nice person. And to trust her. You know. And believe that what she was telling me really. [Deborah, age 58]

10.3.2 The Intervention was challenging

Often the first comment participants made about the intervention related to its challenging nature. Most reported feeling challenged physically, cognitively and emotionally, generally to a greater degree than had been expected.

R: Um, The week of physiotherapy was both mentally and physically exhausting. Especially mentally exhausting. It's been quite a journey. [Deborah, age 58]

R: Um, intense and emotional. Um, it really pushed my boundaries, made me re-evaluate behaviours... So it's been very intense but it's just amazing. [Lisa, age 43]

For those who found the intervention cognitively demanding, this was related to learning and understanding about their symptoms, as well as the concentration required during movement retraining. Some noted the apparent paradox of having to concentrate to not think about their movement (attend to their body).

R: ...I'm feeling mentally tired, I feel it's because I'm thinking, thinking more about not thinking [laughs]. Does that make sense? My focus is obviously on what is going wrong with me and whatever, so I'm trying to retrain myself not to think about it so much. ...There's so much to think about when I'm trying to manoeuvre around. I think that's where I am feeling tired. I'm just thinking constantly about what I'm doing. [Nicole, age 45]

The physical challenges usually related to the intensity with which treatment sessions were scheduled over five consecutive days. This is discussed below.

10.3.3 Treatment intensity

The intensity (frequency) of treatment sessions was considered an important element for success by most. Some participants described how consecutive days of treatment made progress more noticeable and allowed progression without setbacks. Some

contrasted this to their previous experiences of physiotherapy, where sessions had been intermittent and considered less helpful.

R: And it's been continuous, whereas the physio I've had, I have some one week and then the next week, or two weeks later by which time you've forgotten what has happened. But I think it's because it's been five days continuous that I can see an improvement, whereas before I couldn't. [Julie, age 50]

R: I think having it every day really helps. Because I think if I had sort of come for one day, then gone home for a month, then come back again, I don't think that would have been beneficial really. I think having it in a short space of time really helps. [Sarah, age 21]

While the high intensity was generally considered useful, some participants also found this difficult to cope with.

R: I think 5 days was a killer. Um, maybe it could be split, the first week is two sessions and then the second week three sessions and aim to build. I don't know. I don't know if that would work. I'm not sure. [James, age 36]

10.3.4 Coming away from home

In addition to the high intensity of the intervention, coming away from home was reported by several participants to be helpful by preventing interference from normal routines. All were provided with hotel accommodation during the five day intervention, which may also have impacted on how they perceived the treatment.

R: I think it was more about, the good thing about coming away from home was that I was, and not stay in hospital but I was then able to go out and practice. And there wasn't any normal life like interrupting. [Megan, age 22]

However, one participant found staying away from home difficult and reported that this hindered her ability to engage with the intervention.

R: That's the state I was in. I did not want to go through with all this. I just wanted to go home. Get back to my um, comfort zone, normal area. I don't know. [Lynn, age 56]

10.3.5 Focus on education and learning

Participants often commented that the study intervention was different from their previous experiences with physiotherapy because of the emphasis on learning and developing an understanding of their symptoms. Some described how they were intellectually engaged in the treatment process, which included learning the theoretical rationale behind the treatment strategies.

I: And how was it different from your previous experiences with physiotherapy?

R: I had one session and it was very much focused on physical. There was no kind of background to it and what was actually happening. And it was all very focussed on trying to get that one, that movement to happen. And, whereas this was kind of, the link between the kind of theoretical side and then diverting attention a little bit, but in a way where you can still work on things. And, and not slip back into like old movement patterns. That was completely different. [Megan, age 22]

10.3.6 Structure and flexibility

A few participants commented that the intervention was well structured, but importantly it also allowed for some flexibility to meet their needs. This structure gave James the impression of progress, he noted that structure was missing from previous experiences of physiotherapy.

R: it's quite well structured, which I weren't expecting. I was expecting it to be a bit hit and miss. It's not regimental, it's flexible, but it wasn't hit and miss. I was expecting it to be hit and miss because of past experiences of how things happen in hospitals, like timing and stuff and um, having clear goals each session, where in other places it's been, well we've done that today and then you go and repeat it again tomorrow and then you repeat it, and it's not moving forward, it's just like repeating. Whereas this week I've found that it's been quite diverse but also quite structured as well, because we go back but not constantly go back. [James, age 36]

10.3.7 Potential problems and barriers

Participants were generally reluctant or unable to provide much criticism of the study intervention. As previously discussed, the high intensity of treatment and coming away from home to stay in a hotel were considered helpful for most, but a potential barrier to progress for a small minority.

Awareness that the intervention was part of a clinical trial may have influenced the participants' perception of its value and how they reported their outcome.

R: The symptom model [was the most helpful part of treatment], you can't find that anywhere else. Of course you can't because this is a clinical trial. [Michael, age 46]

I: Do you have any other comments or thoughts?

R: Um, not really, I just, it's refreshing, that's the word, it's refreshing to know that there's, there is a treatment out there. Whether at the moment it is still early days and still being trialled, but there is a way out. [James, age 36]

10.4 I Could See It Working

It was common for participants to report that the movement retraining strategies had an immediate impact on their symptoms.

R: ...now when people give you exercises usually, you think oh gosh, I've got to do them, I've got to do them. But when I started doing them I could feel the difference. The physical difference, you know? The head wasn't shaking so badly. [Mary, age 67]

The use of a video and mirrors during physiotherapy sessions enabled participants to see their movement normalise while using the treatment strategies. This appeared to help motivate participants to engage with the intervention.

R: Um, and I get this child like delight when I see the flickers of the muscles working and everything going. It's just, I'm like a kid. [Lisa, age 43]

The perception that the treatment strategies worked and worked immediately seemed to be an important element of the intervention. It challenged participants' beliefs about paralysis or their inability to move normally. This in turn seemed to help them learn to trust their body.

R: Because when I saw, you know I could see in the mirror that there were flickers of movement [in the muscles of my ankle]. And I think if I hadn't had seen that, I wouldn't have trusted. [Lisa, age 43]

10.5 Understanding

Participants reported that the intervention had helped them to understand their problem and this was considered the most valuable intervention outcome by many participants. The value placed on having some understanding corresponds with the participants' focus on the lack of understanding and distress associated with uncertainty that characterised the baseline interviews.

10.5.1 Understanding was multifactorial, with a biological focus

The biological or physical events that dominated most participants' illness narratives in baseline interviews remained important in their understanding, but there was a shift towards recognising that other factors were part of their problem.

I: Can you talk to me about what you think caused the problem in the first place.

R: After this week with [the study physiotherapist], I think there's a few factors, um ... [Nicole, age 45]

In the above quote, Nicole goes on to describe in detail several potential contributing factors which included "possible MS" (confirmed by neuroimaging), a situation where she was stuck in a car unable to move her leg which was anxiety provoking, and her perfectionist personality trait. Another example of a multifactorial formulation is given by Julie below. It is less explicitly biopsychosocial in scope, however it is not singularly biological or structurally focussed, nor is it characterised by mystery as was the case with most participants' understanding at baseline.

I: What do you think caused your symptoms in the first place?

R: After looking at the process this week, I think it's a combination of the, it started when I had the occlusion in my eye, that's when the first tremor started and then, I had quite a bit of illness. Um, and then I was diagnosed with Parkinson's disease and given medication and the symptoms got worse. So I think it's a culmination of all of that. Now having an understanding of functional movement symptoms, um, I think it's just been quite a collective reason um, illness and just circumstance. [Julie, age 50]

When asked to describe their understanding of their problem, most participants articulated that they understood that it was not caused by a structural lesion or a

degenerative process and most were able to suggest several factors that could have contributed to its development. These changes in conceptualisation could be considered as a move towards a biopsychosocial understanding of their illness.²⁰² This differed from the baseline interviews where symptoms were generally seen as mysterious and potentially sinister, and the onus was on the physician to solve the problem.

10.5.2 Understanding provided legitimacy

The intervention, learning that FMD was common and having a perceived improved understanding of their problem appeared to provide legitimacy, validating the participants' illness experiences.

R: I've learned about the disorder, about the symptoms. I know that they are real now. And I know there are other people in the same situation as me. Which I didn't, I thought I was alone in this. So it's been a real enlightening experience. [Julie, age 50]

10.5.3 Understanding reduced threat value of symptoms

In the baseline interviews, several participants described feeling unsettled by or fearful of the uncertainty of their diagnosis and prognosis. In contrast, in post-treatment interviews participants often described feeling relieved and reassured, suggesting the experience of their functional symptoms felt less threatening.

R: Um, I think now I understand it more, I can accept it and it doesn't feel too bad. It's a bit scary that there's something sort of not right in your brain, but I think like there's a lot more things that are worse and it's good because it can get better. And it's not going to like, even though it probably will be quite difficult, but I think I'll be able to manage it more and I know how to deal with it now. [Sarah, age 21]

I: Do you think you will be better equipped to deal with a dip?

R: Yes. I think it's taking it slower. When the tremor starts or when my speech starts, instead of getting into a panic and trying to undo things, it's to take time to think it through... But I think it makes a big difference when you know why something is doing what it's doing. As opposed to being in the dark and now knowing why it's doing that. [Julie, age 50]

10.5.4 Understanding allowed improvement

When asked to comment on why they thought their symptoms had improved, most participants attributed changes to their improved understanding of their disorder. Participants described how their new understanding gave them more control over their symptoms and that it enabled them to become active in their rehabilitation.

I: What do you think, do you have a sense of what's changed to allow these things to improve?

R: I've realised I understand it more and I'm giving my body the opportunity. [Megan, age 22]

R: I don't think it is possible for the muscles to have changed so significantly in a couple of days. I think it's definitely in my understanding in my um, my trust, my confidence in my ankle. [Lisa, age 43]

10.5.5 Understanding was empowering

Together with the perception of improved understanding, there was an apparent shift from the sense of helplessness and powerlessness prevalent in the baseline interviews. Some participants described a new sense of self-reliance and an ability to help themselves.

I: What would it mean if the spasms come back?

R: I work harder trying to beat them. Because I think, being here I have the, I have the skills and knowledge, I have the skills base now and the knowledge to understand what is going on in your body. And the techniques to, counteract this. And get control back. And regain control and maintain control. [Michael, age 46]

10.6 Less Resistance to Considering Psychological Factors

In the baseline interviews, there was resistance towards acknowledging a potential role for psychological factors as part of the problem. This persisted in some participants, but for most there was less expressed resistance to the notion that psychological factors were part of the problem. An example of the softening stance in considering a role for psychological factors can be seen with Michael. In the baseline

interview he stated several times that he did not think that psychological factors were relevant to his problem:

I: How would you describe the relationship between psychology and functional symptoms?

R: There may be a relationship. I can't see it in my case. [Michael, Baseline Interview, age 46]

Post-treatment, he appeared to be less sure.

I: So what do you think caused them in the first place?

R: I don't know. I don't know. It may have been just an accumulation of stress at work, stress at home. I have 3 kids. Was it an accumulation of stress that set it all rolling? It might be. It might have absolutely nothing to do with it at all. It's hard to tell. [Michael, Post-Treatment Interview, age 46]

Several participants described coming to a new realisation that they often felt anxious and that this was part of their problem.

I: And can you speak a bit about, um, the role of mood or anxiety, how it affects symptoms.

R: Yeah. If I feel stressed, then my symptoms do get a lot worse. Sometimes quite uncontrollable. So I, try to become more aware. Um, it's, this week has been of benefit because it has pointed out that. The anxiety. You just don't think about it do you? [Amy, age 43]

R: Yeah, I've definitely learnt a bit this week about it as well, and how I've been. Um, and it's around people, like up in the gym we've been doing some... good walking, but then someone would cross my pathway or I'd be conscious of somebody there and it would all go wrong. Because I felt, I hadn't maybe realised it as much before but I was anxious about it. [Nicole, age 45]

Despite this softening stance, all the participants tended to hold the view that psychological factors, if present, were usually secondary to their motor symptoms or a separate issue and that they were not the cause of their FMD.

10.7 A Work In Progress

Only one participant considered that their movement problem had completely resolved, the remainder considered that they had benefited from the intervention but

that there was more work to be done. Treatment and rehabilitation was seen as a work in progress. They referred to newly acquired knowledge, movement strategies or a treatment plan that would enable them to progress their rehabilitation after discharge. However, they often reported expecting this to be difficult.

R: Yeah, it will take time. It's like everything, with physiotherapy it takes time you need to work at it. I didn't expect a quick fix, I knew I would have to put in a lot of work. Um so, I'm prepared to do a lot of work at home. [Amy, age 43]

10.8 Hope

The improvement that participants experienced during the intervention appeared to give them hope. Most described feeling cautiously optimistic about the future and hopeful for further progress.

R: I still got the shakes but the leg movement is a lot more advanced, as we've seen, it's a lot more advanced as to where it was as the beginning of the week. So I feel there is hope now, where at the beginning of the week I thought that's it. There's no hope. [James, age 36]

R: And so I can just see a light at the end of the tunnel that wasn't there. You know, I just, you know, I believe in it. [Deborah, age 58]

10.9 A Positive Experience

The participants considered that the intervention had been worthwhile and was generally experienced as positive and uplifting. This seemed to be related to it providing legitimacy to their illness experience, introducing a belief and optimism that their symptoms could improve in the future and some resolution of the uncertainty about their problem, which had weighed heavily on their minds prior to the intervention. Their positive experience contrasted sharply with their previous interactions with health care professionals, as described in the previous chapter, which had been mainly negative and unproductive.

I: Do you have any thoughts or comments to finish up?

R: No, just really this week has been brilliant for me. Amazing. Because I really do feel, after this week, that's what's wrong. You know, and it's like, yes [deep sigh]. [Nicole, age 45]

10.10 Outlier

There was one clear outlier amongst the participants, who presented an account that at times was contradictory. She suggested that the intervention had not made any difference to her motor symptoms and that it could not help her because nobody had understood the cause of her movement problems. However, like some of the other participants, she stated that the intervention had helped her come to the realisation that she was anxious. In addition she described renewed confidence that her problem with balance and falls would improve with perseverance and practice.

I: Was there anything about this week that was useful for you?

R: Understanding. Understanding what's happening. And understanding, I didn't think I was an anxious person. But I think this balance has made me very anxious. And I think that's when I get really anxious and I don't want do anything. I just don't feel I can do it. [Lynn, age 56]

R: But I don't, at the beginning of this I didn't know why [this problem started]. What, what built up to it? Why? You know? Out of the blue. If I had fallen, broken my arm or my leg. Then ok, then you, you've broken your leg and it's going to take this long. This has just come out of the blue. That, I find is really strange. Really hard. Because it's just, why? Why did it? Why did it do that? Why? [Lynn, age 56]

10.11 Summary

All the participants described having had at least some benefit from the intervention. Reported improvements in their motor symptoms ranged from modest changes to symptom resolution. They had experienced the intervention as “intense”, challenging them physically, cognitively and emotionally, which was unexpected. The participants identified several components of the intervention that they considered important and these were generally different from their previous experiences of physiotherapy. This included that it was delivered by specialist clinicians, the high treatment intensity, the focus on education and that movement retraining involved redirecting attention away from the body. Another important difference with this experience of treatment was that they could see the treatment strategies having an immediate impact on their symptoms, which left them feeling motivated and hopeful about the future. The participants generally considered the understanding that they had gained from the

intervention to be the most valuable intervention outcome and that this had enabled them to become active in their treatment. As part of their new understanding, most participants were less resistant to the idea that psychological factors were part of their problem. Most found the intervention to be an uplifting experience, in that it legitimised their illness experience, providing clarity to what was previously their 'mysterious illness'. One outlier felt that the intervention had not been helpful, which she related to nobody understanding what was wrong.

Chapter 11 Qualitative Findings: Six Months Follow-up

11.1 Introduction

In this chapter I will explore themes from interviews conducted at the six month follow up. The themes will be explored under the sections: Impact of the Intervention, Putting it into Practice, Perceived Role of Psychological Factors and Feedback. The chapter concludes with a table highlighting themes that were central to each interview time point (baseline, post treatment and follow up).

11.2 Impact of the Intervention

At six months, most of the participants reported that they thought they had benefited from the intervention. It appeared to have had a wide and varied impact on the participants' lives, which is discussed in detail below. It was quite difficult to disentangle physical, social or psychological changes, as well as the extent to which each individual had benefited.

11.2.1 Understanding

In the interviews conducted immediately after the intervention, most participants regarded the understanding they had gained about their problem to be the most valuable intervention outcome. In the six-month follow up interviews, improved understanding appeared to remain the most highly regarded outcome, although most commented that some things remained unexplained or confusing.

There was little change in the participants' conceptualisation of their movement problem from post-treatment to six months. In general they remained convinced that the diagnosis of FMD was correct. Most presented a belief that multiple factors had probably contributed to their problem. This often resembled a biopsychosocial formulation, although most considered biological events, such as a viral illness or injury, to be of primary relevance.

I: ...what do you think caused the problem in the first place?

R: I really believe that, I'd had bronchitis on and off for 2 years. So every 3 months it was the steroids or antibiotics. I was really run down. I believe that amalgamated in a like a semi-collapse [when] I was out one day with a friend. So I believe the two of that together sort of, obviously caused, a reaction in the brain. [Amy, age 43]

11.2.2 Hope

Participants continued to see their problem as one that could improve in the future. The hope and optimism that was prominent immediately post-treatment remained to some extent, but was now tempered with a realisation that future progress was likely to be slow and that their symptoms would persist for the foreseeable future.

I: So what are you expecting to happen over the next 6 to 12 months?

R: Um, if I carry on as I'm on, then hopefully more improvement and more improvement in my knee. And that's, you know, even a small amount. [Nicole, age 45]

11.2.3 Reduced threat

Immediately after the intervention, some participants, having gained a greater understanding of their problem, described feeling less concerned by their symptoms. This perception of reduced threat continued at six months, with several participants describing feeling more at ease with their problem. This change was associated with having an acceptable diagnostic label, a greater understanding of their problem and some resolution of the uncertainty of their prognosis. Several participants had expressed a concern in their baseline interviews that their problem would continue to progress causing increasing disability. Most now considered that their problem was one that could improve, and that they were better equipped to prevent deterioration.

R: Oh yes, you know, when you said what it was, it's a functional tremor, it's not dystonia, it's not Parkinson's. The relief! You know, from head to toe, of somebody telling me after 12 years of going to every doctor... Just knowing what it was, you know, the answer, after all those years. You know. That has made, well made me more relaxed. [Mary, age 67]

11.2.4 Self-Confidence

In the six month follow up interviews, positive outcomes were often associated with having greater self-confidence, particularly when out in public. Confidence was drawn from knowing what was wrong and having strategies to control their symptoms. Conversely, participants who had experienced falls or were concerned about falling often described a need to rebuild their self-confidence in order to progress.

I: What sorts of things can you tell me have changed and what things might not have changed?

R: Um, I think it helped, it's helped me feel a bit more confident, because I understand, sort of semi-understand what's going on now. And it's not me, you know, I'm not intentionally doing these kind of things. So that's been a real great benefit. It's sort of boosted me. I feel more comfortable in public. [Amy, age 43]

R: But I had a couple of falls, which knocked my confidence, just because I was trying to do things too quickly. So I have to really step back and go slowly, but, yeah it's, it's um. Yeah it's been a journey. [Lisa, age 43]

11.2.5 Physical Changes: Impairment, Activity and Participation

The actual changes to their motor symptoms and movement problems at six months varied considerably amongst participants. No one considered that their problem was worse compared to baseline, but one participant reported at six months that their problem had relapsed to baseline levels. Participants who reported ongoing benefits were generally evenly split between three groups; either reporting some setbacks after receiving the intervention but still better than baseline, feeling that their problem was stable, or reporting further improvement. As was the case in the post-treatment interviews, participants reported gradations of benefit, ranging from subtle changes to symptom resolution (one participant only, Michael).

Many described reduced physical disability, such as less reliance on mobility aids and greater ease with mobility and usual activities. One participant reported that he had returned to work with reduced hours after a long period of sickness absence, one had started paid employment for the first time and one had started a college course.

Some participants reported unexpected health benefits from the intervention. Mary had previously been told she was unable to have laser cataract surgery due to her head tremor, but this was now considered viable.

R: But my only problem now more than anything is cataracts... But I'm going on the 4th of November and they're going to try and laser that. They couldn't do it before because of the tremor ...she said, oh I think we can do it [now]. It only takes a few seconds and we'll try. [Mary, age 67]

Michael described how, with some difficulty, he was able to stop taking benzodiazepine medication, which he considered a significant achievement.

I: Could you start off by telling me how things have been over the last 6 months?

R: Since I was here in November, ah, all my spasms have ceased and stopped. At that stage I was on 4mg of clonazepam, on returning home I then cut it down by 1mg per week. Which meant come, I think it was the 20th of December I was drug free. No medication whatsoever. I'd say the down side of it all was, of the medication side was the symptoms, the withdrawal symptoms of clonazepam were horrendous. [Michael, age 46]

11.2.6 Relationships

Several participants noted that their overall improvement over the six months had been accompanied by positive changes in their relationships. Some described less tension and conflict in their family unit. For example, Michael described how his wife was less stressed now that he had a better sleeping pattern and was contributing to household duties and that he was better able to fulfil his role as a father to his young daughters.

I: My wife was very stressed and rightly so. Understandably so. She was getting very tired. I was up most nights, ah, during the day I was going to bed... and my wife coming home found very little done in the house. I just wasn't able to do it... It's, it's much less stressful now. Every house with 3 children has its associated stresses, but not the stress when one of the parents is very unwell. [Michael, age 46]

Some participants described how others had noted a change in their character, specifically being less irritable.

I: What about other people. Have they noticed any differences?

R: Yes my children have. They've noticed that sometimes my movement has better control. Um, they said sometimes I'm less grumpy. [laughs]. [Amy, age 43]

11.2.7 Validation

Validation of their illness was an important outcome immediately after treatment and this remained prominent at six months. Such validation justified dissatisfaction of previous interactions with HCPs (e.g. Sarah, see below). It also made one participant more willing to accept help from others (Deborah, see below), and helped another feel empowered to medically retire (Julie, see below).

I: Is there anything that everybody should know about functional neurological disorder?

R: Um, that they are real symptoms. Because when I was in [my local hospital] some people said to me I was making it up. Because nothing was showing up on my scans, which is fair enough, but I wouldn't make it up. And I think, they are real symptoms that people are having. I think people just need to be sort of like, educated on it because I think people don't understand it. [Sarah, age 21]

R: So it's always been, you know, I am not disabled. I am not disabled, I will drag that suitcase down the corridor, I am not disabled! And now it's like, it's just somebody helping me as you would help any other person. [Deborah, age 58]

R: But I think it's important because for me it was, "am I imagining this?" and because I've spoken to the likes of yourself and [the study neurologist] and the experts at home. I know I'm not imagining that it's there. [Julie, age 50]

11.2.8 Self-Reliance

Immediately after the intervention, it was common for participants to express an expectation of being able to manage their health more independently in the future, with less reliance on consultations with HCPs. In the follow up interviews, this sense of self-reliance continued in many participants. In practical terms, this was related to having an answer for what was wrong (so further investigations were unnecessary), knowing how to help themselves, and having an expectation of further recovery.

I: Do you think you need any other support or help for this to continue to get better?

R: Well, is there anything else? If I can keep doing these exercises as [the study physiotherapist] has taught me. And it is working. Surely if I keep on it will work more, you know?

I: Is there anything else that you think you might need other than physio?

R: Well is there anything else? No, I don't think so. [Mary, age 67]

This sense of self-reliance did not mean that participants felt they no longer required any contact with health services, but that they expected to use them less frequently and more efficiently. For example, participants often expressed relief at knowing they could contact the study team (neurologist and physiotherapist) should questions or new problems arise in the future, as they perceived the team understood their problem and could deal with it more effectively than their local services. This contrasted with their early difficult experiences of finding support in the NHS, which involved long waiting times and multiple consultations with different professionals which they generally had not found helpful.

11.2.9 Moving On

In the baseline interviews, there had been a prominent theme of feeling stuck and powerless to effect change, whilst immediately after the intervention, participants described a sense that they would now be able to move on. This continued at the six month follow-up interviews, where there was a sense amongst most participants that they had moved forward and were getting on with life despite continuing to experience symptoms.

I: Um and any thoughts about the next 6 to 12 months? About what you might think will happen?

R: ...I'd like to think that now I'm just going to start getting on with life... I just want to get on with life. [Lisa, age 43]

11.2.10 Outliers

There were two outliers amongst the participants. Lynn (age 56) was described as an outlier in the post-treatment interviews because she considered that the intervention had not been particularly helpful for her. At six months follow up, she continued to believe this was the case, despite also reporting a significant improvement to her gait

and balance problem. She attributed her improvement to the efforts she had made to practice walking, build confidence and confront her fear of falling. Her perception that the intervention did not help may have been related to an expectation of receiving a treatment that would result in an immediate improvement, and therefore she did not see the value in education and learning about self-management.

R: I don't know why I thought like that but I just thought it would be in a gym.... I thought I'd be doing exercises and things in the gym.

I: And how was it different?

R: Because I wasn't doing exercises in a gym, I had to challenge my working. I don't know why I thought it was going to be like that.

I: So what were you doing instead of exercise?

R: What do you mean? I thought I was going to be doing exercises, but we didn't. We, we, um, we talked about, how to side track my mind. My mind was thinking too much on what my legs are doing... [Lynn, age 56]

The other outlier, Julie (age 50), was an exceptional case because she was the only participant at six months to report that there had been no improvement in her symptoms compared with baseline. She had reported improvement immediately after the intervention but this had relapsed by six months. She attributed the relapse to an unfortunate series of health problems that included a fall resulting in a wrist fracture, an episode of acute labyrinthitis, exacerbation of long term chronic pain, and low mood associated with adjustments to her antidepressant medication. When asked if she considered there were any benefits from having had the intervention, she stated that she continued to use strategies that helped her balance and she was better at pacing her activity to manage fatigue. She also described how the intervention had helped her feel more accepting of her limitations.

I: I was wondering if you didn't have the labyrinthitis and you didn't fracture your wrist, I wonder how things would have been.

R: But the pain would have still been there.

I: So do you think you would have been about the same as you are now?

R: Probably yes. Yeah. And I think because of, because the thing, um, chopping and changing my antidepressants, I don't think that's helped my mood through all of this, and I've just been on a 2 week cruise and I was spending most of the day in the cabin, because I didn't have the energy or the inclination to do anything.

I: And um, do you think you got some benefit, do you think you've had some benefit from the treatment?

R: Definitely. Because it's still in my head, what I did with [the study physiotherapist]. And I've still got the blue book [workbook]. [Julie, age 50]

11.3 Challenges of Putting it into Practice

Common to all participants were reports that adjusting to everyday life post-treatment and putting symptom management plans into practice was more difficult than they had expected. Challenges frequently came with setbacks due to co-existing health problems. This included the side effects of withdrawing from benzodiazepine medication (Michael); a fall resulting in a fractured wrist, labyrinthitis, and chronic pain (Julie); chronic fatigue (Amy, Sarah, Megan); an abdominal mass cancer scare (James); and migraine (Megan).

R: So it just feels like I go 10 steps forward and 10 steps back. And that's what it's been like, a constant battle. But I know that's what I got to deal with. [James, age 36]

Another challenge frequently described by participants was finding the time to implement treatment strategies amongst busy work or family life schedules. Also, for some participants, movement strategies that reduced motor symptoms during the intervention did not work as well at home, causing frustration. Maintaining motivation when progress appeared to plateau was difficult.

Some participants expressed disappointment that they had not made additional progress between discharge and follow up, or that they had failed to meet personal rehabilitation goals.

R: So I haven't got to where I wanted to be. Um, I wanted to be where I maybe still have, like maybe a bit of a limp or, do you know what I mean? But not have no aid. That was my goal, was the next I come and see you guys I can go, "Look!" And to me I feel like that was another thing I was beating myself up about because I wanted to be aid free next time I came to see you. [James, age 36]

Some participants described how they coped with challenges and setbacks by adapting their management plans and resetting their expectations. Participants who displayed

this adaptability may have been more likely to feel as if they were on an overall trajectory of improvement.

I: Ok. Um, and so you had a plan of things to do when you left. Were you easily able to introduce that into your everyday life when you got home?

R: Not really, no. I started doing it like [the study physiotherapist] said, which was very good, doing a little bit each day at home. So just silly things if I got home and I was chopping away or just making a cup a tea and walking around the house and doing that. But I found it was too much to think about. So I was trying to, you know, bend my knee, heel to toe, it was all too much. ...But that is because I've got children and I've got to do school pick up. Drop off and things like that. If I didn't have that and I didn't have my job then maybe I could have every day, I could have set aside more time and done more. So I had to adapt it to my lifestyle. [Nicole, age 45]

11.4 Perceived Role of Psychological Factors

In their baseline interviews, participants had been generally reluctant to consider that psychological factors were relevant to their movement problem. Immediately after the intervention, there was less resistance amongst participants to considering that psychological factors were relevant and some participants described coming to the realisation that they were anxious and that this was part of their problem. In the six month follow up interviews, participants remained open to considering that psychological factors, specifically anxiety and depression, were related to their problem. The level of perceived relevance differed amongst participants.

I: Do you think psychological things are part of functional movement disorder?

R: Part of it, I think there's a chunk of it. I don't think it's all because it's not all. I think it's all, um, because you've got the physical effects that go with it. But I think that maybe I'd say a chunk of it is. [James, age 36]

This role of psychological factors was usually perceived to be as a symptom modulator, in that anxiety and depression made their physical symptoms worse. Participants generally disagreed with the idea their problem was *caused* by psychological factors. This notion was considered either completely wrong or overly simplistic. Some participants considered the converse to be true, and that anxiety and depression were secondary problems caused by their movement disorder.

I: OK and we spoke before about how some people feel that psychological issues are generally part of the problem. What are your thoughts on that?

R: They're probably a risk factor... I don't think it's psychological based. But I think [psychological factors] coupled with the physical probably caused the whole main thing. But I don't think it's just psychological. [Megan, age 22]

Low mood was a particularly prominent theme in the six-month follow up interviews of two participants. Both described how their mood had limited their progress. After completing the study intervention, both participants had sought psychological treatment. This contact for psychological therapy took place over the telephone for both participants and unfortunately both describe a negative experience.

R: And I've spoke to the people on the phone from Friends Life, but they don't know what they're dealing with.

I: What do they do?

R: They just talk to you about it. Talking therapy. It's about, so how do you feel today? And what did you do and how did you make things better. And it's just, you know it's just text book. They're reading off a book or off a sheet. They don't know my actual, what I'm going through. They only know... um, trying to... they only know the textbook side of stuff. Oh he feels depressed today, ok. Do you know? And I feel it's sometimes a waste of time because I get off the phone feeling worse than I did when I got on the phone. Because I feel like they've drained all the energy out of me talking about my problems. And I feel worse than I actually did before they actually called me. [James, age 36]

I: You told me you saw a CBT therapist recently, what did they say?

R: Oh no I didn't see them. No, it was on the telephone.

I: Tell me about that.

R: Well I just thought that [it] was weird. Totally weird [laughs]. "Do you want to commit suicide?" No! "Do you get anxious when you go out the door? Do you feel sick? Do you want to go to the toilet? Do you want to, do you want to kill somebody?" All these stupid questions. Well I say stupid, stupid to me because it, no, my brain didn't think like that. [Lynn, age 56]

11.5 Feedback from Participants

The six-month interviews revealed components of the intervention that participants considered particularly helpful, and also components that were potentially unhelpful.

11.5.1 The Intervention Workbook

Most participants reported finding the intervention workbook valuable. Some described how they referred back to the workbook regularly, while others reread the workbook as a reminder before attending their six month follow up appointment. The intervention workbook contained personalised information about FMD, strategies that helped to normalise movement, and a plan to improve their symptoms in the long term. Several participants described finding this information useful and reassuring. It was also used by participants to help explain their problem to others and some reported that they were able to use it to help local HCPs understand their problem (potentially adding legitimacy to their problem).

R: And when you're actually flung into normality, it's quite hard to improve. But um, I just kept looking back on the plans that we went through and the study-book that we wrote out which, to help me with stuff. So that book that we wrote out came quite in useful, cos you look in back on things that you'd done here. [James, age 36]

I: The workbook that you did in physio. Was it useful when you went home?

R: Yeah. It was really useful to show other people as well. Especially the explanation of how FMD can manifest and how it, the whole, how it happens, the model. ...And going through it with close family and they understand it a lot better. [Megan, age 22]

The workbook also contained a section where participants had been asked to write a reflection on each treatment session. One participant described how this had helped them build a better understanding of their problem, by being encouraged to reflect on the information they had been given and then being given the opportunity to clarify questions in the following session.

R: I thought the writing, the reflective writing [was useful], because sometimes I had questions and I could go back and ask [the study physiotherapist] about them and we then we would, you know, just talk over things as well. [Deborah, age 58]

Several participants found that writing and rereading their reflections gave them new perspectives on their problem and helped them to see how much progress they had made.

R: And all the reflections that we did, when I wrote in my [workbook] each night. Just for me, it put things into perspective really. That I haven't got to make life quite as difficult for myself as I do sometimes. [Deborah, age 43]

R: So reading back, I found all the reflective stuff that I was asked to do each night that was what fascinated me. Um, because I'm at a completely different stage now. [Lisa, age 43]

11.5.2 Preparing Participants for Discharge

As part of the intervention, the study physiotherapist routinely warned participants to expect setbacks and discussed strategies to cope with difficult periods and to get back on track. This was supported in the workbook by a personalised relapse management plan. However the rapid improvement that many participants made over the five day intervention often left them with high expectations for further progress post discharge.

R: I think, when I was here and the week that I was here, I had such amazing results in such a short, almost instantly. That I kind of expected to follow that. So in my head, the improvements um and the, the, I guess connections would be in a similar linear um, process. [Lisa, age 43]

The common experience reported at the six-month follow up interviews was that maintaining rehabilitation gains and making further progress was more difficult than they had expected. Setbacks and periods of symptom exacerbation were common. Despite efforts to manage participants' high expectations, it appeared that some participants were disappointed with their lack of further progress.

I: Do you think, um, we did enough, or do you think there was enough discussion from us as a team about what to expect? Do you think we prepared you?

R: Um, I think [the study physiotherapist] tried. Um, whether or not I would have listened if she would have been more explicit, I don't know. Um, what would have been helpful, would have been some follow up. So perhaps some conversation, whether it was a phone call, or some sort of... whether it was meeting a local physio, just to sort of recap, um, that would have been helpful to kind of amplify the angel voices and stop me running away with my own thoughts. But I, yeah, because I felt that the first, and I don't know. I don't know that the first couple of months after being here. I wouldn't say they were wasted, I needed to go through those emotions, there's no short cut. But whether or not that would have helped me, I guess move forward through those stages a bit quicker, um, I don't know. [Lisa, age 43]

11.5.3 Earlier follow up

Several participants (including the example in the above quotation) suggested that a follow up sooner than six months would have been helpful. Participants described needing some reassurance and wanting an opportunity to practice strategies to control their abnormal movements, to talk through difficulties and revise their management plans. Participants had been encouraged to telephone the physiotherapist if they had any questions or concerns and this was reiterated within the workbook. However less than a quarter of participants actually made contact between discharge and follow up (7 of the 30 intervention participants). One participant described that she wanted to call for advice but was too concerned about being considered a treatment failure.

11.5.4 Patient Interactions

Several participants were curious to hear the stories of other people with FMD and would have liked to have met other trial participants. The desire to meet other patients was related to their sense of isolation and the fact that many had been led to believe their problem was rare and unusual.

11.5.5 Intensity

The high intensity of the study intervention may be an important therapeutic ingredient enabling rapid progress; however some participants found the intensity very challenging. One participant suggested spreading the treatment sessions out over two weeks.

11.6 Summary

At six months follow up all but one participant reported that their symptoms had improved compared to baseline. The reported impact of the intervention on participants' lives was wide, and varied amongst individuals. It was difficult to disentangle physical, psychological and social changes and determine the extent to which each individual benefited. A common theme for all the participants, was that

putting everything into practice was more difficult than they had expected. Challenges came with setbacks due to other health problems, finding the time to implement rehabilitation strategies, maintaining motivation when progress plateaued and managing fatigue. The optimism and expectation of recovery that was prominent in the interviews conducted immediately after the intervention was tempered at six months with a realisation that there was no quick-fix to their problems and that further change would be slow.

In the baseline interviews, participants had described feeling stuck and powerless to help themselves. At six months, most continued to experience symptoms, but with a sense that they had generally moved forward. This seemed to be associated with having a better understanding of their difficulties and thinking that things could improve in the future. Many described feeling more accepting of their problem. The interviews provided an insight into what participants found helpful about the intervention and areas where adjustments may help to improve participant experiences and outcomes.

11.7 Central Themes Across All Three Time Points

There were three themes that were central to the participants' narratives and common to each interview time point (baseline, post treatment, and follow up). These were: (i) the Impact of FMD; (ii) Dissatisfaction with Psychological Explanations for Symptoms; and (iii) Understanding. There were some changes within each theme immediately following the intervention and at six month follow up. This is described in Table 11. 1 below and discussed further in Chapter 12.

Table 11.1. Central Themes Across All Three Time Points

	Prior to Treatment	Post Treatment	Six Months Follow-up
The Impact of FMD	Functional motor disorder had a substantial negative impact, with participants describing both a physical and emotional burden.		
	<p>The physical burden of FMD varied amongst the participants, however, most described experiencing mobility restrictions and/or limitations in completing normal activities of daily living, such as washing, dressing, meal preparation, and sustaining employment.</p> <p>The emotional burden of FMD appeared to be of at least equal if not greater than the physical burden. The emotional burden was expressed as distress associated with frustration related to coping with symptoms, not understanding what was wrong, the unknown prognosis/future, and difficulty finding support from health care professionals.</p> <p>Isolation was a particularly prominent and distressing consequence of living with FMD. This was especially a problem for those who had left work or education due to ill health.</p> <p>Participants commonly described feeling powerless and unable to help themselves.</p>	<p>All but one participant felt the intervention had helped their motor symptoms. There was considerable variation in the perceived extent of change; some described only modest improvements while others described significant changes such as no longer requiring walking aids.</p> <p>In post-treatment interviews, participants often described feeling relieved and reassured, suggesting the experience of their functional symptoms felt less threatening. This was associated with the feeling of having a greater understanding of their problem.</p> <p>The intervention appeared to leave the participants feeling hopeful for their future.</p>	<p>Most participants reported ongoing benefit from the intervention. Often, improvement in symptoms translated to positive changes, such as less reliance on mobility aids, return to work, and less tension in interpersonal relationships.</p> <p>It was common for participants to describe feeling more at ease with their problem. This change was associated with having an acceptable diagnostic label, a greater understanding of their problem and some resolution of the uncertainty of their prognosis.</p> <p>The hope and optimism that was prominent immediately post-treatment remained to some extent, but was now tempered with a realisation that future progress was likely to be slow.</p> <p>The intervention appeared to empower the participants, with many feeling able to manage their problem more independently.</p>
	Outlier: One participant felt their symptoms had relapsed to baseline levels and they had not sustained any physical benefits from the intervention. However, they continued to feel some hope for the future and described feeling less threatened by their symptoms.		

	Prior to Treatment	Post Treatment	Six Months Follow-up
Dissatisfaction with Psychological Explanations for Symptoms	Most participants were unsatisfied with psychological explanations for their movement problem. After the intervention there was less resistance to the idea that psychological factors were part of their problem.		
	<p>Most participants felt dissatisfied with psychological explanations for their symptoms.</p> <p>Physical precipitating events or an initial causal attribution to neurological disease were central to most participants' illness narrative and psychological explanations were generally seen to be at odds with their physical/biological experiences.</p> <p>Those who experienced psychological problems such as anxiety or depression, generally felt that these were separate issues to their movement problem or reactive to their disability.</p> <p>Psychological explanations were often interpreted as meaning there was nothing wrong.</p> <p>Psychogenic illness was commonly associated with pejorative stereotypes, which some participants made efforts to distance themselves from (e.g. I'm not that type of person).</p>	<p>Dissatisfaction with psychological explanations continued in many participants, but for some there was less resistance to the idea that psychological factors could be part of their problem.</p> <p>Some participants described a new realisation that at times they felt anxious which was part of their movement problem.</p> <p>Despite this softening stance, all the participants tended to hold the view that psychological factors, if present, were usually secondary to their motor symptoms or a separate issue and that they were not the cause of their FMD.</p>	<p>Those who were less resistant to considering a role for psychological factors as part of their movement problem post treatment remained open to this idea at follow up.</p> <p>Psychological factors were generally seen as symptom modulators, exacerbating the problem rather than the cause.</p> <p>Some participants remained unconvinced that psychological factors were part of their movement problem.</p> <p>The explanation for FMD used in the study, which can be considered as biopsychosocial in scope with a biological emphasis, appeared to resonate with most participants. The study explanation for FMD appeared to provide validation of the participants' illness experience, in a way that a psychologically focused explanation did not.</p>
	There was a tendency for participants to view their problem from a dualistic mind-body perspective. The division between mind and body blurred for some participants after the intervention.		

	Prior to Treatment	Post Treatment	Six Months Follow-up
Understanding	A central theme relating to the patient's understanding of their problem ran through the participants' interviews at all three time points. Understanding had an important impact on the lived experience of FMD and appeared to affect the ability to engage with the intervention.		
	<p>Most participants reported a lack of understanding of their movement problem and symptoms were commonly described as mysterious.</p> <p>Most participants perceived that doctors did not understand what was wrong.</p> <p>It was common for participants to feel unconvinced by the diagnosis of FMD.</p> <p>Lack of understanding meant that the future prognosis was uncertain, and for some this was frightening to the point of being distressing.</p> <p>Not knowing what was wrong meant that the participants did not know how to help themselves, leaving them feeling powerless.</p>	<p>Most participants reported that the intervention had given them a greater understanding of their movement problem.</p> <p>Most reported feeling that the gain in understanding was the most valuable treatment outcome.</p> <p>Most participants described their problem in a way that resembled a biopsychosocial formulation of FMD.</p> <p>Several participants attributed improvement in their movement problem to having a greater understanding.</p> <p>Understanding appeared to reduce the threat value of symptoms and gave many a sense of empowerment and a perception that they were better equipped to self-manage.</p>	<p>At follow up, the understanding gained with the intervention was still considered the most valuable treatment outcome and most were convinced that the diagnosis of FMD was correct.</p> <p>The participants' understanding of their problem could be described as biopsychosocial in nature, with an emphasis on the biological. Most continued to feel that some things remained unexplained, for example why they developed FMD in the first place.</p> <p>A gain in understanding appeared to be associated with a number of positive treatment outcomes. This included reduced threat value of symptoms, hope for the future, and validation of their illness experience.</p>
	Outlier: Confusion at baseline was common to all, but only one participant felt that her problem remained a complete mystery after the intervention. She described how the intervention could not help because nobody knew what was wrong.		

Chapter 12 Qualitative Study Discussion

This qualitative study involved interviews with a selection of participants from a feasibility trial, who received a specialist physiotherapy intervention for FMD. The participants were interviewed at baseline, after treatment and at six months follow up. Interviews were transcribed and analysed across cases at each time point. The findings from the baseline interviews provided insights into how the participants experienced the onset of FMD, their interactions with the health care system and the impact of FMD on their lives. The interviews conducted immediately after treatment provided insights into how the participants experienced and interacted with the intervention. The six month follow up interviews provided insights into the impact that the intervention had on the participants' lives. The data from all three time points are discussed here and considered in relation to the usefulness and further development of the specialist physiotherapy intervention. This chapter concludes with a discussion of the limitations of the qualitative study and directions for future research.

12.1 The Impact of FMD

Participants' accounts demonstrated the significant negative impact that FMD had had on their life. It was reported that physical disability due to FMD had a wide ranging impact, including limiting the ability to complete normal activities of daily living such as washing and dressing, fulfilling their roles as a parent and sustaining paid employment. This in turn put significant strain on their inter-personal relationships. Perhaps of equal or greater significance to the physical impact was the distress associated with FMD. Participants used emotive language to describe this impact, for example "I've lost virtually a year of my life, because of my condition" [Michael, age 46, table 2]. Distress was associated with not knowing what was wrong, the uncertainty of their prognosis, difficult finding support from health care professionals (HCP) and social isolation.

Other studies of patients with FMD have previously reported high levels of disability and distress. Carson et al (2011)⁶ found that patients with functional neurological symptoms (n=1144) scored higher on measures of disability (SF12) and distress (HADS) than patients whose symptoms were explained by disease (n=2637), in the cohort of

patients taking part in the Scottish Neurological Symptoms Study. Anderson et al (2007)²⁰³ compared patients diagnosed with “psychogenic movement disorder” to those diagnosed with Parkinson’s disease and found equivalent levels of disability and impaired quality of life, while those with psychogenic movement disorder had more anxiety and depression.

12.2 Resistance to psychological explanations

In the baseline interviews, participants were generally resistant to psychological explanations for their symptoms. Even those who acknowledged a history of psychological problems felt that this was unrelated to their physical symptoms. Such a rejection of psychological explanations by patients is widely reported in the literature, backed by evidence from qualitative studies,^{195,196} illness belief questionnaires,²⁰⁴ and anecdotal evidence from neurologists.²⁰⁵ The current study offers data which may help to understand some of the reasons why patients may reject these psychological explanations.

First, most of the participants associated the onset of their symptoms with a physical event, usually an injury or illness that appeared to precipitate their motor symptoms. In addition, they described the impact of FMD in terms of physical disability which they perceived (or reported) to be the cause of secondary distress. Psychological explanations felt at odds with their physical experiences and left them unconvinced by a psychogenic diagnosis. This mismatch in participant experience and diagnostic explanation can be considered in relation to the issue of mind-body dualism. The participants’ conceptualisation of illness appeared to be dominated by a biomedical model, where it is assumed that illness is fully accounted for by deviations from the norm of measurable biological variables.²⁰² Consequently, whatever is not capable of being explained in this way is necessarily excluded from the category of disease. The problem of mind-body dualism was likely reinforced by monothematic psychological explanations given by HCPs, leaving an explanatory gap between the (physical) experience and (psychological) diagnosis that left participants feeling misunderstood and dismissed. The issue of mind-body dualism may also help to explain previous

reports of patients diagnosed with “conversion disorder” holding onto beliefs of having an underlying and as yet undiagnosed neurological disease.²⁰⁴

Second, psychological explanations were interpreted by many participants as meaning that the doctor did not believe they had a real or significant problem. Some even felt that HCPs thought that they were faking their symptoms. These interpretations of psychological explanations were associated with pejorative stereotyped beliefs about mental health issues; which included that they are trivial problems that could be overcome if the person wanted to get better, that the patient is at fault and that mental illness is a character flaw. Participants either endorsed such views or projected them onto others. The implication was that psychological problems are generally not considered a legitimate cause of illness.

Third, psychological explanations felt dangerous for some participants. Several potential sources of perceived danger were identified, such as the danger that the neurologist had honed into their past history of psychological problems, preventing them from performing a thorough assessment and therefore potentially missing underlying disease. There was reported danger of being subjected to unhelpful and undesirable treatment (reported experiences included the frightening experience of being locked in a psychiatric hospital ward and the prescription of unhelpful sedating and addictive medications). Finally, there was a perceived danger of being excluded from active treatment (such as physiotherapy and botulinum toxin).

The attitude and behaviour of clinicians may also have played a role in the participants' resistance to psychological accounts of FMD. Mental health problems notoriously lack equality with physical health problems. This point is made vividly by Sarah, who reports being told by a doctor “you haven't got a brain tumour and you haven't got cancer, I've got other patients.” Whether or not this is an accurate representation of the consultation between Sarah and her doctor, this was the message she took from it and the inequality and undesirability of problems related to psychological factors is made clear. In support of the possibility that patients may pick up on negative attitudes that are held by clinicians is a survey of neuroscience nurses.¹⁹³ One in six did not think that functional motor symptoms were real and one in 10 thought that

patients with FMD wasted doctors' and nurses' time and were not deserving of the same level of care as patients with organic disease. Also, in a survey of 349 British neurologists,²⁰⁵ it was found that many harbour a belief or concern that patients with FMD are feigning their symptoms. Only 44% felt that conversion disorder was completely distinct from feigning, the remaining 56% felt there was an overlap between the two or that one was a subset of the other. The attitudes and prejudices of both patients and clinicians help to explain the reluctance of patients to consider psychological factors as part of their problem.

12.2.1 A change in perspective post-treatment

A novel finding of this study was that there was less resistance to considering a role for psychological factors amongst some participants after receiving the study intervention. There may be several explanations for this finding. First, some participants described how the intervention had helped them to come to a new realisation that they felt anxious and that this was part of their problem. Their previous lack of recognition of feelings of anxiety may be explained by the psychological construct of alexithymia, which is defined as an inability to accurately recognise one's own emotions.²⁰⁶ Patients with FMD have been found to score higher on scales of alexithymia compared to the general population.^{37,207} However, this does not appear to account for the change in perception of all participants. Another potential explanation is that some participants felt "safer" disclosing experiences of psychological symptoms, having had their primary concern, that is, their physical symptoms, taken seriously and that they would not be dismissed. Patient recognition of psychological factors may be an important part of successful treatment. In an epidemiological study of 716 patients with neurological symptoms that were "not at all" or "only somewhat" explained by disease, non-attribution of symptoms to psychological factors by patients was found to be a strong independent predictor of poor outcome, with an odds ratio of 2.0.¹⁵³

12.3 The importance of understanding

Understanding of their problem was a central theme in each participant's narrative. The role and relevance of this understanding changed in the post treatment and follow up interviews.

12.3.1 Understanding at baseline

In the baseline interviews, participants considered their problem to be mysterious and they generally perceived that doctors had not understood what was wrong. This perception was reinforced when participants felt their diagnosis had been made based on negative investigations. Other experiences that undermined the participants' confidence in HCPs included receiving conflicting information from different specialists and prescription of treatment that they felt had caused them harm. Some saw psychological explanations for their symptoms as further evidence that the doctor did not understand their problem, whilst others who had received more biological explanations, such as the "*software not hardware*" analogy, were left feeling confused about what was wrong. Difficulty understanding the diagnosis amongst patients with functional neurological symptoms is well reported in the literature.^{195,196}

There were significant negative consequences associated with this perceived lack of understanding. Several participants described how not knowing what was wrong and the uncertainty about their prognosis was frightening. Not knowing while waiting for appointments, investigations and a diagnosis was a particularly distressing experience for some. A lack of understanding was also associated with powerlessness. Participants described that, as they did not know what was wrong, they were unable to help themselves and therefore they were stuck in their distressing situation.

12.3.2 Understanding after treatment

Most participants reported that the intervention had helped them to develop an understanding of their problem. The understanding reported by participants was not a complete picture of FMD, however most recognised that the aetiology was multifactorial and included factors other than structural disease. This may be

considered a move from a biomedical model of illness towards a biopsychosocial conceptualisation of their problem. Improved understanding was usually considered the most valuable treatment outcome, which emphasises the negative effect that not knowing had had prior to treatment. When interviewed at six months, improved understanding remained the most highly regarded treatment outcome.

12.3.3 The study explanation for FMD

The study explanation for FMD appeared to resonate with the participants. A potential reason is that the explanation did not attempt to challenge pre-existing beliefs held by participants about their problem by enforcing a psychological explanation. Instead, the explanation aimed to build on their existing beliefs and develop a broader biopsychosocial understanding of their problem. This was achieved by formulating the participant's experiences into a coherent (evidence based) biopsychosocial model, thereby providing an explanation that was perceived as relevant, legitimate, personally acceptable and amenable to treatment. A key element of the explanation was that the physical events that, at baseline, most participants perceived to have precipitated their problem, became central to the personalised explanation. Psychological factors were included in the explanation, usually described as "fight or flight" reactions, which may be considered a biological 'spin' on psychological processes. The extent to which psychological factors were included in any participant's personalised formulation was dependent on the extent to which they considered them relevant to their problem.

These findings are similar to those reported by Karterud et al (2015).²⁰⁸ They conducted semi-structured interviews with 11 adolescent and young adult inpatients who had been diagnosed with non-epileptic (functional) seizures. They found that the use of a multifactorial biopsychosocial model helped patients to accept the diagnosis by providing a recognisable reflection of their lives. In addition, they reported that the biopsychosocial model preserved self-esteem, in a way that psychologically heavy explanations and associations with mental illness did not.

Another potentially important factor in the acceptability of the explanation is that the aetiological model on which it was based was connected to a corresponding

treatment. This may have alleviated the participants' previous concerns of feeling marginalised and abandoned by the NHS, and also a perception reported by some participants that there are no available effective treatment for psychogenic problems.

12.3.4 The impact of the perception of having a greater understanding

The perception of having a greater understanding of their problem was associated with several positive consequences: feeling reassured, feeling empowered, feeling hopeful and increased self-confidence.

Most participants described how understanding their problem left them feeling reassured. This related to the fear associated with the unknown that was reported in the baseline interviews. In the post treatment and follow up interviews it was common for participants to describe feeling less threatened by their symptoms, which may be an important mechanism by which the intervention worked. Studies of mediators of change in patients undergoing treatment for chronic pain and chronic fatigue (both of which may be aetiologically related to FMD), have found that a perception of reduced threat of their symptoms mediated improvement.^{209,210}

Understanding was perceived by many participants to be the reason that they improved with the intervention. This relates to the perception in the baseline interviews of being stuck and unable to help themselves due to not knowing what was wrong. Post-treatment, participants described a sense of self-reliance in managing their health problem. In this way, understanding was empowering. This finding may also be related to the role of illness belief as an aetiological mechanism that causes FMD, whereas a change in understanding may be associated with a positive change in problematic illness beliefs and expectations.

12.4 Legitimation and Validation

The intervention appeared to give validation and legitimacy to the participant's illness experience. This is likely to be an important factor in making the intervention acceptable and helpful.

The sociological concept of the “sickness role”, first described by Talcott Parsons in 1951, is an influential theory of illness behaviour that may help to understand the significance of having illness validated.²¹¹ Parsons described how the sick role provides an exemption from normal social roles and obligations. The sick role comes with an assumption that the person requires specialist help to get better and that access to the sick role requires legitimisation of the illness from a doctor. An argument can be made that previous interactions with HCPs had not resulted in a diagnosis that the participant or society had considered a legitimate reason for entering a sick role. Exclusion from the sick role may then have prevented the commencement of convalescence and recovery or adaptation. Instead, the participant is left searching for a diagnosis that validates their illness experience and grants them access to the sick role. The concept of the sick role has been criticised over the years for being a simplistic representation of behaviour that does not necessarily represent all cultural, ethnic and socioeconomic groups.²¹¹ However, it remains an influential concept, which may help to understand conflict between patients with FMD and clinicians.

12.5 From passivity to becoming active participants in rehabilitation

In the baseline interviews, participants were generally passive in their illness role. They described feeling stuck and unable to move forward with their problem. This changed in the post treatment and follow up interviews. Participants described a sense of self-reliance in managing their health and many reported an expectation of future improvement in their FMD symptoms following self-management. In this way, they can be described as active participants in their rehabilitation. Ingredients that appeared to be important in empowering participants to become active in their own rehabilitation were; receiving an acceptable diagnosis, having an understanding of their problem, validation and support from clinicians whom they trusted and they considered to understand their problem, having strategies that they believed improved their motor symptoms and having hope for the future.

12.6 The impact of the intervention

The longitudinal design of this study provided insights into how the participants were able to use the intervention. In general, all participants described having had at least some benefit.

Perhaps unexpectedly for a physiotherapy delivered intervention, “non-physical” benefits appeared to be more prominent than physical benefits in the participants’ accounts. As discussed above, most reported that the greatest value of the intervention was gaining a greater understanding of their problem. In many cases, this appeared to be associated with other benefits including resolution of the distress of uncertainty, hope for the future, and empowerment of the individual. Some participants also described improvement in their inter-personal relationships, which had been negatively affected by FMD. The non-physical benefits that participants attributed to the intervention appeared to reduce the general distress associated with FMD and may account for the new sense of feeling at peace with their diagnosis described by several participants.

Other studies have reported similar findings. Sharpe et al (2011),⁷⁷ in a RCT of guided self-help for functional neurological symptoms, based on Cognitive Behavioural Therapy principles, found that the intervention was associated with reduced health anxiety (as measured by the Whiteley Index). Also, Demartini et al (2014),⁷¹ in a cohort study of 66 patients with mixed functional neurological symptoms, reported that multidisciplinary rehabilitation led to a statistically significant improvement in the Illness Perceptions Questionnaire subscales of illness coherence (understanding) and emotional representation (distress caused by symptoms).

Participants also reported physical benefits from the intervention. Immediately after treatment all participants reported an improvement to their physical symptoms. At six months, all but one continued to report physical benefit. Some considered that their symptoms continued to improve after treatment ceased, while others reported maintaining the benefits gained with treatment but felt there had been little further

progress, and some reported improvement from baseline but with some loss of the treatment effect.

There were also reports of broader health benefits from the intervention; this included qualifying for cataract surgery when previously a head tremor precluded treatment and coming off benzodiazepine medication.

In the post-treatment and follow-up interviews, there was a sense amongst many participants that they had become more self-reliant in managing their symptoms. This was associated with feeling as if they understood their problem, knowing how to manage it, as well as knowing there was backup support from the study clinicians if needed in the future. An implication of a new sense of self-reliance is the potential for a reduction in future use of health resources and therefore future cost savings. The reports of returning to work and coming off medication also suggest the potential for cost-benefit.

12.7 Barriers to recovery and limitations of the intervention

Participants' accounts of the intervention and how they fared in the period between treatment and follow-up revealed potential barriers to recovery and limitations of the intervention.

Comorbidity was commonly reported as a cause of symptom relapse or ongoing disability. Reported comorbidities that were associated with ongoing problems were fatigue, persistent pain, recurrent migraine, transient illness, low mood and a cancer scare (abdominal mass found and biopsied). Most patients with FMD have comorbidities, such as neurological disease, hypermobility syndromes, anxiety, depression and other "medically unexplained" symptoms such as persistent pain and fatigue. Comorbidity can be formulated into the biopsychosocial understanding of FMD, contextualised as predisposing, precipitating and perpetuating factors.¹³ The clinical implication is that addressing comorbidity is an important part of treatment of FMD. The presence of comorbidity may influence the suitability of a patient to a particular treatment option. Therefore, patients with, for example, significant

comorbid anxiety, depression and pain may benefit from a more multidisciplinary approach to treatment. Comorbidity may also influence long term prognosis and outcome with treatment.

A potential limitation of the intervention relates to the finding that many participants found that implementing their self-management plan was more difficult than they had anticipated, leading to frustration and declining motivation. Participants were warned to expect “ups and downs” after treatment and that progress might not be as rapid as during the five days of the intervention. However, it appeared that many continued to have very high expectations in regard to their recovery which went unmet. This may indicate a need to re-evaluate how we prepare patients to cope post treatment. An alternative view about this issue is that high expectations of recovery may be helpful. This would be predicted by the underpinning aetiological model for FMD, in which expectation is considered a mechanism that drives abnormal movement. Supporting the aetiological role of expectation in FMD is the finding that expectations of non-recovery predict a poor outcome.¹⁵³ It may follow that high expectations of recovery predict a good treatment outcome and therefore expectations of recovery are therapeutic. In this case, some disappointment at lack of symptom resolution or the slowness of improvement may be a necessary inevitability.

12.8 Clinical and Research Implications

The findings from this research have several implications for clinical practice.

12.8.1 The importance of understanding the diagnosis

The participants in this study considered that developing an understanding of their problem was important for them to improve. This highlights the importance of providing a diagnostic explanation that patients find acceptable and relevant. In the current study, explaining the diagnosis started with the neurology consultation at the time of potential recruitment to the study and was backed up during the specialist physiotherapy intervention.

12.8.2 Vulnerability and risk of iatrogenic harm

Reports of receiving previous treatments that participants perceived had caused them harm were alarmingly common (6 out of 11 participants). This highlights the vulnerability of this patient group to iatrogenic harm. It is probable that the marginalisation of these patients from mainstream medicine, as perceived by this cohort and as has been reported in previous studies,¹⁹⁵ contributes to this vulnerability. The availability of specialist services for patients with FMD, where they can receive an early diagnosis and be directed towards a range of evidence based treatments that reflect the heterogeneity of this group may help limit iatrogenic harm.

12.8.3 Who is suitable?

Most of the participants described having had some benefit from the study intervention, but there was little data from the feasibility trial or qualitative study which would help determine in the future who is most suitable for this type of treatment. This may be a reflection of the limited sample size and participant selection bias in the qualitative study. Alternatively it may indicate that the study eligibility criteria were appropriate.

12.8.4 Discussing psychological factors with patients

Several participants described having had particularly negative experiences with previous experiences with HCPs that left them feeling ashamed of their problem. These experiences were usually related to a perceived implication that their symptoms were psychological and not real. Other studies have found that explanations for functional neurological symptoms that imply mental illness can be an affront to self-esteem.²⁰⁸ The explanation given to participants in the current study was based on the clinical observation of neurologists Jon Stone and Mark Edwards, that patients are more accepting of the diagnosis when it is explained in terms of the physical diagnostic signs displayed by the patient.¹⁵⁴ They suggest that the explanation should note that psychological factors are an important part of the problem for many people, but that

the initial consultation is not usually an appropriate time to open up a dialogue about the sensitive issue of mental health and past traumatic experiences.

The study data supports the clinical approach described above. In the baseline interviews, participants were resistant to the idea that their problem was related to psychological factors. However, it was found that this resistance to psychological explanations often softened in post treatment and follow up interviews. The intervention helped participants to develop a broader biopsychosocial understanding of their problem, which allowed a role for psychological factors which was not necessarily causal. Other factors that may be important for the softening stance against psychological factors are that they had secured a diagnosis, granting them access to the sick role (i.e. validating and legitimising their illness), and knowing that they would not be dismissed as was their reported previous experience (for example, Lynn's experience of being told "we think it's psychological and there is nothing wrong with you"). The intervention may also have helped participants to recognise the presence and impact of anxiety.

The clinical implication is that there may be value in "going in easy" with psychological explanations in the early stage of diagnosis, in order to engage the patient. Commencing physiotherapy may for some patients be an important precursor to psychological therapy. It should be noted that the resistance to psychological explanations may be particular to certain populations, such as patients seeking support in tertiary neurology clinics, and there may be patients with FMD who actively seek psychological support, who are not represented in the current study.

12.8.5 Measuring change in clinical and research settings

Typically, when assessing the impact of a physiotherapy intervention, the focus is on measuring physical impairment and disability; non-physical changes may be an afterthought or even ignored. The findings from this research suggest that non-physical changes make up a substantial part of the positive impact of the intervention on participants' lives and therefore this should be considered in outcome measurement. Specific non-physical changes found in the qualitative data that were

associated with the intervention were: (i) a perception of increased understanding of the problem; (ii) an expectation of recovery; (iii) reduced distress from the lack of understanding of FMD, the uncertain prognosis and lack of support from HCPs; (iv) reduced threat value of symptoms; (v) a perception of self-efficacy; and (vi) improved interpersonal relationships. These findings require further validation and assessment.

Choosing outcome measures that reflect the impact of treatment and are sensitive to change in patients with FMD is problematic. For example, patients with FMD are recognised as having high levels of distress, which may improve with effective treatment. A prominent source of distress found in the baseline interviews of this study was the perception of being abandoned by the health care system. This source of distress will not necessarily be captured by generic anxiety or depression scales, nor by commonly used measures for illness distress, such as the Illness Perception Questionnaire, where none of the 87 items appear to take this specific issue into account.²¹²

12.8.6 Important elements of the intervention

Elements of the intervention that participants valued and which may be important therapeutic ingredients were: (i) the focus on explaining the diagnosis; (ii) the specific symptom-explanatory model; (iii) the focus on self-management; (iv) consecutive days of treatment (although some participants found the intensity challenging); (v) staying in accommodation away from home; and (vi) the intervention workbook.

It was common for participants to comment after treatment that they held the study clinicians in high regard. Higher levels of therapeutic alliance have consistently been shown to be associated with better outcomes in both medicine and psychology, and there is some evidence for this effect in physiotherapy for chronic low back pain.²¹³ It is therefore highly likely that the therapeutic alliance may have been an important therapeutic ingredient in the intervention.

12.8.7 Refining the intervention

Based on feedback given by participants during their interviews, some elements of the intervention could be refined to improve the patient experience, and possibly improve the intervention outcome. The most commonly made suggestion was that the follow up appointments should be scheduled earlier than six months. The six months interval was chosen in the feasibility study in order for this appointment to coincide with the final outcome measurement. In clinical practice, a follow up interval of around four to six weeks may be more appropriate. The participant suggestion of a scheduled telephone call at one or two weeks may have additional therapeutic benefits. Other suggestions for the intervention that warrant investigation and assessment are the inclusion of group sessions and flexibility to reduce the intensity of scheduled sessions (e.g. delivery of the intervention over two weeks).

12.9 Limitations

There are several limitations to this study that should be acknowledged.

The findings reported represent the views of a relatively small sample with some limitations in diversity. A purposive sampling plan was predefined in an effort to recruit a representative sample of patients. This planned representation was to some extent achieved in that no predefined characteristic went unrepresented in the final cohort. However, there were only two men and one person over the age of 60. The sample also had limited representation of patients with poor treatment outcomes, in that only one of the 11 participants reported at six months that their symptoms had not been improved with the intervention and all participants reported getting at least some benefit from the intervention. The longitudinal study design with interviews commencing prior to treatment meant that it was not possible to purposively select participants based on poor treatment outcome. Therefore there was limited data available for an exploration of themes associated with poor outcome.

The study design was confined to exploring the views of participants receiving the study intervention. There would have been value in also exploring the views of

participants in the control group who received standard physiotherapy, patients who were unaccepting of the diagnosis and those that refused to participate in the study.

My identity as a specialist physiotherapist and perceived connection to the study clinicians is a potential limitation. This may have influenced the way participants answered questions. For example, participants may have felt obliged to be more complementary about the intervention and less inclined to disclose information that may suggest poor outcome. In addition, my background as a physiotherapist with experience in FMD will have influenced the way I interacted with participants, the way I led the interview and interpreted the responses.

My connection with the study intervention is a potential conflict of interest and source of bias. This bias was minimised by the qualitative analysis team consisting of two additional researchers who were independent of the intervention.

An important limitation is the transferability of the findings. Extrapolating the findings beyond the highly selected participants of this study should be done with caution. The particular characteristics of this cohort are patients attending a tertiary movement disorder clinic specialising in FMD, who met the eligibility criteria of the feasibility study. Therefore, the findings may not apply to patients who are only seen in psychiatric settings or primary and secondary care, those with high levels of comorbid pain, fatigue or psychopathology, and those who have more significant disability (i.e. disability to the extent that assistance is required for toileting). A final limitation was the relatively short follow-up interval of six months.

12.10 Future Research

Future studies should seek to explore the views of a greater diversity of patients, with consideration of age, gender, ethnicity, and socioeconomic class. Studies exploring characteristics and views that may account for good and poor treatment outcomes would be a valuable avenue of future research, with implications for refining interventions and helping to direct patients towards the most suitable treatment option. Data from patients who receive standard (non-specialist) physiotherapy

treatment may provide insights into the additional benefits of the study intervention or indeed, absence of additional benefit. The views of physiotherapists treating patients with FMD would be important to understand if the intervention were to be rolled out across the NHS.

The non-physical positive impacts of the intervention found in this study, such as reduced threat from symptoms, could be validated in future research and used to inform the creation of outcome measures specifically for FMD.

12.11 Conclusion

The participants in this study reported high levels of disability and distress associated with having FMD. They had had multiple previous interactions with HCPs, which they generally regarded as unsatisfactory and which was usually associated with receiving psychological explanations for their symptoms. Participants rejected such psychological explanations for a number of reasons. Firstly, they were unable to reconcile their physical experiences with a psychological explanation. Secondly, many interpreted psychological explanations as meaning the doctor had not believed that they had a real problem. This was related to various pejorative stereotypes of mental illness. Thirdly, some felt that “going down the psychological route” was fraught with danger and was unlikely to lead to recovery. And finally, a perceived negative attitude of clinicians towards problems associated with psychological factors left some participants feeling unworthy and ashamed. A novel finding of this study was that there was less resistance to considering a role for psychological factors as part of FMD after the participants had completed the intervention. This appeared to be related to the participants gaining a broader biopsychosocial understanding of their problem, where psychological factors could be considered as part of their problem, but not necessarily the cause.

The study intervention was highly praised by the participants and was associated with reports of improvement to their physical symptoms. Perhaps somewhat unexpectedly, most participants considered having an improved understanding of their problem was the most valuable treatment outcome. This perception was associated with feeling less

distressed by their problem, a sense of self-reliance and feeling hopeful for the future. The specific biopsychosocial explanation for their symptoms that was part of the study intervention appeared to resonate with the participants. The explanation appeared to give legitimacy to the participants' illness experiences, which may have been important for engaging them in the specialist physiotherapy intervention and future self-management.

Only one participant reported a complete resolution of their motor symptoms, the remaining participants reported gradations of improvement from no change to near-resolution. A prominent barrier to recovery was the presence of comorbid symptoms, which included fatigue, pain, migraine and other illness.

The clinical implications from this study include the importance of providing a diagnostic explanation for symptoms which patients are able to reconcile with their problem. In this instance, this meant focusing on physical and biological factors, with secondary reference to psychological factors. The study intervention was found to have a positive impact on the participants' lives at six months; in particular participants felt less concerned and more in control of their health problem. An implication for future research concerns measuring treatment outcome in terms of non-physical changes, such as levels of understanding of the problem and changes in illness-associated distress.

Chapter 13 Final Thesis Discussion and Conclusions

13.1 Introduction

In this thesis, I have explored a specialist physiotherapy-based intervention for FMD using a mixed methods research approach. A randomised feasibility study was conducted with the primary aim of determining the feasibility of testing the intervention in an RCT. A longitudinal qualitative study of patients receiving the intervention was embedded into the feasibility study, providing insights into how the intervention worked, as well as the lived experience of FMD and its clinical implications. Feasibility was demonstrated with high rates of participant recruitment, retention and intervention acceptability. The clinical outcomes were promising, suggesting that an appropriately powered RCT has a good chance of demonstrating clinical and health-economic effectiveness. This chapter will draw together the research findings with a synthesis of the quantitative and qualitative data.

13.2 Summary of Findings by Chapter

The topic of FMD was introduced in Chapter 1. FMD was described as a multifactorial problem affecting movement that is best understood with a biopsychosocial framework. It was argued that the awareness and understanding of FMD was surprisingly limited when it is seen in the context of the frequency with which patients present and the burden of symptoms to the patient and society.

In Chapter 2, I argued that the historical context to some extent explains the low levels of understanding, interest and status of FMD within the health care system. FMD was embraced by neurology, under the influence of the French neurologist Charcot in the late 1800's, before moving towards psychiatry under the influence of Sigmund Freud and his contemporaries. However, since the "heyday of hysteria" in the late 1800's, both neurologists and psychiatrists have been notoriously uninterested in FMD, leaving patients in a no-mans-land.¹⁰⁴ Concerns over malingering and the risk of missing organic disease are thought to have led to a virtual disappearance of the diagnosis of FMD from medicine. A resurgence of scientific interest started in the mid

1990's, bolstered by findings from functional imaging studies. Today, the scientific literature appears to be increasing exponentially, as if making up for lost time. This surge of scientific interest may appear as if history is repeating itself, echoing the late 19th century. Hopefully this time around we will learn from the past and avoid the neurology-psychiatry all-or-nothing divide, which I have argued, stunted the development of physiotherapy for FMD and the holistic treatment of FMD in general. There is reason to be hopeful, multidisciplinary research and treatment teams for FMD are starting to become the norm. This has led to fruitful collaborations between neurology, psychiatry and rehabilitation specialists, producing outputs such as the Stepped Care approach to the treatment of functional neurological symptoms, published by NHS Scotland,²¹⁴ and consensus recommendations for physiotherapy practice.⁴¹

Chapter 3 is a systematic review of the literature for physical rehabilitation for FMD. Only 35 studies inclusive of physical rehabilitation were found representing treatment of 564 people; of whom 79% were women, the average age was 35, and the average symptom duration was 3 years. Overall the quality of the evidence for physical rehabilitation was considered low due to the limitations in the study methodologies. However, the reported results were encouraging. There is controlled evidence from one study that physical based rehabilitation can provide at least short term improvement for patients with functional gait disorders with a symptom duration less than 5 years. There is also uncontrolled evidence from several large cohort studies that treatment involving physical rehabilitation is beneficial for the majority (55-72%) of patients with mixed FMD symptoms selected for treatment, and treatment effects have been reported to last up to two years. However, it appears that most patients remain symptomatic to some extent after rehabilitation and return to work is rare. Measures of physical function tended to show medium effect sizes, while measures of mental health tend not to change. In summary, the major limitation of the literature is the absence of controlled evidence for rehabilitation beyond four weeks. There are multiple unanswered questions including: What are the most important ingredients of rehabilitation? Which are the most suitable outcome measures? What are the most suitable treatment parameters in regards to setting, duration and intensity? And, how generalisable is the literature to the NHS?

The important first stages of developing and evaluating a complex intervention, according to the MRC, is to ensure the intervention has a coherent theoretical basis that it is described in full and is implementable in a clinical environment. This is the focus of Chapter 4. A theoretical, evidence based model for FMD which is amenable to physiotherapy is described. The model is biopsychosocial in design, but emphasises the biological sphere, which provides a specific rationale for physiotherapy. The key mechanisms by which physiotherapy intervention are proposed to affect change are: a reduction of self-focused attention during movement; and a change in the patient's belief and expectation that their movement will be abnormal.

The randomised feasibility study of the specialist physiotherapy intervention is reported in Chapters 5 to 7 (methods, results and discussion). In short, 60 consecutive patients with FMD who met the selection criteria were randomised to receive either the study intervention or a control consisting of a referral to community neuro-physiotherapy. The feasibility of conducting a trial was demonstrated by high rates of recruitment, retention and the acceptability of the intervention, with no reported serious adverse events. The clinical and economic outcomes were promising, in that the intervention was associated with a moderate to large treatment effect size across a range of physical and quality of life outcome measures. Together with demonstrated feasibility, the positive trial outcomes further support the progression to a large scale, definitive trial.

The feasibility study provides a template for the design of a definitive trial. The inclusion criteria appeared to work well. While only 32% of patients with FMD seen in the neurology movement disorders clinic where recruitment took place met the eligibility criteria, recruitment was completed in nine months, which reflects the high volume of patients suitable for physiotherapy treatment and meeting eligibility. The high rate of intervention-patients reporting improvement at six months (72%), suggests that patients unsuited to physiotherapy were appropriately excluded. The most suitable primary outcome measure for a large RCT of the intervention was found to be the SF36 Physical Function and the study provides data for an RCT sample size calculation. The CSRI was found to have limitations, in that there was insufficient data to accurately estimate service use costs and changes in engagement with employment.

These issues should be addressed by adapting the CSRI in any future studies. An employment specific questionnaire, such as the Work Productivity and Activity Impairment Questionnaire may be a valuable addition for health economic analysis.¹⁸⁹ Based on the experience of uneven rates of recruitment between the intervention and control groups, a future study should also consider block randomisation.

The qualitative study methods, findings and discussion are reported in Chapters 8 to 12. Data from interviews conducted prior to treatment provided insights into the qualitative study participants' views and experiences of developing FMD and accessing the health care service. Common themes emerged around a lack of understanding of what was wrong and resistance to psychological explanations for their symptoms. Data from interviews conducted after treatment and at six months provide insights into how the participants engaged with the study intervention and the impact that the intervention had had on their lives. A central theme related to gaining understanding of their problem ran through the interviews at all three time points. The lack of such understanding was implicated in the participants' ongoing problems prior to treatment. Post-treatment, most participants felt they had gained an understanding of their problem, which they perceived to be the most valuable outcome of treatment, despite most also reporting improvement to their motor symptoms. At six months follow-up this understanding remained the most important outcome and was associated with positive changes, such as a perception of greater self-reliance in managing their movement problem, reduced concern over their movement problem and hope for the future. The study findings could help to refine the intervention, for example, providing greater support during the transition from treatment to discharge with earlier follow up and bolstering self-management skills. How this is best achieved would be a valuable focus for future research.

13.2.1 Limitations of the Research

The limitations of both the feasibility and qualitative study are discussed in detail in the relevant chapters. A limitation that warrants repeating is that the clinical trial was designed to assess feasibility; therefore interpreting clinical outcomes as controlled evidence for effectiveness would be extrapolating the data beyond its value. The

limiting factors include that the study was not powered to detect a treatment effect, a primary outcome measure was not predefined and assessment was not blinded. However, given the lack of clinical trials in FMD, the feasibility study outcome data make a significant contribution to the evidence base that supports the use of specialist physiotherapy for FMD. The findings should, however, be interpreted with caution as generalisability is not assured, due to the single recruiting centre, and the neurologist and physiotherapist had a special interest in FMD, which may not always be the case if the intervention were to be rolled out across the NHS.

13.3 Synthesis of Quantitative and Qualitative Data

13.3.1 Outcome Measurement

An important aim of the research projects included in this thesis was to identify a suitable primary outcome measure for a RCT of physiotherapy for FMD. The SF36 Physical Function domain was found to be the most suitable candidate from the measures assessed in the study. It showed a large effect size, which appeared to be a clinically important change; it had the advantage of being a self-report measure, which is less likely to be affected by the variable nature of FMD severity over short periods of time (associated with self-focused attention); and was applicable to patients independent of symptom location (i.e. symptoms affecting gait, upper limb function or posture). Measures of gait impairment had a larger effect size but were not necessarily applicable to patients with only upper limb or head and neck symptoms. The qualitative data supports the relevance of the SF36 Physical Function to the impact of FMD on patients and change with treatment. The SF36 Physical Function domain considers mobility and activities of daily living requiring upper limb dexterity (bathing and dressing, vacuum cleaning and carrying groceries). In the qualitative interviews, the participants reported that physical symptoms were the primary problem, causing disability which resulted in secondary distress. In post-treatment interviews, the qualitative study participants often described their improvement in terms of their ability to walk and access their environment.

In the qualitative interviews, it was common for participants to report that the most valuable outcome of the intervention was having a greater understanding of their problem. This perception of greater understanding was also found in some of the outcomes from the feasibility study. The “Coherence” dimension of the B-IPQ asks, “How well do you feel you understand your illness?” with answers ranging from 0 (don’t understand at all) to 10 (understand very clearly). The intervention group mean scores were 6.2 (SD 2.6) at baseline, 8.8 (SD 1.6) post-treatment and 7.8 (SD 1.9) at six months, with little change in the control group 4.9 (SD 3.1), 5.7 (SD 3.5), 5.9 (SD 3.1) (see Appendix 7, page 240). Given the thesis finding that understanding of the problem appears to be important in improvement of FMD, an assessment of patients’ understanding should be considered in future interventional studies.

The qualitative data may help to explain the feasibility study finding of an increase in the number of cases of anxiety in the intervention group (and not the control group) at six months (n=4 at baseline, n=10 at six months). The increase appeared counterintuitive, given that the intervention group reported significant improvement on measures of physical disability and a change in the total B-IPQ score suggestive of a reduction in the perceived threat posed by illness. Possible explanations for the increased cases of anxiety are that it is a chance occurrence in the data, that some participants became more anxious, or that the intervention helped some participants develop insight into their problem and possible links to anxiety. The qualitative data support this last explanation. Several interview participants reported in post-treatment interviews, that they had come to a realisation that they were anxious or that anxiety was part of their problem. This realisation may have led them to score themselves higher on the HADS. Recognition of the role of anxiety may be a helpful outcome of the physiotherapy intervention. It may help some patients with FMD to more correctly attribute sensations and involuntary movement to the sensorimotor consequences of anxiety, rather than symptoms of disease. It may also help some patients who could benefit from a psychological intervention engage with treatment when they are initially reluctant to do so. These hypotheses are consistent with data from a large cohort study that found that non-attribution of the symptoms of FMD to psychological factors predicts poor outcome, with an odds ratio of two.¹⁵³

Both the feasibility study and qualitative study found that the intervention was associated with decreased distress associated with symptoms (reduction in B-IPQ total score and interview reports of feeling at more at ease with ongoing symptoms). The qualitative data provided possible reasons for this finding. This included the perception of knowing what was wrong, knowing how to manage the problem, resolution of the uncertainty of the prognosis, and resolution of the perception of feeling abandoned by the NHS, despite having a problem that was debilitating and at times frightening.

The health economic analysis found that the incremental cost per QALY gained with the intervention was £12,087, significantly less than the £20,000 upper limit of cost effectiveness suggested by NICE.¹⁷⁷ The qualitative data gave weight to the probability of cost effectiveness. The interview participants commonly reported acceptance of the diagnosis and a sense of self-reliance which included an expectation of less dependence on HCPs. This suggests the potential for a reduction in health service utilisation, such as seeking further investigations, second opinions, and alternative treatments.

13.3.2 How did the intervention work?

Seventy-two percent of the intervention group participants reported improvement in their symptoms at six months. This corresponded with significant improvement in measures of physical function (e.g. SF36 Physical Function domain and measures of balance, mobility and upper limb function). Data from both the feasibility study and qualitative study provide clues as to how the changes may have come about.

In the feasibility study discussion, I suggested that the reduction in B-IPQ total score in the intervention group may indicate a reduced threat of symptoms and that this may be an important mediator of change. The qualitative data supported the suggestion of reduced threat from symptoms. These findings are consistent with intervention-mechanism studies of chronic fatigue syndrome and persistent pain treatments.^{209,210}

Data from the feasibility and qualitative study suggest that a perception of having an increased understanding of the problem may be an important mediator of

improvement and may help by reducing the threat of symptoms. Such understanding appeared to be associated with self-efficacy and hopefulness, which may also be important factors that mediate improvement with this intervention.

Given the association of FMD with psychological disturbance, it would be reasonable to hypothesise that changes in mental health scores may mediate improvement. However, this was not found in the data. Measures of mental health did not change with treatment. However, this may be related to the subclinical levels reported at baseline or reflect limitations with the assessments.

13.3.3 What Constitutes a Good Treatment Outcome?

The feasibility and quantitative studies raise the question: what constitutes a good treatment outcome? Standard definitions of FMD usually highlight that symptoms are not related to structural damage or disease and therefore there is a potential for complete symptom resolution. This may lead to unrealistic expectations, and a perception that lack of symptom resolution is a treatment failure. In the current studies, symptom resolution at six month follow-up was rare. This is also the case for other interventional studies.⁷⁵ However, the current study found improvements in physical function, perceived understanding, distress and measures of cost-utility. These findings emphasise the importance of using sensitive outcome measures and measuring multiple domains of health.

The challenge for physiotherapists treating patients with FMD may be in recognising the value of non-physical treatment outcomes, such as reduced illness distress and cost-benefit, in situations where there has been no change in disability. The expectation that patients should get completely better and that failure to do so constitutes failure, is unrealistic and arguably suggests an underlying uncertainty regarding the genuine nature of functional disorders.

13.4 Thesis Conclusions

FMD is a common cause of disability and distress, for which there are few treatment options available. The findings from the studies described in this thesis support the use

of a specialised physiotherapy-led intervention for FMD and add to the body of evidence for specialist physiotherapy for FMD. The key components of the intervention were education about the diagnosis (provided in neurology and physiotherapy sessions), demonstrating to the patient how they can move normally when distracted, using distraction to develop strategies that normalise movement, and developing a long term personalised symptom management plan.

The explanation given to patients about the diagnosis was the important first step in treatment. The explanation was biopsychosocial in scope, but emphasis was placed on the biological domain. The explanation resonated with most of the study participants and appeared to be important in helping them to accept the diagnosis and engage with treatment and self-management. The finding that many participants were less resistant to acknowledging a role for psychological factors in their problem after treatment may be significant in the intervention success and may have clinical implications for designing multidisciplinary treatment pathways (with recognition that generalisability beyond patients attending tertiary neurology clinics is not assured).

The research identified several factors that may mediate a good treatment outcome. These were: (i) a change in illness belief; (ii) a perception of improved understanding of their movement problem; (iii) reduced threat value of symptoms; (iv) a perception of increased self-efficacy; and (v) hopefulness. These potential mediators of change could be targeted in future interventions; however confirmation of their role in mediating change with further research is needed. Consecutive days of treatment and high intensity treatment appeared to be an important element of the intervention that warrants further exploration.

The intervention cohort improved despite an average symptom duration of 5 years. Given the substantial evidence that symptom chronicity is associated with a poor prognosis,⁷⁰ it is possible that if the intervention was delivered earlier in the course of the participants disorder, it may have been more effective.

This thesis has explored a specific physiotherapy intervention. Physiotherapy is only one of a number of different treatment approaches that may be effective for patients

with FMD. Other potentially effective treatments available in the NHS include specific types of psychological therapy, occupational therapy and multidisciplinary rehabilitation. In addition, there may be other interventions that are not available on the NHS and cultural specific treatments that may also prove to be effective. Given the heterogeneity of patients with FMD, it follows that a variety of different treatments are necessary to suit the needs and preferences of people with this diagnosis. A key priority therefore in future work is to identify criteria that predict which patients are most likely to benefit from each of these different interventions, to improve the effective triage of patients into treatment.

13.4.1 Progression to a definitive trial

The findings of this thesis support the progression from the feasibility study to a pragmatic multicentre RCT. Additionally, the findings should inform the design of this trial. See Appendix 8: Reflections on the Methodology (page 242), for a list of methodological considerations for a future trial.

Refining, standardising and documenting the intervention so that it is both implementable and reproducible in a clinical trial will be a challenging but necessary step. A useful approach to achieve this objective is Intervention Mapping^{215,216}. Intervention Mapping is a systematic approach to intervention development that enables integration of theory, research evidence and practical implementation issues. Additionally, Intervention Mapping aims to link intervention components to theoretical mechanisms of change, which may help to understand the intervention and improve outcomes. Utilisation of Intervention Mapping is likely to make the scaling up of the single-site-intervention described here to a multicentre trial intervention more successful.

Given the high satisfaction ratings and positive treatment outcomes, a future trial should aim to reproduce as closely as possible the conditions of the study intervention. An addition that may improve the intervention is a six week physiotherapy follow up session.

The SF36 Physical Function domain was found to be the most suitable primary outcome measure. The adjusted difference between groups for this measure was 19.8 (95% CI 10.2, 29.5). When calculating a sample size for a future pragmatic trial, a more conservative group difference should be selected, for example a difference of 10 points.

Finally, a future study design could consider a secondary mediation analysis to explore potential treatment mechanisms. Potential mediators that could be investigated using standardised questionnaires include: expectation of recovery, perceived level of understanding of the health problem, distress caused by symptoms, and perceived level of threat posed by symptoms. Reproducing the study intervention in a pragmatic multicentre trial will be challenging. However, my experiences over the past three years with inspiring mentors, collaborators and patients have left me feeling confident that it is possible and motivated to try.

Appendix 1 Quality Appraisal of Studies with Subject Numbers of 10 or More

Quality Appraisal Checklist for Case-Series Studies²¹⁷ (The authors of this tool state that empirical evidence does not support defining quality by a cut-off score. Additionally the validity of scoring each criterion with equal weighting is yet to be determined)	Demartini et al	Jordbru et al	Czarnecki et al	Nielsen et al	Moene et al	Shapiro & Teasell	Matthews et al	Heruti et al	McCormack et	Saifee et al	Ferrara et al	Dalocchio et al	Espay et al	Speed
1. Was the hypothesis/aim/objective of the study clearly stated?	1	1	1	1	1	1	1	1	1	1	0	1	1	1
2. Was the study conducted prospectively?	1	1	0	1	1	1	1	0	0	0	1	1	1	0
3. Were the cases collected in more than one centre?	0	0	0	0	1	0	0	1	0	0	0	0	0	0
4. Were patients recruited consecutively?	1	1	1	1	1	1	1	1	1	1	0	1	0	1
5. Were the characteristics of the patients included in the study described?	1	1	1	1	1	1	1	1	1	1	1	1	1	1
6. Were the eligibility criteria for entry into the study clearly stated?	0	1	1	1	1	0	0	1	1	0	1	0	0	1
7. Did patients enter the study at a similar point in the disease?	0	0	0	0	0	0	0	1	0	0	0	0	0	0
8. Was the intervention of interest clearly described?*	1	1	1	1	1	1	1	0	1	1	1	1	1	1
9. Were additional interventions clearly described?														
10. Were relevant outcome measures established a priori?	1	1	0	1	1	0	1	0	1	1	1	1	1	1
11. Were outcomes assessors blinded to the intervention that patients received?	0	0	0	0	1	0	0	0	0	0	1	1	1	0
12. Were the relevant outcomes measured using appropriate objective/subjective methods?	1	1	0	1	1	0	1	0	1	1	1	1	1	1
13. Were the relevant outcome measures made before and after the intervention?	1	1	0	1	1	0	1	0	1	1	1	1	1	1
14. Were the statistical tests used to assess the relevant outcomes appropriate?	1	1	1	1	1	0	1	0	1	0	1	1	1	1
15. Was follow-up long enough for important events and outcomes to occur?	1	1	1	0	0	0	0	0	0	0	0	0	0	0
16. Were losses to follow-up reported?	1	1	0	1	0	0	0	0	0	0	1	1	0	0
17. Did the study provide estimates of random variability in the data analysis of relevant outcomes?	0	1	0	1	0	0	0	0	1	0	0	1	1	1
18. Were the adverse events reported?	0	0	0	1	0	0	0	0	0	0	1	1	0	0
19. Were the conclusions of the study supported by the results?	1	1	1	1	1	1	1	1	0	1	1	1	1	1
20. Were both competing interests and sources of support reported?	1	0	1	1	0	0	1	0	1	0	0	1	1	0
TOTAL	13	14	9	15	13	6	11	7	11	8	12	15	12	11

* Clarity of description of the interventions was considered within the scope of the trial report. However, this level of information is generally insufficient to reproduce complex interventions such as those described in these studies.

Appendix 2 Treatment Protocol

Assessment

A thorough subjective history and physical assessment is important to understand the patient's problems; to recognise predisposing, precipitating (triggering), and perpetuating factors; to identify factors that may be amendable to physiotherapy; and to create an individualised explanation of the diagnosis for the patient. Important elements include:

1. Explore the onset of symptoms and potential precipitating factors which may help to facilitate the patient's understanding of the diagnosis.
2. Compile a comprehensive list of symptoms and health related problems. For each problem, details about frequency, severity, exacerbating factors and easing factors are sought.
3. Develop an impression of the impact of symptoms on daily life by charting a typical 24 hour routine.
4. Enquire about previous experiences with treatment and health care professionals.
5. Explore beliefs and understanding of symptoms.
6. Observe the impact of symptoms on posture, movement, tasks and activities. This includes sitting, standing, transfers, gait and upper limb tasks.
7. Explore the nature of symptoms, note variations in severity with distraction, and any manoeuvres that dampen or exacerbate symptoms.

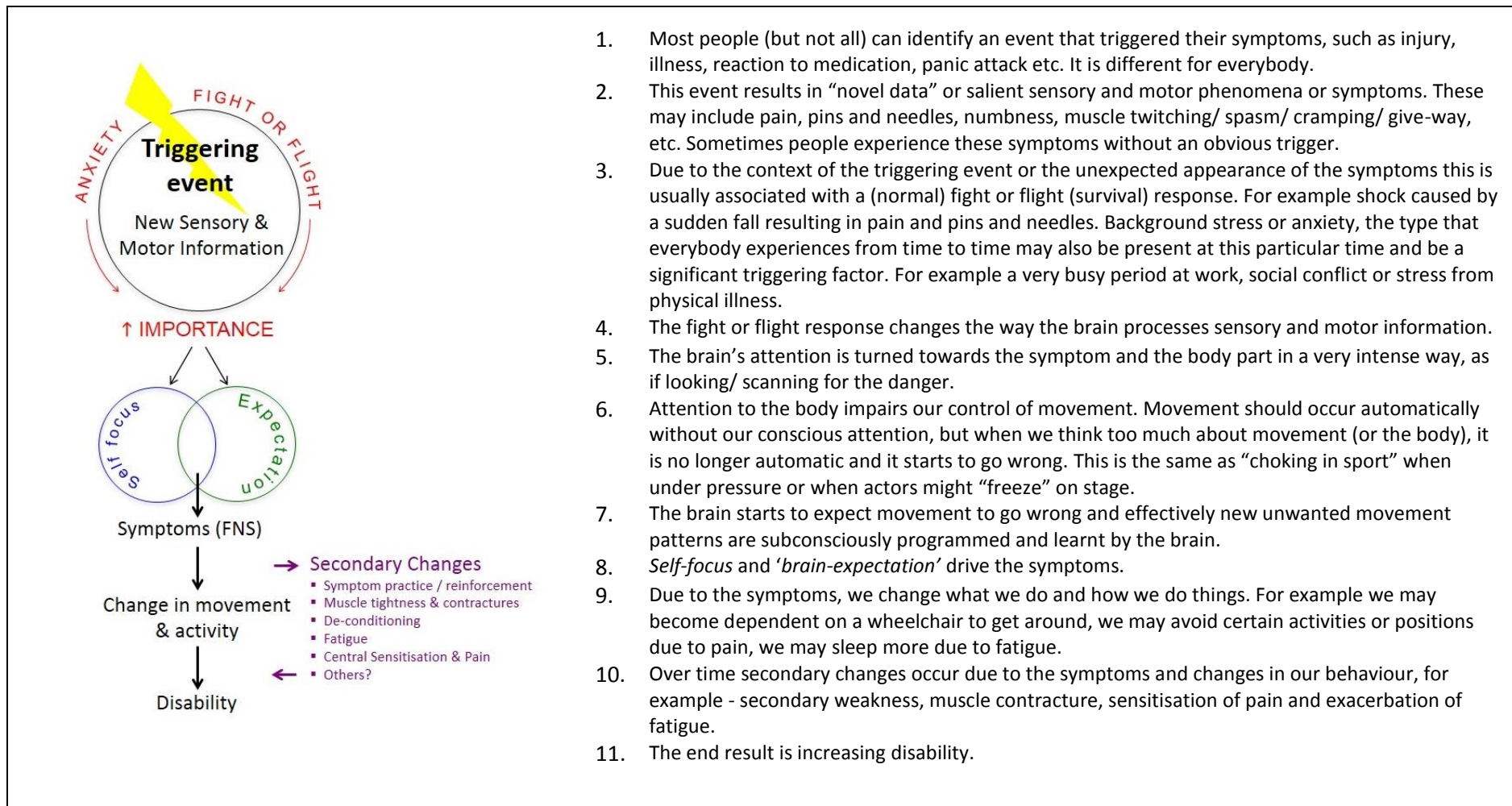
Education

Helping the patient develop an understanding and insight into their symptoms is an important first step to prepare them for treatment. This starts with a comprehensive explanation of the diagnosis by the neurologist,¹⁵⁴ which is built on during physiotherapy sessions. The educational components of treatment have four key objectives:

1. To address unhelpful illness beliefs that may form part of the mechanism driving symptoms.
2. Develop an understanding that symptoms are not caused by structural damage or a degenerative process, thereby potentially lowering the threat value of symptoms and highlighting reversibility.
3. To prepare the patient for a symptom management approach to treatment according to a biopsychosocial model of illness.²⁰² This is where the patient assumes the responsibility for understanding the diagnosis and “administering the treatment”. As opposed to a traditional biomedical model where the patient is the passive recipient of treatment.
4. To equip the patient with the knowledge and skills necessary to manage their symptoms in the longer term.

During treatment, the patient and physiotherapist should come to a shared understanding of the problem in order to collaborate on a management approach. The symptom model (see Figure 1, below) is used to explain how triggering events lead to abnormal self-focussed attention and expectations which drives symptoms; and that secondary changes occur which perpetuate the problem. A thorough individualised symptom formulation is collaboratively built around the framework of the symptom model. The formulation is used to explain the rationale for the treatment. This explanation forms most of the first physiotherapy session, and is reviewed and expanded in subsequent sessions. The patient should be given daily opportunities to ask questions and should feel safe to express concerns or doubt about the diagnosis. The patient may start to feel more confident in the diagnosis as treatment progresses

Figure 1. Developing a personalised symptom formulation. Reproduced from Nielsen et al 2015¹



Movement retraining

Movement retraining aims to restore normal movement patterns, primarily by:

1. Altering unhelpful beliefs and expectations about movement.
2. Reducing abnormal self-focus during movement and restoring “automatically” generated movement.
3. Changing maladaptive compensatory habitual postures, movement patterns and behaviours.

Movement retraining addresses unhelpful beliefs by demonstrating to the patient that normal movement can occur. This is also a powerful way to help the patient understand the diagnosis and convince them that it is correct. This starts in the neurology consultation by showing the patient their clinical signs of FMD, with an explanation that clinical signs such as Hoover’s sign and tremor entrainment demonstrate intact “wiring”.¹⁵⁴ In the physiotherapy gym, normal movement can be produced in the context of meaningful activity. The key to normalise movement is to redirect the patient’s focus of motor attention. For example a functional gait disturbance often normalises when the patient is asked to walk backwards, or to slide feet along the floor when stepping forward. Tasks that normalise movement usually involve novel or unfamiliar movements and re-direction of attention is required to achieve the task. A list of other strategies that may be helpful to normalise movement for different symptoms (and form part of movement retraining) can be found in Table 1, below. Normal movement is demonstrated to the patient with the aid of mirrors and video.

Movement retraining usually involves a sequential approach, where elementary symptom free components of movement are established, and built on in successive stages to gradually reshape normal movement patterns.^{75,128} Asking the patient to watch themselves in the mirror as they move may help to redirect attention away from their body, to their reflection. The mirror provides feedback to help shape movement and provides evidence of normal movement to the patient (which may influence beliefs and expectations). Movement retraining occurs in the context of functional

tasks, such as standing from a chair and transferring to a bed (as opposed to hip and knee flexion-extension exercises) may provide a more implicit approach to motor learning, where the movement is more automatic and the patient is less aware of the “mechanics” of movement with focus directed towards the goal of movement.

Retraining the sit-to-stand and stand-to-sit movement pattern is often an appropriate starting point. Strategies that normalise movement are developed during physiotherapy sessions and practiced. The patient is encouraged to use strategies that normalises their movement during their usual routine. This is in preference to setting aside time at home to practice physiotherapy exercises. Consolidation and generalisation of movement retraining is achieved by gradually increasing the difficulty of tasks, changing the environment (e.g. outdoors, busy environments), varying speeds and multitasking. The Figure 2 shows examples of movement retraining in action.



Figure 2. Examples of movement retraining

Table 1. Movement retraining strategies for different motor symptoms

Leg weakness	<ul style="list-style-type: none"> • Early weight bearing to trigger automatic muscle recruitment. • Side to side weight shifting in a safe environment (e.g. parallel bars/ raised plinths on either side) with attentional focus towards the rhythmical movement of centre of mass (rather than towards the legs). • Crawling in 4-point/ 2-point kneeling. • Treadmill walking with or without body weight support harness and feedback from a mirror.
Ankle weakness	<ul style="list-style-type: none"> • Anterior-posterior weight shifting often triggers dorsiflexor activity via the body's automatic ankle strategy. It is useful for the patient to see this in the mirror. • Walking backwards, or sliding feet along the floor often elicits dorsiflexors activity. • Electrical stimulation can be useful to provide the patient with the experience of muscle activation.
Upper limb weakness	<ul style="list-style-type: none"> • Weight bearing in sitting, against a wall, or in 4-point kneeling can help to switch on muscles. Crawling can also be very effective. • Stimulate automatic upper limb postural responses, for example sitting on an unstable surface such as a therapy ball resting upper limbs on a supporting surface. • Minimise habitual non-use by using the weak upper limb to stabilise objects during tasks or assist the other hand. • Practice bilateral hand tasks that are familiar or important to the patient that may not be associated with their symptoms e.g. use of mobile phone or computer tablet.
Gait disturbance	<ul style="list-style-type: none"> • Build up a normal gait from simple achievable components that progressively approximate normal walking. For example, side to side weight shift, continue this weight shift motion to allowing feet to 'automatically' advance forwards small amounts, progressively increase the step length with the focus on maintaining rhythmical weight-shift, rather than the conscious action of stepping. • Try novel movement such as walking backwards, sideways, sliding feet as if skiing. • Treadmill walking with or without a body weight support harness and feedback from a mirror. • Increase walking speed can help to 'bypass attentional focus' and tap into more automatic muscle recruitment (in some cases this may worsen walking pattern).
Upper limb tremor	<ul style="list-style-type: none"> • Training in front of a mirror seems to aid a sense of agency and control over the tremor. It can also shift attentional focus away from the internal sensory experience of the tremor. • Change habitual 'coping strategies' that often paradoxically just increase muscle tension and worsen a tremor, such as sitting on the hand or overly tensing the limb in an attempt to keep it still. • Make the movement 'voluntary' by asking the patient to actively do the tremor, then change the movement to larger amplitude and slower frequency. From here the patient can try and slow the movement down towards stillness. • Performing competing movements that require re-direction of motor attention can be useful. Shoulder rolling, smooth running of palms of hands over thighs, clapping to a rhythm or moving the symptomatic arm as in large fluid movements as if conducting an orchestra. • Teach the patient to find ways to relax and switch of muscles. EMG biofeedback can aid this. Actively contracting and relaxing muscles with progressive relaxation can be useful, as can breathing relaxation exercises.

Lower limb tremor	<ul style="list-style-type: none"> • Side to side or anterior-posterior weight shift. When the tremor has reduced, slow the weight shift to stillness. • Competing movements in sitting, such as toe tapping or gently sliding feet forwards and backwards on the floor. • Ensure even weight distribution when standing. This can be helped using weighing scales or a mirror for feedback. • Changing problematic habitual postures. For example patients with a 'bouncing knee tremor' often sit with only the toes in contact with the floor (which drives the tremor via the clonus reflex). Encouraging full foot contact with the floor usually reduces or resolves this type of tremor.
Fixed Functional Dystonia	<ul style="list-style-type: none"> • Change habitual sitting and standing postures to prevent prolonged periods in end of range joint positions and promote postures with good alignment. • Teach positioning strategies to turn down overactive muscles in sitting and lying e.g. a limb 'hanging' in mid air is much more likely to be active than if in contact with a supporting surface. Cushions or folded towels may be needed to bring the supporting surface up and bridge gaps. • Normalise movement patterns (e.g. sit to stand, transfers, walking) with an external or altered focus of attention (i.e. not the dystonic limb). • Gentle weight-bearing (as tolerated) though the fixed limb can often help to normalise muscle activity around a joint. Sometimes gentle 'on-off' weight-bearing motions can help turn down dystonic muscles, probably via reciprocal inhibition when antagonist muscles are activated. • Discourage unhelpful protective avoidance behaviours and encourage normal sensory experiences (e.g. wearing shoes and socks, weightbearing as tolerated, not having their arm in a 'protected' posture across their lap). • Consider examination under anaesthetic/ sedation, especially if completely fixed or concerned about contractures. • Electrical muscle stimulation to the antagonist muscles can sometimes be helpful in achieving improved limb posture during therapy.
Jerks/ Myoclonus	<ul style="list-style-type: none"> • Address habitual postures that might be feeding into tense/ overactive muscle groups e.g. a patient with an upper limb jerk, might tend to hold the shoulder in a protracted posture causing overactive/ shortened pectoral muscles. • Practicing opposing movements that interfere with the abnormal movement pattern. E.g. if a patient has abdominal 'up and down' jerks, then performing little trunk rotations, or deep thoracic expansion breathing might interfere with the jerk. • Desensitise and lengthen overactive/shortened muscles through therapeutic resting postures (rather than direct stretching) e.g. a patient with functional cervical dystonia towards the left, might be able to achieve therapeutic lengthening of the dystonic muscles in right sided lying. • Relaxation and sensory grounding can be useful, especially if the patient can identify warning signs or certain situations that bring on their symptoms.

Self-management & the Treatment Workbook

The self-management approach to treatment recognises that FMDs are often chronic conditions with multiple contributing factors that can require ongoing attention in order to sustain improvement and make further progress. Self-management ensures the patient is empowered to put knowledge and skills into practice to improve their health and wellbeing. It is an ideological shift away from the biomedical model of illness, where patients are passive recipients of treatment to patients being active partners in the effective management of their health.²¹⁸

The Treatment workbook is designed to support self-management. The patient and physiotherapist collaboratively complete the workbook throughout the five days of treatment. The contents of the workbook include:

1. An explanation of the diagnosis using the physical-biased symptom explanatory model, with reference to the patient's personal experience.
2. Goals for physiotherapy.
3. Space for the patient to reflect on treatment sessions, noting what was covered, things that were easy, things that were difficult and new insights.
4. A list of symptom management strategies that were developed and practiced during the physiotherapy programme.
5. Explanation and management plan for chronic pain (if relevant).
6. Explanation and management plan for fatigue (if relevant).
7. Understanding difficulties with memory, concentration and attention.
8. Markers of progress, such as achieved goals and scores on outcome measures.
9. A symptom management plan that aims to sustain and progress improvements made with treatment. This may include a list of therapeutic activities and behaviours; un-therapeutic behaviours to avoid; when and how to use specific symptom management strategies; activity-pacing plan with progressions; a plan to return to usual activities; etc.
10. What to do on difficult days and setbacks.
11. An end-of-treatment summary, including the most relevant information and management strategies, and a list of achievements.

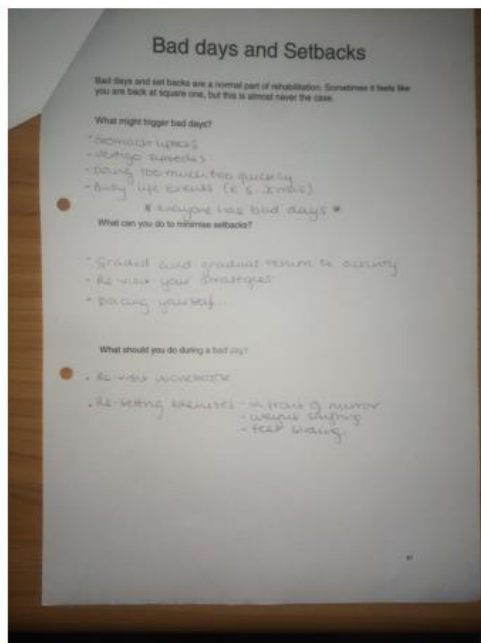
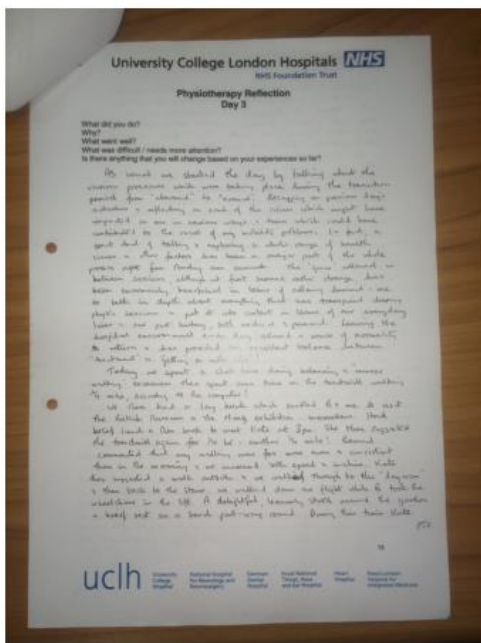
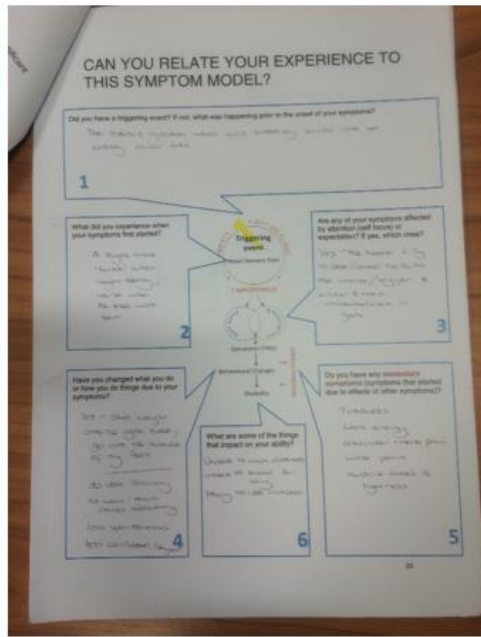
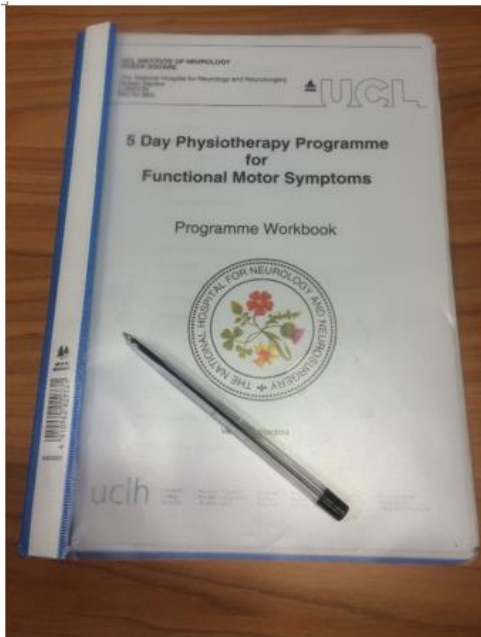


Figure 3. Examples of the workbook

Appendix 3 Patient Information Sheet: Feasibility Study

UCL INSTITUTE OF NEUROLOGY
QUEEN SQUARE

THE NATIONAL HOSPITAL FOR NEUROLOGY AND
NEUROSURGERY
QUEEN SQUARE
LONDON WC1N 3BG



PATIENT INFORMATION SHEET

Version 2, 8/03/2014

Feasibility Study of Physiotherapy for Functional Motor Symptoms

We are inviting you to take part in a research study. Before you decide it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and discuss it with others if you wish. Please ask us if there is anything that is not clear or if you would like more information. Take your time to decide whether or not you wish to take part.

Thank you for taking the time to read this information sheet

What is the purpose of the study?

Physiotherapy is often prescribed for people with functional motor symptoms (nervous system symptoms such as weakness, tremor or muscle spasms that are not due to structural problem of the nervous system) but there is little scientific evidence to show that it may be effective. One of the reasons why the evidence is limited is that there are many unanswered questions about how best to conduct a fair scientific trial of physiotherapy in this condition. In this study we aim to answer these questions, which will enable us to conduct a full trial.

Do I have to take part?

It is up to you to decide whether or not to take part. You are free to withdraw at any time. If you decide to take part we will ask you to sign a consent form indicating your willingness to participate in the study. A decision to withdraw, or a decision not to take part, will not affect the standard of care you receive. If you chose not to take part or to withdraw from this study you will be given the option of being referred to your local physiotherapy service.

What will I have to do if I take part?

If you choose to take part you will be asked to complete a fairly brief questionnaire booklet to give us more information about your health – we can help you complete this if need be. You will be asked to complete a balance test and if able, a timed 10 metre walk. We will also ask to film 30 seconds of your walking and movement with a digital camera. This will take about 1 hour in total. You will then be allocated by chance to one of 2 groups.

The video and all other information you provide will remain strictly confidential, will be stored securely and will only be seen by the research team.

Treatment groups

It is important to have 2 treatment groups in the study in order to compare peoples' experience of routine physiotherapy with the research intervention.

Group 1

Participants in this group will be referred to their local physiotherapy service. If you are currently receiving physiotherapy or have recently been discharged, a letter summarising your consultation with Dr Edwards will be sent to your local physiotherapy team. You will receive a copy of this letter. We will ask you to come back in approximately 6 weeks and again at 6 months at a time convenient to you to redo the questionnaires, video, balance and walking assessment.

Version 2, 8/03/2014

UCL Institute of Neurology • National Hospital for Neurology & Neurosurgery • Queen Square • London WC1N 3BG

☎ +44 (0)20 3448 3718 • www.ion.ucl.ac.uk

Director: Professor A Thompson MD, FRCP, FRCPI

Institute Secretary: R.P Walker BSc (Econ)

The Institute of Neurology promotes teaching and research of the highest quality in neurology and the neurosciences.

You will also be given the option of being referred to an inpatient treatment programme. This will involve meeting the inpatient team for an initial appointment to see if their programme is suitable for you. The waiting list to attend this programme is currently longer than 12 months.

Group 2

Participants allocated to group 2 will receive physiotherapy over 5 consecutive days at The National Hospital for Neurology and Neurosurgery. They will be asked to redo the questionnaires, video, balance and walking assessment on the 5th day and again at a follow up appointment in 6 months time.

If participants in this group continue to experience significant symptoms following the physiotherapy programme, they will be referred to the inpatient treatment programme.

How do you decide which group I am in?

This study is what we call 'randomised'. At the moment we do not know whether usual physiotherapy treatments will be more helpful in improving your symptoms than the intensive 5 day physiotherapy programme we have developed. We need to compare the two by selecting people to go into groups receiving one of these two approaches. A computer will be used to select names at random and put them into one of the two groups. The computer has no information about people, so selection is by chance. You will have a one-in-two chance of being in either group.

How long will I be in the study?

If you agree to take part you will be in the study for 6 months. After this you will return to standard NHS care. If you have been involved in previous clinical research you may wish to reconsider your participation considering the extra time commitment.

Expenses and Payments

We will reimburse any travel expenses that you incur that are in addition to your normal NHS care.

What are the benefits of taking part?

We cannot promise that the study will help you, but the information from this study may help improve future physiotherapy treatment provision of functional motor symptoms. We also hope to increase awareness of functional motor symptoms amongst physiotherapists and other health care professionals.

What are the possible risks?

Physiotherapy is considered a standard and safe treatment for people with functional motor symptoms. We believe there are no additional risks in participating in this research study.

What if there is a problem?

We do not anticipate anybody to come to any harm by taking part in this study. If you have a concern about any aspect of this study, you should ask to speak to the chief investigator (Glenn Nielsen, 02034483718) who will do his best to answer your questions. If your concerns are not resolved you could contact Dr Edwards (Tel 02034484295). If you remain unhappy and wish to complain formally, you can do this via the NHS Complaints Procedure. The Patient Advice and Liaison Service (PALS) can support you through this process. Contact PALS on 020 3447 3042.

Will my taking part be kept confidential?

Yes. We will follow ethical and legal practice and all information about you will be handled in confidence. You will be allocated a unique study code in order to keep any information stored about you confidential. All research staff not directly involved with you will only know you by this code. All information will be stored on password protected computer databases and will only be accessible to the research team and potentially by regulatory authorities for auditing

and monitoring purposes. When the results of the study are reported, individuals who will have taken part will not be identified in any way.

Will my GP be informed about my participation?

Yes, we will send a letter to your GP informing them of your participation. Any medical or physiotherapy letters or reports will be copied to you and your GP.

How will the information be used?

The findings of this research will be published in scientific journals and presented at conferences. You will be informed about the results by the researchers, who will provide you with a one page A4 lay summary and will let you know how to access the scientific publications. The lay summary will also be sent to the patient support group FND Hope [www.fndhope.org].

Who is funding this research?

Funding for this research is provided by the National Institute for Health Research (NIHR).

Who has reviewed this study?


This study has been reviewed and approved by the National Institute for Health Research (NIHR). All research in the NHS is looked at by independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given favourable opinion by the City Road & Hampstead Research Ethics Committee.

Contact details

If you have any questions or concerns, please contact –
Glenn Nielsen (chief investigator).
Tel: 02034483718
Email: g.nielsen@ucl.ac.uk

This research project is registered on the following database –
<http://www.clinicaltrials.gov>

Appendix 4 Consent Form: Feasibility Study

<p>UCL INSTITUTE OF NEUROLOGY QUEEN SQUARE</p> <p>THE NATIONAL HOSPITAL FOR NEUROLOGY AND NEUROSURGERY QUEEN SQUARE LONDON WC1N 3BG</p>		
CONSENT FORM		
Study Number: 14/LO/0573 ClinicalTrials.gov ID: NCT02275000		
Title of project: Feasibility Study of Physiotherapy for Functional Motor Symptoms		
Please initial box		
1. I confirm that I have read and understand the information sheet dated 8/03/2014 (version 2) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	<input type="checkbox"/>	
2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my medical care or legal rights being affected.	<input type="checkbox"/>	
3. I understand that relevant sections of my medical notes and data collected during the study, may be looked at by individuals from University College London, from regulatory authorities or from the NHS Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records.	<input type="checkbox"/>	
4. I agree to my GP being informed of my participation in the study.	<input type="checkbox"/>	
5. I consent to being videoed as part of the assessment process in this study.	<input type="checkbox"/>	
6. I agree to take part in the above study	<input type="checkbox"/>	
Please send me a summary of the project outcomes to		
Address: _____		

OR Please do not send me a copy of the project outcome <input type="checkbox"/>		
_____ Name of Participant	_____ Date	_____ Signature
_____ Name of Person taking consent	_____ Date	_____ Signature
When completed, copy for participant, copy for research file, original to be filed in medical notes.		
Version 2, 8/03/2014		
UCL Institute of Neurology • National Hospital for Neurology & Neurosurgery • Queen Square • London WC1N 3BG ☎ +44 (0)20 3448 3718 • www.lon.ucl.ac.uk Director: Professor A Thompson MD, FRCP, FRCPI Institute Secretary: R P Walker BSc (Econ)		
The Institute of Neurology promotes teaching and research of the highest quality in neurology and the neurosciences.		

Appendix 5 Client Services Receipt Inventory (version used in this study)

University College London Hospitals NHS Foundation Trust

Functional symptoms can affect people's lives in many ways. The following questions will help us to understand the effect on you, particularly financially. All your answers will remain completely confidential.

Firstly, please could you tell us about the health care which you have received -

1) In the last 3 months, have you used any of the services below?

Please tick 'yes' or 'no' for each line. If you answer 'yes' to any of them, please tell us how many times you used the service and, when applicable, tick if the service was private.

	GP and Practice nurse	No	Yes	Number of times
1.1	Seen GP at the surgery	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
1.2	Seen GP at home	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
1.3	Phoned GP for advice	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
1.4	Seen practice nurse at the surgery	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
1.5	Phoned practice nurse for advice	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
	Social services			
1.6	Got meals on wheels	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
1.7	Home help came around	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
1.8	Seen social worker	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
1.9	Phoned social worker	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
	Physiotherapist			
1.10	Seen at the hospital (NHS)	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
1.11	Seen at home (NHS)	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
1.12	Seen at the GP surgery or a clinic (NHS)	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
1.13	PRIVATE Physiotherapist at home	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
1.14	PRIVATE Physiotherapist Outpatient	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
1.15	PRIVATE Physiotherapist hospital	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
	Occupational therapist			Tick if private
1.16	Seen at the hospital	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂	<input type="checkbox"/>
1.17	Seen at home	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂	<input type="checkbox"/>
	Other services (e.g. alternative therapies, voluntary services)			
1.18	Please describe.....	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂	<input type="checkbox"/>
1.19	Please describe.....	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂	<input type="checkbox"/>

PLEASE CHECK YOU HAVE ANSWERED ALL QUESTIONS

2) **In the last 3 months**, have you been to hospital?

Please do not include physiotherapy or occupational therapy appointments.

Please tick 'yes' or 'no' for each one. If you answer 'yes' to any, please tell us how many times you used the service.

	No	Yes	
2.1 Been to accident and emergency (casualty)	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂	Total number of visits:
2.2 Elective hospital overnight stay	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂	Total number of nights:
2.3 Non-elective hospital overnight stay	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂	Total number of nights:
2.4 Overnight hospital stay in intensive care/ high dependency unit	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂	Total number of nights:
2.5 Had a hospital outpatient appointment	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂	Total number of appointments:.....
2.6 Day patient procedure/test	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂	Total number of tests/procedures:.....

We would now like to find out about what health costs you and others financially

3) **In the last 3 months**, what medicines have you used and how did you pay for them?

Drug Name (generic or brand)	Duration of use (days)	Daily Dose (Dose/no. per day)

4) **In the last 3 months**, have you, your relatives/friends, the NHS or social services paid for any of the following?

Please tick 'yes' or 'no' for each line and tell us how much it cost

	No	Yes	How much has this cost altogether in the last 3 months?	Who paid for this?
4.1 Employing extra help (e.g. cleaning)	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
4.2 Transport to get to healthcare appointments (e.g. to go to your GP surgery or hospital)	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
4.3 Transport to get to self-help groups	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
4.4 Health care (e.g. private or alternative treatments)	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
4.5 Special equipment (e.g. kitchen equipment)	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
4.6 Changes to your home (e.g. stairlift)	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
4.7 Financial support (please do <u>not</u> include state benefits)	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
4.8 Any other costs	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂

5) If you are currently employed, have you taken any time off work **in the last 3 months**?
Please note: Include any time taken off because of ill health or using any health services such as those listed in questions 1 & 2 (e.g. GP/hospital appointments).

- 5.1 Yes ₁ *If yes: Please give details below*
 No ₂ *If no: Please go straight to question 6*
 I have not been employed in the last 3 months ₃ *If not employed: Please go straight to question 6*

Please tell us either the number of days or the number of hours you took off in the last 3 months

	No	Yes	Number of whole working days	Or Number of hours
5.2 Took sick leave from work	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
5.3 Used your paid holiday time from work	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
5.4 Took unpaid leave from work	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
5.5 Just made up the time at work	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
5.6 Other arrangement (please describe)	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂

What is your gross income (i.e. before tax) per week? £.....

6) **In the last 3 months**, have friends and relatives helped you with tasks at home which you couldn't do?

6.1 Yes ₁ *If yes: Please tick below the tasks they helped you with and for how many hours per week.*
 No ₂

	No	Yes	Typically, how many hours per week?
6.2 Personal care (e.g. bathing, dressing)	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
6.3 Housework / laundry	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
6.4 Providing transport/taking you out	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
6.5 Preparing meals	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
6.6 Gardening	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
6.7 Shopping	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
6.8 Looking after pets	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
6.9 DIY/home improvements	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
6.10 Other (Please describe)	<input type="checkbox"/> ₁	<input type="checkbox"/> ₂
.....			

7) **In the last 3 months**, have friends and relatives stayed off work to help you?
Please include any time they took off to look after you, take you to healthcare appointments or visit you in hospital.

7.1 Yes ₁ *If yes: How many whole days or hours did they take off work in the last 3 months?*

No ₂

8) Which of the following best describes your **current** situation?
 Please read the whole list first and then write '1' in the box that applies. If other categories apply, write '2', '3' etc. to indicate the order that best describes your situation.

- Retired ₁
- Unable to work ₂ → Please tick reason for this
- _{2.1} Functional symptoms
- _{2.2} Other illness (please describe)

- _{2.3} Other reason (please describe)

- Made redundant / took early retirement ₃ → Please tick reason for this
- _{3.1} Functional symptoms
- _{3.2} Other illness (please describe)

- _{3.3} Other reason (please describe)

- Working full time (30 hours or more/ week) ₄
- Working part time (less than 30 hours/week) ₅
- Unemployed and looking for work ₆
- Volunteer ₇
- Job training / apprentice ₈
- At home and not looking for work (e.g. looking after home and or family) ₉
- Other: Please describe ₁₀

9) If you are currently **not working because of your health**, how long have you been unable to work because of your health condition?

Please specify in nearest number of years, months or days

10) Do you receive any state benefits? Yes ₁ No ₂

If yes: Please tick below which benefits you get and tell us how much you get altogether

- 10.2 ₁ Income support ₇ Invalidity allowance
- ₂ Family credit ₈ Disability working allowance
- ₃ Jobseeker's allowance ₉ Disability living allowance
- ₄ Housing benefit ₁₀ Incapacity benefit
- ₅ Statutory sick pay ₁₁ Attendance allowance
- ₆ Pension ₁₂ Others (please describe)

.....
 Thank you for completing this questionnaire. Your answers will remain strictly confidential.

Appendix 6 Feedback Questionnaire and Results

1.	How likely are you to recommend this programme to family and friends if they need similar treatment?					
	Extremely likely				27 (93%)	
	Likely				2 (7%)	
	Neither likely or unlikely				0	
	Unlikely				0	
	Extremely unlikely				0	
	Don't know				0	
2.	Please can you tell us the main reason for the score you have given?					
	See below					
3.	How did you find the intensity of the programme?					
	Too intense				2 (7%)	
	Very intense, but manageable				14 (48%)	
	About right				11 (38%)	
	Ok, but not very intense				1 (3%)	
	Not intense enough				1 (3%)	
4.	Overall, how satisfied were you with the physiotherapy programme?					
	Completely satisfied				25 (86%)	
	Satisfied				4 (14%)	
	Neither satisfied or dissatisfied				0	
	Dissatisfied				0	
	Completely dissatisfied				0	
5.	To what extent do you agree with the following statements:					
		Strongly disagree	Disagree	Neutral	Agree	Strongly agree
	The programme helped me to better understand my symptoms	0	0	0	2 (7%)	27 (93%)
	The programme helped me gain more control over my symptoms	0	0	2 (7%)	3 (10%)	24 (83%)
	The programme has helped me create a plan to improve my symptoms	0	0	0	3 (10%)	26 (90%)
	The programme included information about thoughts, feelings and psychological influences on my symptoms	0	0	1 (3%)	3 (11%)	24 (86%)

6.	How easy did you find completing the questionnaires?	
	Very Easy	17 (59%)
	Easy	7 (24%)
	Ok	3 (10%)
	Difficult	1 (3%)
	Very difficult	1 (3%)
7.	Do you have any comments about the questionnaires?	
	See below	
8.	Do you have any other comments or suggestions?	
	See below	

Answers to Open Ended Questions

Question 2. Please can you tell us the main reason for the score you have given? (How likely are you to recommend this programme to family and friends if they need similar treatment?)

- Gain an understanding concerning FND through medical information give, physio and how to try to improve this
- This has given me back control of my muscles and given me techniques and strategies on how to gain control over future episodes. I have been given a symptom model to explain my condition
- Very informative about diagnosis, staff know what they are talking about. Excellent explanation, motivational, assist with unaided walking
- Great one to one support from the whole team
- The improvement in my mobility this week has been beyond my wildest dreams. I still have to pinch myself to realise that I arrived in a wheelchair completely unable to maintain an upright posture and now I'm walking upright freely and on our way home the wheelchair will carry our luggage!
- Found it very helpful and didn't know that help like this was out there. If it has helped me, I'd like to think it can help other people
- Everything was explained clearly and it wasn't a pressured environment. Not once have I felt I was forced, I got on very well with my physio and that DEFINITELY helped
- It helped my problem so much more than I imagined possible in such a short period of time.
- Think once I get home I could answer this better
- I have learnt techniques that I can employ in my everyday life all of the time which will not impact on my day to day life but enhance it
- This programme is well thought out. It helps with physical symptoms as well as the mental thinking process. I feel more than able to cope with the instructions given
- This has shown that I can be helped with the support given. It has given me the tools to help myself get better or certainly improve my daily life
- I found it very educational and useful in every way. Even if it will make a slight difference in managing my condition, I would recommend this programme.

- Because I have found some things very helpful. And I can put into place when having a mild XXX (?)
- I have found all the information and tools given extremely helpful and already they are having a huge impact on my symptoms
- The information given on this issue has been amazing! I feel so empowered to deal with this. Kate has been amazing, I would recommend anyone with this condition to see her. She has helped me so much in understanding the condition
- I have made so much improvement this week, to the point I am not trembling. For 2 reasons, 1. Strategies to cope with the tremors, 2. educational aspects
- team understand you, make you normal
- Well explained reasons for condition and good advice and help
- Explained fully, treated with understanding and explanation. Everything that was told worked. Expertise of staff
- I have learnt a lot during the 5 day physio programme. I am more educated about my illness and have a very very good plan to take away with me for further rehab
- Patient centred completely and explain all the theory behind the practice. Excellent and patient approach to treatment. Feel so much more empowered myself
- I feel so relieved and relaxed. This was exactly what I needed, very informative and useful week.
- The week has helped me understand the reasons why I walk like I do. it has given me strategies to use if things became too awkward. It has given me hope that I can reverse this movement problem. I have been able to talk through my anxieties about it
- I feel that now I have a great understanding of my condition and what has caused it. Very important to me. I understand how and why I have got it. I understand that over focus is part of the problem
- Treatment has really helped me improve and everything was explained and organised really well. Kate was really nice
- It has improved my symptoms so quickly and has taught me how to carry on using the techniques to carry on the improvements
- Miraculous. Compassionate. Patient. You have helped me push my comfort zones, think and work outside the box and results are incredible. Outstanding
- Because it's worked when I thought there was no help for change at all

Question 7. Do you have any comments about the questionnaires?

- Very useful in helping to concentrate on specific issues
- Some of the questions are not clear enough
- I found it difficult to mark myself due to the fact that some days my symptoms are much worse than others
- It is difficult to answer the past week due to being in hospital and staying at the hotel. I would suggest a new one for patients who have stayed in hotels with questions more adapted to this environment

Question 8. Do you have any other comments or suggestions?

- Keeping a record over the next few weeks/months will feel important. Everything has improved so quickly it will be necessary to consolidate the whole learning process, both in terms of actual walking and fully taking on board all the changes that have happened and will go on happening over a period of time. I guess many adjustments will be needed both in practice and perception.
- Would be good to have a chance to feedback about the physio themselves. I cannot praise or thank Kate enough for help this past week. She was amazing.
- An excellent programme that has given a good understanding of the condition
- thank you for the support
- I just wanted to say THANK YOU to all the team members that put an effort in inspiring and changing the way I manage my condition
- Thank you!
- I would have enjoyed more literature to go away with and read. Kate was brilliant and did email me some, but I would like more.
- follow up within 3 months as outpatient or optional day case
- worked well, going forward will be a real boom
- if possible it might be nice to meet other participants on the programme to see how they got on and what they got out of it
- I was worried hotel experience would compromise treatment. Admissions team could have helped by providing better logistics and hotel could have been loser - had to move room 3 times last night. Would not recommend the hotel. If treatment had not been so good, got off to a v rocky start due to the hotel being such a bad experience. And a welcome pack or phone call the week before would have helped to reduce anxiety.
- Keep up the good work helping people with these symptoms and difficulties. Thank you so much :)
- thank you for the opportunity, support, motivation and understanding
- Keep doing what you do please!
- Just thank you

Appendix 7 Brief Illness Perception Questionnaire Results

Brief Illness Perception Questionnaire, dimensions

	Dimension	Group	BASELINE Mean (SD)	4 WEEK Mean (SD)	6 MONTH Mean (SD)
1	Consequences	Intervention	7.9 (2.1)	6.2 (2.5)	6.2 (2.9)
		Control	8.3 (2.1)	7.9 (2.2)	7.8 (2.3)
2	Timeline	Intervention	6.5 (2.2)	4.8 (2.4)	5.8 (2.9)
		Control	7.1 (2.3)	7.5 (2.2)	7.3 (2.3)
3	Personal control	Intervention	3.3 (2.8)	6.0 (2.4)	5.3 (2.9)
		Control	2.0 (2.1)	3.4 (2.5)	2.9 (2.2)
4	Treatment control	Intervention	6.9 (2.3)	9.0 (1.6)	7.4 (1.9)
		Control	6.3 (2.4)	6.2 (2.6)	6.1 (2.5)
5	Identity	Intervention	7.9 (2.0)	6.0 (2.7)	6.4 (2.4)
		Control	7.9 (1.5)	7.8 (1.8)	7.6 (2.3)
6	Concern	Intervention	7.9 (2.0)	5.3 (2.8)	5.5 (3.0)
		Control	7.7 (2.5)	7.4 (3.0)	7.2 (3.2)
7	Coherence	Intervention	6.2 (2.6)	8.8 (1.6)	7.8 (1.9)
		Control	4.9 (3.1)	5.7 (3.5)	5.9 (3.1)
8	Emotional Response	Intervention	6.1 (2.9)	5.0 (3.0)	5.8 (3.4)
		Control	6.8 (3.2)	6.5 (3.1)	6.3 (3.3)

The dimensions are derived from the following questions. Each question is rated on a scale of 0 to 10.

1. **Consequences:** How much does your illness affect your life?
2. **Timeline:** How long do you think your illness will continue?
3. **Personal Control:** How much control do you feel you have over your illness?
4. **Treatment Control:** How much do you think your treatment can help your illness?
5. **Identity:** How much do you experience symptoms from your illness?
6. **Concern:** How concerned are you about your illness?
7. **Coherence:** How well do you feel you understand your illness?
8. **Emotional Response:** How much does your illness affect you emotionally? (e.g. does it make you angry, scared, upset or depressed?).

In general a higher score represents greater threat, except for Items 3, 4 and 7 where lower scores represent greater threat.

Table. Brief Illness Perception Questionnaire items. Mean baseline and follow up scores are presented for control and intervention groups. The regression coefficient represents the difference between groups adjusted for baseline scores.

	Intervention Group mean (SD)		Control Group mean (SD)		Regression coefficient for group, baseline as covariate (95% CI)	Cohen's <i>d</i>
	BASELINE	FOLLOW UP	BASELINE	FOLLOW UP		
Consequences	7.9 (2.1)	6.2 (2.9)	8.3 (2.1)	7.8 (2.3)	-1.3 (-2.4, -0.1), p=0.034	0.48
Timeline	6.5 (2.2)	5.8 (2.9)	7.1 (2.3)	7.3 (2.3)	-1.1 (-2.3, 0.3), p=0.055	0.40
Personal Control	3.3 (2.8)	5.3 (2.9)	2.0 (2.1)	2.9 (2.2)	2.0 (0.7, 3.3), p=0.004	0.71
Treatment Control	6.9 (2.3)	7.4 (1.9)	6.3 (2.4)	6.1 (2.5)	1.2 (0.1, 2.3), p=0.038	0.52
Identity	7.9 (2.0)	6.4 (2.4)	7.9 (1.5)	7.6 (2.3)	-1.2 (-2.3, -0.1), p=0.031	0.49
Concern	7.9 (2.0)	5.5 (3.0)	7.7 (2.5)	7.2 (3.2)	-1.8 (-3.1, -0.5), p=0.007	0.57
Coherence	6.2 (2.6)	7.8 (1.9)	4.9 (3.1)	5.9 (3.1)	1.5 (0.2, 2.7), p=0.025	0.55
Emotional Response	6.1 (2.9)	5.8 (3.4)	6.8 (3.2)	6.3 (3.3)	0.4 (-1.3, 1.4), p=0.958	0.12

Appendix 8 Reflections on the Methodology

Randomisation

There was an issue with online the randomisation application. Participants were allocated to the control group at a faster rate than the intervention group. This is illustrated in the figure below. The company providing the randomisation application was contacted twice with these concerns, but they assured us that it was working correctly and that these differences were due to chance.

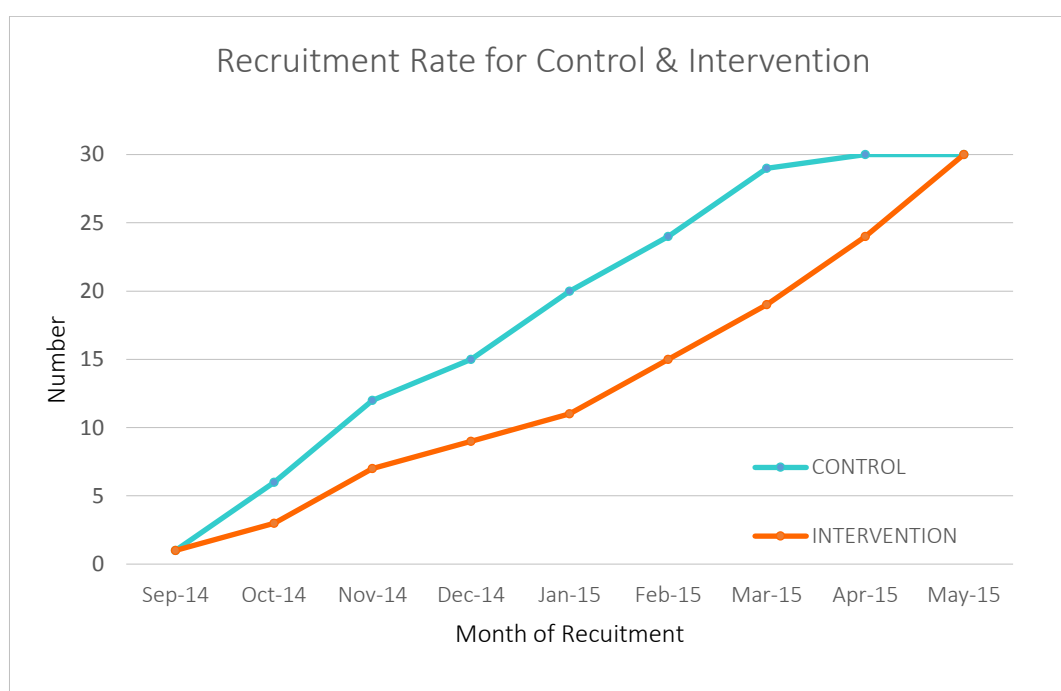


Figure 1. Recruitment rate for control and intervention groups

The result was that the control group was fully recruited (n=30) one month prior to the intervention group. Therefore the final 10 participants recruited to the study were allocated to the intervention group, which opens a possibility of participant selection bias. This issue could have been avoided using block randomisation, and this is an important consideration for a future trial.

Selection Criteria

The selection criteria appeared to work well and there were no obvious examples of participants for whom the intervention was inappropriate.

Excluding patients that score in range of “caseness” for anxiety and depression HADS is not supported by the data. Caseness of anxiety or depression was not statistically associated with outcome according to the CGI. Additionally, there was no data to support the need to exclude patients on the basis of long symptom duration or age.

There was a higher proportion of participants with “severe to extreme” self-rated pain in those that had a poor outcome compared to good (87% vs 33%). The eligibility criteria excluded patients for whom pain was the dominant symptom, but still 47% of the participants rated their pain as severe to extreme at baseline. Therefore, excluding patients with high levels of pain from a future study may limit the generalisability of the findings. In a clinical setting, the decision to exclude patients from specialist physiotherapy for FMD based on pain may depend on the availability of more suitable alternative treatments.

We excluded participants with significant comorbid psychopathology contributing to their symptoms. This was because inpatient multidisciplinary rehabilitation is more appropriate and this treatment is available from two London Hospitals.^{71,72} The exclusion criterion was left as a clinical decision rather than operationalised as a cut-off score, as no outcome measure has been identified as suitable for this purpose. The decision to exclude based on psychopathology takes into account multiple factors such as a level of anxiety/depression, and other psychiatric conditions such as personality disorder, post-traumatic stress disorder, obsessive compulsive disorder, etc. We could have been more explicit and transparent in what we considered to be significant psychopathology, for example excluding “incapacitating anxiety”, self-harming behaviour and recent suicidal ideation.

The Intervention

Given the high satisfaction ratings and positive treatment outcomes, a future trial should aim to reproduce the conditions of the study intervention. However, in a pragmatic multicentre trial some compromise will be necessary. For example, other NHS centres may not be able to deliver a physiotherapy intervention over five consecutive days and the intervention may need to be adapted to be delivered over several weeks. However, given the qualitative study finding that most participants felt that the high intensity of treatment was helpful, it would be wise to keep the study intervention higher than what would normally be delivered in standard physiotherapy.

A future iteration of the intervention should consider scheduling a follow up session approximately four to six weeks after completion of the programme, with the aim of addressing the qualitative study finding that putting management plans into practice post discharge was difficult.

Follow up

Several participants in the qualitative study found that the six month follow up period was too long and would have liked to have been seen earlier. A future trial should consider a physiotherapy follow up appointment at four to six weeks after completion of the physiotherapy programme. The benefit of telephone follow up could be explored as a potential time and cost saving measure.

The trial design could have benefited from including a 12 month telephone assessment to extend the follow up period.

An additional avenue of exploration could have been to assess how participants coped in the weeks following discharge from physiotherapy. Data collection could have included information on setbacks and symptom relapses. This data may have revealed issues that could be addressed in future iterations of the intervention to improve outcome. Collection of this data could have taken the form of post cards that are sent weekly by the participant or scheduled telephone calls.

Addressing Psychological Problems

The qualitative study found that some patients, following receiving the study intervention, came to a realisation that anxiety was part of their problem. Additionally, several participants were less resistant to considering a role for psychological factors as part of their problem after treatment. Therefore, adding information about treatment and self-help for psychological problems to the workbook may be useful. This could include internet resources for more information and treatment options available in the NHS.

Safety Reporting

Safety issues and adverse events were discussed in detail during six month follow up appointments, and more informally in telephone contact to arrange appointments. No adverse incidents were reported. However the trial design would have benefited from a systematic and formal method for identifying adverse events throughout the trial period. A future trial should build regular formal assessment of adverse events into the trial design.

The Client Services Receipt Inventory (CSRI)

A future iteration of the CSRI should aim to address the following issues: missing data regarding employment earnings; insufficient data regarding hours of sickness absence from work; and insufficient data regarding the type (and therefore cost) of hospital treatment received. Specific issues relating to the CSRI were outlined in the discussion section of the feasibility study.

Appendix 9 Qualitative Study Patient Information Sheet

UCL INSTITUTE OF NEUROLOGY
QUEEN SQUARE

THE NATIONAL HOSPITAL FOR NEUROLOGY AND
NEUROSURGERY
QUEEN SQUARE
LONDON WC1N 3BG



PATIENT INFORMATION SHEET Version 2, 8/03/2014

Patient Experiences and Perceptions of Diagnosis and Treatment of Functional Motor Symptoms

We are inviting you to take part in a research study. Before you decide it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and discuss it with others if you wish. Ask us if there is anything that is not clear or if you would like more information. Take time to decide whether or not you wish to take part. Thank you.

What is the purpose of the study?

As part of our feasibility study of physiotherapy for functional motor symptoms, we are keen to hear about the experiences and opinions of people who have undergone our treatment programme. We are interested in both positive and negative experiences since you first started experiencing symptoms. We will use the results to inform improvements to treatment design and provision for people with functional motor symptoms.

Why have I been chosen?

We are interviewing people who are part of our Feasibility of Physiotherapy study, who are allocated to the 5 day physiotherapy programme group.

Do I have to take part?

No. Taking part is voluntary. You can withdraw from the study at any point without giving a reason and there will be no pressure on you to try and change your mind. Your decision of whether or not to take part or withdraw from this study will not affect your current or future treatment.

You may withdraw before participating or at any point during the interview. If you withdraw after the interview has taken place we will include your responses in our analysis unless you request otherwise.

What will I have to do if I take part?

The research involves 3 interviews which will be conducted (i) prior to commencing the physiotherapy programme, (ii) on the final day of the physiotherapy programme and (iii) on the day of your 6 month follow up. The interviews will take place at the hospital, will take around 1 hour and will be audio recorded. You will be asked questions about how your symptoms have affected your everyday life and about your experiences with health professionals. Anything you say will remain confidential to the research team and will not be passed onto any of the other health professionals you are seeing.

UCL Institute of Neurology • National Hospital for Neurology & Neurosurgery • Queen Square • London WC1N 3BG
☎ +44 (0)20 3448 3718 • www.io.n.ucl.ac.uk
Director: Professor A Thompson MD, FRCP, FRCPI
Institute Secretary: R P Walker BSc (Econ)

The Institute of Neurology promotes teaching and research of the highest quality in neurology and the neurosciences.

However if you disclose any information pertaining to potential harm to yourself or others, I am obliged to pass this information on to the relevant authority.

How long will I be in the study?

After your third interview, your commitment to this study is complete. If you have been involved in previous clinical research you may wish to reconsider your participation considering the extra time commitment.

Expenses and Payments

We will reimburse any travel expenses that you incur that are in addition to your normal NHS care.

What are the benefits of taking part?

Your views will help inform improvements to our physiotherapy treatment programme. This research may also influence treatment of functional motor symptoms on a larger scale.

What are the possible risks?

Some people may feel upset, distressed or angry talking about their experiences. If this should happen we will stop for a break and you can decide whether or not you want to continue with the interview. The interviews will be carried out by a researcher with a background in physiotherapy.

Will my taking part be kept confidential?

The interviews will be recorded using a digital voice recorder and then transcribed using codes to protect anonymity. After the study period the audio files will be deleted. All information you give us will be kept confidential. The data will be collected and stored in accordance with the Data Protection Act 1998 and will be disposed of in a secure manner. The information will be used in a way that will not allow you to be identified individually.

How will the information be used?

The findings of this research will be published in scientific journals and presented at conferences. You will be informed about the results by the researchers, who will provide you with a one page A4 lay summary and will let you know how to access the scientific publications. The lay summary will also be sent to the patient support group FND Hope [www.fndhope.org].

Who is funding this research?

Funding for this research is provided by the National Institute for Health Research (NIHR).

Who has reviewed this study?

This study has been reviewed and approved by the National Institute for Health Research (NIHR). All research in the NHS is looked at by independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given favourable opinion by the City Road and Hampstead Research Ethics Committee.

What if there is a problem?

We think that it is unlikely that things will go wrong as all we are doing is talking to people about their experiences. However if you have a complaint or a question about the research please contact Glenn Nielsen (glenn.nielsen@uclh.nhs.uk tel 02034483718) in the first instance. If he is unable to help, please contact Fiona Stevenson on tel 020 7794 0500, ext 31008. If you remain unhappy and wish to complain formally, you can do this via the NHS Complaints Procedure. The Patient Advice and Liaison Service (PALS) can support you through this process. Contact PALS on 020 3447 3042.


Contact details

If you have any questions or concerns, please contact –
Glenn Nielsen (chief investigator).
Tel: 02034483718
Email: g.nielsen@ucl.ac.uk

This research project is registered on the following database –
<http://www.clinicaltrials.gov>

ClinicalTrials.gov ID: NCT02275000

Appendix 10 Qualitative Study Consent Form

<p>UCL INSTITUTE OF NEUROLOGY QUEEN SQUARE</p> <p>THE NATIONAL HOSPITAL FOR NEUROLOGY AND NEUROSURGERY QUEEN SQUARE LONDON WC1N 3BG</p>		
CONSENT FORM		
Study Number: 14/LO/0573 ClinicalTrials.gov ID: NCT02275000		
Title of project: Patient Experiences and Perceptions of Diagnosis and Treatment of Functional Motor Symptoms		
Please initial box		
1. I confirm that I have read and understand the information sheet dated 8/03/2014 (version 2) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	<input type="checkbox"/>	
2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my medical care or legal rights being affected.	<input type="checkbox"/>	
3. I understand that relevant sections of my medical notes and data collected during the study, may be looked at by individuals from University College London, from regulatory authorities or from the NHS Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records.	<input type="checkbox"/>	
4. I consent to the interview being audio recorded.	<input type="checkbox"/>	
6. I agree to take part in the above study	<input type="checkbox"/>	
Please send me a summary of the project outcomes to		
Address: _____		

OR Please do not send me a copy of the project outcome <input type="checkbox"/>		
_____ Name of Participant	_____ Date	_____ Signature
_____ Name of Person taking consent	_____ Date	_____ Signature
When completed, copy for participant, copy for research file, original to be filed in medical notes.		
Version 2, 8/03/2014		
UCL Institute of Neurology • National Hospital for Neurology & Neurosurgery • Queen Square • London WC1N 3BG ☎ +44 (0)20 3448 3718 • www.ion.ucl.ac.uk Director: Professor A Thompson MD, FRCP, FRCPI Institute Secretary: R P Walker BSc (Econ)		
The Institute of Neurology promotes teaching and research of the highest quality in neurology and the neurosciences.		

Appendix 11 Coding Framework

1. Acceptance of the problem
2. Awareness of own role in symptoms
3. Barriers to recovery & ongoing issues
4. Being part of a trial
5. Control
6. Critical incidents
7. Dealing with symptoms coping adapting
8. Decreased threat value
9. Employment
10. Engagement with clinicians and the system
 - a. Worthiness and the system
 - b. Waiting and diagnostic limbo
 - c. Nobody is listening
11. Expectation, Hope & Future
12. Explanation, understanding & knowledge
13. Explaining to others
14. Externalisation
15. Fatigue
16. Fear of falling
17. Feeling relief
18. Good to know I'm not alone
19. How it feels and impact
20. Invisible illness
21. Isolation
22. Mystery
23. Non-physical outcomes
24. Pain
25. Relationships and roles
26. Perceived mechanism of change
27. Physical or biological cause
28. Physical outcomes
29. Presentation of self
30. Psychological factors
 - a. Anxiety and panic
 - b. Depression and low mood
 - c. Psychological means...
31. Self reliance & self efficacy
32. Shame
33. Trust
34. About the treatment
 - a. About the physiotherapist
 - b. Differences from previous physio
 - c. Difficulties
 - d. Emotional response to treatment
 - e. Expectation going into treatment
 - f. Helpful components of treatment
 - g. It was full on
 - h. Mirror, video and visual
 - i. Praise and general comments
 - j. Suggestions & criticism
 - k. The workbook

Appendix 12 Qualitative Interview Topic Guide

PATIENT INTERVIEW TOPIC GUIDE

Patient Experiences of diagnosis and treatment of FMD

Interviewer: Glenn Nielsen

Version 2, 8/03/2014

INTERVIEW ONE: PRE-TREATMENT

[Introductory Script]

Thank you for your time and agreeing to talk to me about your experiences and for me to record this interview. I'm part of a research team investigating functional motor symptoms. We would like to learn about how it feels to be diagnosed with FMD, how they affect your life and what experiences you have had with treatment – specifically physiotherapy. We are interested in both positive and negative experiences. All information you provide me will remain anonymous. I have to let you know that if you disclose information about risk of harm to yourself or others, I am obliged to let the relevant authority know. If you would like to pause the interview at any stage, let me know and if you would like to stop the interview at any stage, that is fine.

1. Patient Narrative

- a) Could you start by telling me your story
- b) When did you first become unwell
- c) How was your health before these symptoms started

2. Illness Experience

- a) What are the different symptoms that you experience
- b) How do they affect your every day life
 - What do you need help with
 - Sleeping
 - Work
 - Looking after yourself and others
 - What would you like to be able to do
- c) Which are the most disabling symptoms
- d) Do you have any control or influence over your symptoms?

3. Receiving the diagnosis

- a) Tell me about receiving the diagnosis of FMD
- b) How did they come to the conclusion that your diagnosis was FMD
- c) How did they explain the diagnosis
- d) Was any treatment offered?
- e) What do you think is causing your symptoms?

4. Treatment

- a) Tell me about what treatments you have had prior to this programme
- b) What has been helpful so far,
- c) What has been unhelpful
- d) What do you think you need to get better?
- e) What are your expectations about this physiotherapy programme?

5. Recovery

- a) What would getting better look like to you?
- b) What is realistic to expect – from this programme? With recovery in general?
- c) What would you like to change / be able to do?
- d) Who is important in your recovery?

6. Psychological & Emotional factors

- a) How does it make you feel to have FMD
- b) Were these feelings present prior to experiencing symptoms
- c) Some people believe that psychological factors such as low mood or anxiety have a part to play in this diagnosis. How do psychological factors relate to your symptoms?

7. Free comments

Do you have anything you would like to say about your experiences?

INTERVIEW TWO – POST-TREATMENT

[Introduction]

1. Experience of the physiotherapy programme

- a) Tell me about your experience of the physiotherapy programme
- b) What did you do during the week
- c) What was most helpful
- d) What was least helpful
- e) What was different from previous experiences of physiotherapy

2. Illness Experience

- a) Has the physiotherapy programme had any impact on your symptoms
- b) What, if anything could have made the programme more effective
- c) What do you expect to happen over the next few weeks? Months?

3. Mechanism

- a) What do you think caused your symptoms
- b) What do you think is responsible for any change? (mechanism of change)
- c) What else can be done / what else do you need?

4. Recovery & The future

- a) Do you have any goals for the future
- b) How do you think you will be in 6 months? – symptom change, ability, occupations, dependence
- c) What would recovery mean / how would you be different if you improved

5. Psychological and Emotional Factors

- a) How does it feel to have FMD
- b) Are psychological factors such as low mood or anxiety relevant to your symptoms
- c) Do you have any control over your symptoms?

6. Free comments

INTERVIEW THREE – 6 MONTH FOLLOW UP

[Introduction]

1. Illness experience

- a) How have you been since completing the programme?
- b) What has changed? - ability
- c) How did you feel on initially going home
- d) Were you able to maintain improvement made, why/why not
- e) Was this what you expected?

2. Illness experience & Mechanism of change

- a) How are your symptoms now compared to before treatment
- b) Why do you think they are better / same / worse
- c) What is caused / causes your symptoms

3. Recovery & The future

- a) What help or support do you need now for your symptoms?
- b) What do you expect to happen to your symptoms over the next 6 – 12 months... and beyond

4. Physiotherapy programme

- a) Tell me about your experience of the physiotherapy programme
- b) Who do you think would most benefit from this programme

5. Free comments

References

- 1 Nielsen G, Ricciardi L, Demartini B, Hunter R, Joyce E, Edwards MJJ. Outcomes of a 5-day physiotherapy programme for functional (psychogenic) motor disorders. *J Neurol* 2015; **262**: 674–81.
- 2 Craig P, Dieppe P, Macintyre S, Michie S, Nazareth I, Petticrew M. Developing and evaluating complex interventions: the new Medical Research Council guidance. *BMJ* 2008; **337**: a1655.
- 3 Stone J, Warlow C, Sharpe M. The symptom of functional weakness: a controlled study of 107 patients. *Brain* 2010; **133**: 1537–51.
- 4 Stone J, Carson A, Duncan R, *et al.* Who is referred to neurology clinics?-the diagnoses made in 3781 new patients. *Clin Neurol Neurosurg* 2010; **112**: 747–51.
- 5 Pearce JMS. Before Charcot. *Front Neurol Neurosci* 2014; **35**: 1–10.
- 6 Carson A, Stone J, Hibberd C, *et al.* Disability, distress and unemployment in neurology outpatients with symptoms ‘unexplained by organic disease’. *J Neurol Neurosurg Psychiatry* 2011; **82**: 810–3.
- 7 Fahn S, Olanow CW. ‘Psychogenic movement disorders’: they are what they are. *Mov Disord* 2014; **29**: 853–6.
- 8 American Psychiatric Association. Diagnostic and statistical manual of mental disorders (5th ed.). Arlington, VA: American Psychiatric Publishing, 2013.
- 9 World Health Organisation. ICD-11 Beta Draft. 2016. <http://apps.who.int/classifications/icd11/browse/f/en#/http%3A%2F%2Fid.who.int%2Ficd%2Fentity%2F1882771459>.
- 10 Trimble MR. Functional diseases. *Br Med J (Clin Res Ed)* 1982; **285**: 1768.
- 11 Edwards MJ, Bhatia KP. Functional (psychogenic) movement disorders: merging mind and brain. *Lancet Neurol* 2012; **11**: 250–60.
- 12 Stone J, Wojcik W, Durrance D, *et al.* What should we say to patients with symptoms unexplained by disease? The ‘number needed to offend’. *BMJ* 2002; **325**: 1449–50.
- 13 Stone J. The bare essentials: Functional symptoms in neurology. *Pr Neurol* 2009; **9**: 179–89.
- 14 World Health Organisation. ICD-10 Version:2016. 2016. <http://apps.who.int/classifications/icd10/browse/2016/en#/F40-F48> (accessed March 6, 2017).
- 15 Stone J, Carson A. Functional neurologic disorders. *Contin (Minneap Minn)* 2015; **21**: 818–37.
- 16 Espay AJ, Goldenhar LM, Voon V, Schrag A, Burton N, Lang AE. Opinions and clinical practices related to diagnosing and managing patients with psychogenic movement disorders: An international survey of movement disorder society members. *Mov Disord* 2009; **24**: 1366–74.
- 17 Broussolle E, Gobert F, Danaila T, Thobois S, Walusinski O, Bogousslavsky J.

- History of physical and ‘moral’ treatment of hysteria. *Front Neurol Neurosci* 2014; **35**: 181–97.
- 18 Mayou R. Is the DSM-5 chapter on somatic symptom disorder any better than DSM-IV somatoform disorder? *Br J Psychiatry* 2014; **204**: 418–9.
- 19 Coggon D, Rose G, Barker D. Epidemiology for the Uninitiated 4th edition. 1997. <http://onlinebooks.library.upenn.edu/webbin/book/lookupid?key=olbp14466> (accessed March 6, 2017).
- 20 Carson A, Lehn A. Epidemiology. In: Hallett M, Stone J, Carson A, eds. Functional Neurologic Disorders, Vol 139 of the Handbook of Clinical Neurology series. Amsterdam: Elsevier, 2016: 47–60.
- 21 Carson A, Brown R, David AS, *et al.* Functional (conversion) neurological symptoms: research since the millennium. *J Neurol Neurosurg Psychiatry* 2012; **83**: 842–50.
- 22 Goodin D. The epidemiology of multiple sclerosis: insights to disease pathogenesis. In: Goodin D, ed. Multiple Sclerosis and Related Disorders, Vol 122 of the Handbook of Clinical Neurology. Amsterdam: Elsevier, 2014: 231–66.
- 23 de Lau LM, Breteler MM. Epidemiology of Parkinson’s disease. *Lancet Neurol* 2006; **5**: 525–35.
- 24 Binzer M, Andersen PM, Kullgren G. Clinical characteristics of patients with motor disability due to conversion disorder: a prospective control group study. *J Neurol Neurosurg Psychiatry* 1997; **63**: 83–8.
- 25 Akagi H, House A. The epidemiology of hysterical conversion. In: Halligan P, Bass C, Marshall J, eds. Contemporary approaches to the study of hysteria: clinical and theoretical perspectives. Oxford: Oxford University Press, 2001: 73–87.
- 26 Bermingham SL, Cohen A, Hague J, Parsonage M. The cost of somatisation among the working-age population in England for the year 2008-2009. *Ment Health Fam Med* 2010; **7**: 71–84.
- 27 Kranick S, Ekanayake V, Martinez V, Ameli R, Hallett M, Voon V. Psychopathology and psychogenic movement disorders. *Mov Disord* 2011; **26**: 1844–50.
- 28 Stone J, Edwards MJ. How ‘psychogenic’ are psychogenic movement disorders? *Mov Disord* 2011; **26**: 1787–8.
- 29 Stone J, Carson A, Aditya H, *et al.* The role of physical injury in motor and sensory conversion symptoms: a systematic and narrative review. *J Psychosom Res* 2009; **66**: 383–90.
- 30 Pareés I, Kojovic M, Pires C, *et al.* Physical precipitating factors in functional movement disorders. *J Neurol Sci* 2014; **338**: 174–7.
- 31 Boeckle M, Liegl G, Jank R, Pieh C. Neural correlates of conversion disorder: overview and meta-analysis of neuroimaging studies on motor conversion disorder. *BMC Psychiatry* 2016; **16**: 195.
- 32 Voon V, Gallea C, Hattori N, Bruno M, Ekanayake V, Hallett M. The involuntary nature of conversion disorder. *Neurology* 2010; **74**: 223–8.

- 33 Aybek S, Vuilleumier P. Imaging studies of functional neurologic disorders. In: Hallett M, Stone J, Carson A, eds. *Functional Neurologic Disorders*, Vol 139 of the *Handbook of Clinical Neurology* series. Amsterdam: Elsevier, 2017: 73–84.
- 34 Macerollo A, Chen J-C, Pareés I, Kassavetis P, Kilner JM, Edwards MJ. Sensory Attenuation Assessed by Sensory Evoked Potentials in Functional Movement Disorders. *PLoS One* 2015; **10**: 1–6.
- 35 Pareés I, Brown H, Nuruki A, *et al.* Loss of sensory attenuation in patients with functional (psychogenic) movement disorders. *Brain* 2014; **137**: 2916–21.
- 36 Teodoro T, Edwards MJ. Functional movement disorders. *Curr Opin Neurol* 2016; **29**: 519–25.
- 37 Demartini B, Petrochilos P, Ricciardi L, Price G, Edwards MJ, Joyce E. The role of alexithymia in the development of functional motor symptoms (conversion disorder). *J Neurol Neurosurg Psychiatry* 2014; **85**: 1132–7.
- 38 Edwards MJ, Adams RA, Brown H, Parees I, Friston KJ. A Bayesian account of ‘hysteria’. *Brain* 2012; **135**: 3495–512.
- 39 Edwards MJ, Fotopoulou A, Parees I. Neurobiology of functional (psychogenic) movement disorders. *Curr Opin Neurol* 2013; **26**: 442–7.
- 40 Edwards MJ, Bhatia KP, Cordivari C. Immediate response to botulinum toxin injections in patients with fixed dystonia. *Mov Disord* 2011; **26**: 917–8.
- 41 Nielsen G, Stone J, Matthews A, *et al.* Physiotherapy for functional motor disorders: a consensus recommendation. *J Neurol Neurosurg Psychiatry* 2015; **86**: 1113–9.
- 42 Daum C, Hubschmid M, Aybek S. The value of ‘positive’ clinical signs for weakness, sensory and gait disorders in conversion disorder: a systematic and narrative review. *J Neurol Neurosurg Psychiatry* 2014; **85**: 180–90.
- 43 Nielsen G, Edwards MJ, Stone J. Functional disorders presenting to the stroke service. In: Caplan L, Biller J, Leary M, *et al.*, eds. *Primer on Cerebrovascular Diseases*, Second edi. Elsevier, 2017: in press.
- 44 Stone J, Reuber M, Carson A. Functional symptoms in neurology: mimics and chameleons. *Pr Neurol* 2013; **13**: 104–13.
- 45 Fahn S, Williams DT. Psychogenic dystonia. *Adv Neurol* 1988; **50**: 431–55.
- 46 Gupta A, Lang AE. Psychogenic movement disorders. *Curr Opin Neurol* 2009; **22**: 430–6.
- 47 Espay AJ, Lang AE. Phenotype-Specific Diagnosis of Functional (Psychogenic) Movement Disorders. *Curr. Neurol. Neurosci. Rep.* 2015; **15**: 32.
- 48 Stone J, Carson A, Duncan R, *et al.* Which neurological diseases are most likely to be associated with ‘symptoms unexplained by organic disease’. *J Neurol* 2012; **259**: 33–8.
- 49 Stone J, Smyth R, Carson A, *et al.* Systematic review of misdiagnosis of conversion symptoms and ‘hysteria’. *BMJ* 2005; **331**: 989.
- 50 Kanaan R, Armstrong D, Barnes P, Wessely S. In the psychiatrist’s chair: how neurologists understand conversion disorder. *Brain* 2009; **132**: 2889–96.

- 51 Stone J, Zeman A, Simonotto E, *et al.* fMRI in patients with motor conversion symptoms and controls with simulated weakness. *Psychosom Med* 2007; **69**: 961–9.
- 52 Stone J, Aybek S. Functional limb weakness and paralysis. In: Hallett M, Stone J, Carson A, eds. *Functional Neurologic Disorders*, Vol 139 of the *Handbook of Clinical Neurology* series. Amsterdam: Elsevier, 2017: 213–28.
- 53 Gargalas S, Weeks R, Khan-Bourne N, *et al.* Incidence and outcome of functional stroke mimics admitted to a hyperacute stroke unit. *J Neurol Neurosurg Psychiatry* 2015; **88**: 2–6.
- 54 Nazir FS, Lees KR, Bone I. Clinical features associated with medically unexplained stroke-like symptoms presenting to an acute stroke unit. *Eur J Neurol* 2005; **12**: 81–5.
- 55 Tobin WO, Hentz JG, Bobrow BJ, Demaerschalk BM. Identification of stroke mimics in the emergency department setting. *J Brain Dis* 2009; **1**: 19–22.
- 56 Vroomen PC, Buddingh MK, Luijckx GJ, De Keyser J. The incidence of stroke mimics among stroke department admissions in relation to age group. *J Stroke Cerebrovasc Dis* 2008; **17**: 418–22.
- 57 Shellhaas RA, Smith SE, O’Tool E, Licht DJ, Ichord RN. Mimics of childhood stroke: characteristics of a prospective cohort. *Pediatrics* 2006; **118**: 704–9.
- 58 Tsivgoulis G, Zand R, Katsanos AH, *et al.* Safety of intravenous thrombolysis in stroke mimics: prospective 5-year study and comprehensive meta-analysis. *Stroke* 2015; **46**: 1281–7.
- 59 Lewandowski C, Mays-Wilson K, Miller J, *et al.* Safety and outcomes in stroke mimics after intravenous tissue plasminogen activator administration: a single-center experience. *J Stroke Cerebrovasc Dis* 2015; **24**: 48–52.
- 60 Zinkstok SM, Engelter ST, Gensicke H, *et al.* Safety of thrombolysis in stroke mimics: results from a multicenter cohort study. *Stroke* 2013; **44**: 1080–4.
- 61 Factor S a, Podskalny GD, Molho ES. Psychogenic movement disorders: frequency, clinical profile, and characteristics. *J Neurol Neurosurg Psychiatry* 1995; **59**: 406–12.
- 62 Bhatia KP, Schneider SA. Psychogenic tremor and related disorders. *J. Neurol.* 2007; **254**: 569–74.
- 63 Dreissen YEM, Cath DC, Tijssen MAJ. Functional jerks, tics, and paroxysmal movement disorders. In: Hallett M, Stone J, Carson A, eds. *Functional Neurologic Disorders*, Vol 139 of the *Handbook of Clinical Neurology* series. Amsterdam, 2016: 247–58.
- 64 Erro R, Edwards MJ, Bhatia KP, Esposito M, Farmer SF, Cordivari C. Psychogenic axial myoclonus: Clinical features and long-term outcome. *Park Relat Disord* 2014; **20**: 596–9.
- 65 Schrag A, Trimble M, Quinn N, Bhatia K. The syndrome of fixed dystonia: an evaluation of 103 patients. *Brain* 2004; **127**: 2360–72.
- 66 Ganos C, Edwards MJ, Bhatia KP. The Phenomenology of Functional

- (Psychogenic) Dystonia. *Mov Disord Clin Pract* 2014; **1**: 36–44.
- 67 Fung VSC. Functional gait disorder. In: Hallett M, Stone J, Carson A, eds. *Functional Neurologic Disorders*, Vol 139 of the *Handbook of Clinical Neurology* series. Amsterdam: Elsevier, 2016: 263–70.
- 68 Jankovic J. Diagnosis and treatment of psychogenic parkinsonism. *J Neurol Neurosurg Psychiatry* 2011; **82**: 1300–3.
- 69 Brown R, Reuber M. Psychological and psychiatric aspects of psychogenic non-epileptic seizures (PNES): A systematic review. *Clin Psychol Rev* 2016; **45**: 157–182.
- 70 Gelauff J, Stone J, Edwards MJ, Carson A. The prognosis of functional (psychogenic) motor symptoms: a systematic review. *J Neurol Neurosurg Psychiatry* 2014; **85**: 220–6.
- 71 Demartini B, Batla A, Petrochilos P, Fisher L, Edwards MJ, Joyce E. Multidisciplinary treatment for functional neurological symptoms: a prospective study. *J Neurol* 2014; **261**: 2370–7.
- 72 McCormack R, Moriarty J, Mellers JD, *et al*. Specialist inpatient treatment for severe motor conversion disorder: a retrospective comparative study. *J Neurol Neurosurg Psychiatry* 2013; **85**: 895–900.
- 73 Jordbru AA, Smedstad LM, Klungsøyr O, Martinsen EW. Psychogenic gait disorder: A randomized controlled trial of physical rehabilitation with one-year follow-up. *J Rehabil Med* 2014; **46**: 181–7.
- 74 Saifee TA, Kassavetis P, Parees I, *et al*. Inpatient treatment of functional motor symptoms: a long-term follow-up study. *J Neurol* 2012; **259**: 1958–63.
- 75 Czarnecki K, Thompson JM, Seime R, Geda YE, Duffy JR, Ahlskog JE. Functional movement disorders: successful treatment with a physical therapy rehabilitation protocol. *Park Relat Disord* 2012; **18**: 247–51.
- 76 Baumgartner J, Duffy JR. Psychogenic stuttering in adults with and without neurologic disease. *J Med Speech Lang Pathol* 1997; **5**: 75–95.
- 77 Sharpe M, Walker J, Williams C, *et al*. Guided self-help for functional (psychogenic) symptoms: a randomized controlled efficacy trial. *Neurology* 2011; **77**: 564–72.
- 78 Hubschmid M, Aybek S, Maccaferri GE, *et al*. Efficacy of brief interdisciplinary psychotherapeutic intervention for motor conversion disorder and nonepileptic attacks. *Gen Hosp Psychiatry* 2015; **37**: 448–55.
- 79 Kompolti K, Wilson B, Stebbins G, Bernard B, Hinson V. Immediate vs. delayed treatment of psychogenic movement disorders with short term psychodynamic psychotherapy: randomized clinical trial. *Park Relat Disord* 2014; **20**: 60–3.
- 80 Goldstein LH, Mellers JDC, Landau S, *et al*. Cognitive behavioural therapy vs standardised medical care for adults with Dissociative non-Epileptic Seizures (CODES): a multicentre randomised controlled trial protocol. *BMC Neurol* 2015; **15**: 98.
- 81 Pollak TA, Nicholson TR, Edwards MJ, David AS. A systematic review of

- transcranial magnetic stimulation in the treatment of functional (conversion) neurological symptoms. *J Neurol Neurosurg Psychiatry* 2014; **85**: 191–7.
- 82 Stone J, Hoeritzauer I, Brown K, Carson A. Therapeutic sedation for functional (psychogenic) neurological symptoms. *J Psychosom Res* 2014; **76**: 165–8.
- 83 Moene FC, Spinhoven P, Hoogduin KAL, van Dyck R. A randomised controlled clinical trial on the additional effect of hypnosis in a comprehensive treatment programme for in-patients with conversion disorder of the motor type. *Psychother Psychosom* 2002; **71**: 66–76.
- 84 Tasca C, Rapetti M, Carta MG, Fadda B. Women and hysteria in the history of mental health. *Clin Pract Epidemiol Ment Heal* 2012; **8**: 110–9.
- 85 Crommelinck M. Neurophysiology of conversion disorders: A historical perspective. *Neurophysiol Clin* 2014; **44**: 315–21.
- 86 Lamb W (translated by). Plato in Twelve Volumes, Vol 9. Perseus Digit. Libr. 1925.
<http://www.perseus.tufts.edu/hopper/text?doc=Perseus:abo:tlg,0059,031:91c> (accessed Oct 23, 2016).
- 87 Gomes M da M, Engelhardt E. A neurological bias in the history of hysteria: from the womb to the nervous system and Charcot. *Arq Neuropsiquiatr* 2014; **72**: 972–5.
- 88 Ng B-Y. Hysteria: a cross-cultural comparison of its origins and history. *Hist Psychiatry* 1999; **10**: 287–301.
- 89 Cheyne G. English Malady or a Treatise on Nervous Diseases of all kinds, as Spleen, Vapours, Lowness of Spirits, Hypochondriacal and Hysterical Distempers. Internet Arch. 1733. <https://archive.org/details/englishmaladyort00cheyuoft> (accessed May 4, 2017).
- 90 Micale MS. On the ‘disappearance’ of hysteria. A study in the clinical deconstruction of a diagnosis. *Isis* 1993; **84**: 496–526.
- 91 Trimble M. Somatization disorder: Briquet’s hysteria. In: Hallett M, Fahn S, Jankovic J, Lang AE, Cloninger CR, Yudofsky SC, eds. *Psychogenic Movement Disorders: Neurology and Neuropsychiatry*. Lippincott Williams & Wilkins, 2006: 180–5.
- 92 Briquet P. *Traité clinique et thérapeutique de l’hystérie*. Internet Arch. 1859. <https://archive.org/details/traitcliniquee00briq> (accessed May 4, 2017).
- 93 American Psychiatric Association., American Psychiatric Association. Task Force on DSM-IV. *Diagnostic and statistical manual of mental disorders : DSM-IV*. American Psychiatric Association, 1994
https://books.google.co.uk/books/about/Diagnostic_and_Statistical_Manual_of_Men.html?id=F-JGAAAAMAAJ&redir_esc=y (accessed May 4, 2017).
- 94 Reynolds JR. Remarks on paralysis and other disorders of motion and sensation dependent on idea. *Br Med J* 1869; **2**: 483–5.
- 95 Goetz CG. Charcot, hysteria, and simulated disorders. In: Hallett M, Stone J, Carson A, eds. *Functional Neurologic Disorders*, Vol 139 of the Handbook of Clinical Neurology series. Amsterdam: Elsevier, 2016: 11–23.

- 96 Gomes M da M, Engelhardt E. Hysteria to conversion disorders: Babinski's contributions. *Arq Neuropsiquiatr* 2014; **72**: 318–21.
- 97 Bogousslavsky J. Hysteria after Charcot: Back to the future. *Fronti* 2010; **29**: 137–61.
- 98 Hinson VK, Cubo E, Comella CL, Goetz CG, Leurgans S. Rating scale for psychogenic movement disorders: scale development and clinimetric testing. *Mov Disord* 2005; **20**: 1592–7.
- 99 Stone J. Neurologic approaches to hysteria, psychogenic and functional disorders from the late 19th century onwards. In: Hallett M, Stone J, Carson A, eds. *Functional Neurologic Disorders*, Vol 139 of the Handbook of Clinical Neurology series. Amsterdam: Elsevier, 2016: 25–36.
- 100 Kanaan RAA. Freud's hysteria and its legacy. In: Hallett M, Stone J, Carson A, eds. *Functional Neurologic Disorders*, Vol 139 of the Handbook of Clinical Neurology series. Amsterdam: Elsevier, 2016: 37–44.
- 101 Tomlinson C. Freud and psychogenic movement disorders. In: Hallett M, Fahn S, Jankovic J, Lang AE, Cloninger CR, Yudofsky SC, eds. *Psychogenic Movement Disorders: Neurology and Neuropsychiatry*. Philadelphia: Lippincott Williams & Wilkins, 2006: 14–9.
- 102 Linden SC, Jones E, Lees AJ. Shell shock at Queen Square: Lewis Yealland 100 years on. *Brain* 2013; **136**: 1976–88.
- 103 Teasell RW, Shapiro AP. Strategic-behavioral intervention in the treatment of chronic nonorganic motor disorders. *Am J Phys Med Rehabil* 1994; **73**: 44–50.
- 104 Stone J, Hewett R, Carson A, Warlow C. The 'disappearance' of hysteria: historical mystery or illusion? *J R Soc Med* 2008; **101**: 12–8.
- 105 Stone J, Warlow C, Carson A, Sharpe M. Eliot Slater's myth of the non-existence of hysteria. *JRSM* 2005; **98**: 547–8.
- 106 Slater E. Diagnosis of hysteria. *Br Med J* 1965; **1**: 1395–9.
- 107 Crimlisk HL, Bhatia K, Cope H, David A, Marsden CD, Ron MA. Slater revisited: 6 year follow up study of patients with medically unexplained motor symptoms. *BMJ* 1998; **316**: 582–6.
- 108 Munts AG, Koehler PJ. How psychogenic is dystonia views from past to present. *Brain* 2010; **133**: 1552–64.
- 109 van der Salm SMA, Erro R, Cordivari C, *et al*. Propriospinal myoclonus: clinical reappraisal and review of literature. *Neurology* 2014; **83**: 1862–70.
- 110 Nielsen G, Stone J, Edwards MJ. Physiotherapy for functional (psychogenic) motor symptoms: A systematic review. *J Psychosom Res* 2013; **75**: 93–102.
- 111 Shapiro AP, Teasell RW. Behavioural interventions in the rehabilitation of acute v. chronic non-organic (conversion/factitious) motor disorders. *Br J Psychiatry* 2004; **185**: 140–6.
- 112 Matthews A, Brown M, Stone J. Inpatient Physiotherapy for Functional (Psychogenic) Gait Disorder: A Case Series of 35 Patients. *Mov Disord Clin Pract* 2016; **28**: 93–6.

- 113 Heruti RJ, Reznik J, Adunski A, Levy A, Weingarden H, Ohry A. Conversion motor paralysis disorder: analysis of 34 consecutive referrals. *Spinal Cord* 2002; **40**: 335–40.
- 114 Ferrara J, Stamey W, Strutt AM, Adam OR, Jankovic J. Transcutaneous electrical stimulation (TENS) for psychogenic movement disorders. *J Neuropsychiatry Clin Neurosci* 2011; **23**: 141–8.
- 115 Dallochio C, Arbasino C, Klersy C, Marchioni E. The effects of physical activity on psychogenic movement disorders. *Mov Disord* 2010; **25**: 421–5.
- 116 Espay AJ, Edwards MJ, Oggioni GD, *et al.* Tremor retraining as therapeutic strategy in psychogenic (functional) tremor. *Park Relat Disord* 2014; **20**: 647–50.
- 117 Speed J. Behavioral management of conversion disorder: retrospective study. *Arch Phys Med Rehabil* 1996; **77**: 147–54.
- 118 Moene FC, Hoogduin KA, Van Dyck R. The inpatient treatment of patients suffering from (motor) conversion symptoms: a description of eight cases. *Int J Clin Exp Hypn* 1998; **46**: 171–90.
- 119 Weiser HI. Motor sensory dysfunction of upper limb due to conversion syndrome. *Arch Phys Med Rehabil* 1976; **57**: 17–9.
- 120 Delargy MA, Peatfield RC, Burt AA. Successful rehabilitation in conversion paralysis. *Br Med J (Clin Res Ed)* 1986; **292**: 1730–1.
- 121 Withrington RH, Wynn Parry CB. Rehabilitation of conversion paralysis. *J Bone Jt Surg Br* 1985; **67**: 635–7.
- 122 Cardenas DD, Larson J, Egan KJ. Hysterical paralysis in the upper extremity of chronic pain patients. *Arch Phys Med Rehabil* 1986; **67**: 190–3.
- 123 Fishbain DA, Lewis JE, Gao J, Cole B, Steele Rosomoff R. Is chronic pain associated with somatization/hypochondriasis? An evidence-based structured review. *Pain Pr* 2009; **9**: 449–67.
- 124 Silver FW. Management of conversion disorder. *Am J Phys Med Rehabil* 1996; **75**: 134–40.
- 125 Watanabe TK, O'Dell MW, Togliatti TJ. Diagnosis and rehabilitation strategies for patients with hysterical hemiparesis: a report of four cases. *Arch Phys Med Rehabil* 1998; **79**: 709–14.
- 126 Behr J. The role of physiotherapy in the recovery of patients with conversion disorder. *Physiother Can* 1996; **48**: 197–202.
- 127 Ness D. Physical therapy management for conversion disorder: case series. *J Neurol Phys Ther* 2007; **31**: 30–9.
- 128 Trieschmann RB, Stolov WC, Montgomery ED. An approach to the treatment of abnormal ambulation resulting from conversion reaction. *Arch Phys Med Rehabil* 1970; **51**: 198–206.
- 129 Vatine J, Milun M, Avni J. Inpatient rehabilitation management of conversion disorder with motor dysfunction. *J Rehabil Sci* 1996; **9**: 122–5.
- 130 Glennon Á. A practical approach to the physiotherapy assessment and treatment of conversion disorders. *R Coll Surg Irel Stud Med J* 2011; **4**: 57–61.

- 131 Atan C, Seckin U, Bodur H. Hysterical paralysis. *Rheumatol Int* 2007; **27**: 873–4.
- 132 Oh DW, Yoo EY, Yi CH, Kwon OY. Physiotherapy strategies for a patient with conversion disorder presenting abnormal gait. *Physiother Res Int* 2005; **10**: 164–8.
- 133 Sullivan M, Buchanan D. The treatment of conversion disorder in a rehabilitation setting. *Can J Rehabil* 1989; **2**: 175–80.
- 134 Khalil TM, Abdel-Moty E, Asfour SS, Fishbain DA, Rosomoff RS, Rosomoff HL. Functional electric stimulation in the reversal of conversion disorder paralysis. *Arch Phys Med Rehabil* 1988; **69**: 545–7.
- 135 Findlater KA. A behavioural approach to the rehabilitation of longstanding hysterical quadriplegia: a case report. *Physiother Can* 1986; **38**: 216–22.
- 136 Klein MJ, Kewman DG, Sayama M. Behavior modification of abnormal gait and chronic pain secondary to somatization disorder. *Arch Phys Med Rehabil* 1985; **66**: 119–22.
- 137 MacKinnon JL. Physical therapy treatment of a patient with a conversion reaction. A case report. *Phys Ther* 1984; **64**: 1687–8.
- 138 Jenkinson C, Coulter A, Wright L. Short form 36 (SF36) health survey questionnaire: normative data for adults of working age. *BMJ* 1993; **306**: 1437–40.
- 139 Hughes S, Alltree J. A behavioural approach to the management of functional disorders. *Physiotherapy* 1990; **76**: 255–8.
- 140 Fishbain DA, Goldberg M, Khalil TM, *et al*. The utility of electromyographic biofeedback in the treatment of conversion paralysis. *Am J Psychiatry* 1988; **145**: 1572.
- 141 Deaton A V. Treating conversion disorders: Is a pediatric rehabilitation hospital the place? *Rehabil Psychol* 1998; **43**: 56.
- 142 Keurentjes JC, Van Tol FR, Fiocco M, Schoones JW, Nelissen RG. Minimal clinically important differences in health-related quality of life after total hip or knee replacement: A systematic review. *Bone Joint Res* 2012; **1**: 71–7.
- 143 Clement ND, MacDonald D, Simpson AHRW. The minimal clinically important difference in the Oxford knee score and Short Form 12 score after total knee arthroplasty. *Knee Surg Sports Traumatol Arthrosc* 2016; **22**: 1933–9.
- 144 Beninato M, Gill-Body KM, Salles S, Stark PC, Black-Schaffer RM, Stein J. Determination of the Minimal Clinically Important Difference in the FIM Instrument in Patients With Stroke. *Arch Phys Med Rehabil* 2006; **87**: 32–9.
- 145 Edwards MJ, Stone J, Nielsen G. Physiotherapists and patients with functional (psychogenic) motor symptoms: a survey of attitudes and interest. *J Neurol Neurosurg Psychiatry* 2012; **83**: 655–8.
- 146 Rose M, Stanley I, Peters S, Salmon P, Stott R, Crook P. Wrong Problem, Wrong Treatment Unrecognised inappropriate referral to physiotherapy. *Physiotherapy* 1999; **85**: 322–8.
- 147 Kenney C, Diamond A, Mejia N, Davidson A, Hunter C, Jankovic J. Distinguishing

- psychogenic and essential tremor. *J Neurol Sci* 2007; **263**: 94–9.
- 148 Parees I, Kassavetis P, Saifee TA, *et al.* Failure of explicit movement control in patients with functional motor symptoms. *Mov Disord* 2013; **28**: 517–23.
- 149 Jackson, Robin C.Ashford, Kelly J.Norsworthy G. Attentional Focus, Dispositional Reinvestment, and Skilled Motor Performance Under Pressure. *J Sport Exerc Psychol* 2006; **28**: 49–68.
- 150 Masters RSW. Knowledge, knerves and know-how: The role of explicit versus implicit knowledge in the breakdown... *Br J Psychol* 1992; **83**: 343–58.
- 151 Masters R, MacMahon K, Pall H. Implicit Motor Learning in Parkinson's Disease. *Rehabil Psychol* 2004; **49**: 79–82.
- 152 Orrell AJ, Eves FF, Masters RSW. Motor learning of a dynamic balancing task after stroke: implicit implications for stroke rehabilitation. *Phys Ther* 2006; **86**: 369–80.
- 153 Sharpe M, Stone J, Hibberd C, *et al.* Neurology out-patients with symptoms unexplained by disease: illness beliefs and financial benefits predict 1-year outcome. *Psychol Med* 2010; **40**: 689–98.
- 154 Stone J, Edwards MJ. Trick or treat? Showing patients with functional (psychogenic) motor symptoms their physical signs. *Neurology* 2012; **79**: 282–4.
- 155 Stone J, Warlow C, Sharpe M. Functional weakness: clues to mechanism from the nature of onset. *J Neurol Neurosurg Psychiatry* 2012; **83**: 67–9.
- 156 Carson A, Ludwig L, Welch K. Psychologic theories in functional neurologic disorders. In: Hallett M, Stone J, Carson A, eds. *Functional Neurologic Disorders*, Vol 139 of the *Handbook of Clinical Neurology* series. Amsterdam: Elsevier, 2016: 105–20.
- 157 Voon V, Cavanna AE, Coburn K, Sampson S, Reeve A, LaFrance WC. Functional Neuroanatomy and Neurophysiology of Functional Neurological Disorders (Conversion Disorder). *J Neuropsychiatry Clin Neurosci* 2016; **28**: 168–90.
- 158 National Institute for Health and Care Excellence. Chronic fatigue syndrome/myalgic encephalomyelitis (or encephalopathy): diagnosis and management. *NICE Guidel* 2007.
<https://www.nice.org.uk/guidance/cg53/resources/chronic-fatigue-syndromemyalgic-encephalomyelitis-or-encephalopathy-diagnosis-and-management-975505810885>.
- 159 National Institute for Health and Care Excellence. Low back pain in adults: early management. *NICE Guidel* 2009.
<https://www.nice.org.uk/guidance/cg88/resources/low-back-pain-in-adults-early-management-975695607493>.
- 160 Butler DS, Moseley GL. *Explain Pain*. Adelaide: Noigroup Publications, 2003.
- 161 Lamb SE, Lall R, Hansen Z, *et al.* A multicentred randomised controlled trial of a primary care-based cognitive behavioural programme for low back pain. The Back Skills Training (BeST) trial. *Health Technol Assess (Rockv)* 2010; **14**: 1–128.
- 162 Nielsen G. Physical treatment of functional neurologic disorders. In: Hallett M,

- Stone J, Carson A, eds. Functional Neurologic Disorders, Vol 139 of the Handbook of Clinical Neurology series. Amsterdam: Elsevier, 2016: 555–69.
- 163 McHorney CA, Ware JE, Raczek AE. The MOS 36-Item Short-Form Health Survey (SF-36): II. Psychometric and clinical tests of validity in measuring physical and mental health constructs. *Med Care* 1993; **31**: 247–63.
- 164 Jenkinson C, Wright L, Coulter A. Criterion validity and reliability of the SF-36 in a population sample. *Qual Life Res* 1994; **3**: 7–12.
- 165 Zahra D, Qureshi A, Henley W, *et al*. The work and social adjustment scale: reliability, sensitivity and value. *Int J Psychiatry Clin Pract* 2014; **18**: 131–8.
- 166 Zigmond AS, Snaith RP. The hospital anxiety and depression scale. *Acta Psychiatr Scand* 1983; **67**: 361–70.
- 167 Broadbent E, Petrie KJ, Main J, Weinman J. The brief illness perception questionnaire. *J Psychosom Res* 2006; **60**: 631–7.
- 168 Carson A, Best S, Postma K, Stone J, Warlow C, Sharpe M. The outcome of neurology outpatients with medically unexplained symptoms: a prospective cohort study. *J Neurol Neurosurg Psychiatry* 2003; **74**: 897–900.
- 169 Hudak PL, Amadio PC, Bombardier C. Development of an upper extremity outcome measure: the DASH (disabilities of the arm, shoulder and hand) [corrected]. The Upper Extremity Collaborative Group (UECG). *Am J Ind Med* 1996; **29**: 602–8.
- 170 Velstra IM, Ballert CS, Cieza A. A systematic literature review of outcome measures for upper extremity function using the international classification of functioning, disability, and health as reference. *PM R* 2011; **3**: 846–60.
- 171 Graham HK, Harvey A, Rodda J, Natrass GR, Pirpiris M. The Functional Mobility Scale (FMS). *J Pediatr Orthop* 2004; **24**: 514–20.
- 172 Peters DM, Fritz SL, Krotish DE. Assessing the reliability and validity of a shorter walk test compared with the 10-Meter Walk Test for measurements of gait speed in healthy, older adults. *J Geriatr Phys Ther*; **36**: 24–30.
- 173 Berg KO, Wood-Dauphinee SL, Williams JI, Maki B. Measuring balance in the elderly: validation of an instrument. *Can J Public Health* 1992; **83 Suppl 2**: S7-11.
- 174 La Porta F, Caselli S, Susassi S, Cavallini P, Tennant A, Franceschini M. Is the Berg Balance Scale an internally valid and reliable measure of balance across different etiologies in neurorehabilitation? A revisited Rasch analysis study. *Arch Phys Med Rehabil* 2012; **93**: 1209–16.
- 175 Nielsen G, Ricciardi L, Meppelink A, Holt K, Teodoro T, Edwards MJ. A simplified version of the Psychogenic Movement Disorders Rating Scale: The Simplified Functional Movement Disorders Rating Scale (S-FMDRS). *Mov Disord Clin Pract* 2017; : online first.
- 176 Herdman M, Gudex C, Lloyd A, *et al*. Development and preliminary testing of the new five-level version of EQ-5D (EQ-5D-5L). *Qual Life Res* 2011; **20**: 1727–36.
- 177 National Institute for Health and Clinical Excellence (NICE). Social value judgements: principles for the development of NICE guidance (2nd edn.). 2008.

<https://www.nice.org.uk/media/default/about/what-we-do/research-and-development/social-value-judgements-principles-for-the-development-of-nice-guidance.pdf> (accessed April 12, 2016).

- 178 Chisholm D, Knapp MR, Knudsen HC, Amaddeo F, Gaité L, van Wijngaarden B. Client Socio-Demographic and Service Receipt Inventory--European Version: development of an instrument for international research. EPSILON Study 5. European Psychiatric Services: Inputs Linked to Outcome Domains and Needs. *Br J Psychiatry Suppl* 2000; : s28-33.
- 179 Sedgwick P. Effect sizes. *BMJ* 2012; **345**: e7370.
- 180 Devlin N, Shah K, Feng Y, Mulhern B, van Hout B. Valuing health-related quality of life: an EQ-5D-5L value set for England. 2016. <https://www.ohe.org/publications/valuing-health-related-quality-life-eq-5d-5l-value-set-england> (accessed July 12, 2016).
- 181 Hunter RM, Baio G, Butt T, Morris S, Round J, Freemantle N. An educational review of the statistical issues in analysing utility data for cost-utility analysis. *Pharmacoeconomics* 2015; **33**: 355–66.
- 182 Curtis L, Burns A. Unit costs of health and social care 2015. *Pers Soc Serv Res Unit, Univ Kent* 2015. <http://www.pssru.ac.uk/project-pages/unit-costs/2015/> (accessed April 28, 2016).
- 183 Ahmad O, Ahmad KE. Functional neurological disorders in outpatient practice: An Australian cohort. *J Clin Neurosci* 2016; **28**: 93–6.
- 184 Nelson MC, Cordray DS, Hulleman CS, Darrow CL, Sommer EC. A procedure for assessing intervention fidelity in experiments testing educational and behavioral interventions. *J Behav Health Serv Res* 2012; **39**: 374–96.
- 185 Ware J, Kosinski M, Keller SD. A 12-Item Short-Form Health Survey: construction of scales and preliminary tests of reliability and validity. *Med Care* 1996; **34**: 220–33.
- 186 National Institute for Health and Clinical Excellence (NICE). National Institute for Health and Clinical Excellence (November 2012) The guidelines manual. www.nice.org.uk (accessed June 16, 2016).
- 187 McCrone P, Sharpe M, Chalder T, *et al.* Adaptive Pacing, Cognitive Behaviour Therapy, Graded Exercise, and Specialist Medical Care for Chronic Fatigue Syndrome: A Cost-Effectiveness Analysis. *PLoS One* 2012; **7**: e40808.
- 188 Lamb SE, Hansen Z, Lall R, *et al.* Group cognitive behavioural treatment for low-back pain in primary care: a randomised controlled trial and cost-effectiveness analysis. *Lancet* 2010; **375**: 916–23.
- 189 Lenneman J, Schwartz S, Giuseffi DL, Wang C. Productivity and health: an application of three perspectives to measuring productivity. *J Occup Environ Med* 2011; **53**: 55–61.
- 190 Broadbent E, Wilkes C, Koschwanez H, Weinman J, Norton S, Petrie KJ. A systematic review and meta-analysis of the Brief Illness Perception Questionnaire. *Psychol Health* 2015; **30**: 1361–85.
- 191 Nielsen G, Buszewicz M, Stevenson F, *et al.* Randomised Feasibility Study of

- Physiotherapy for Patients with Functional Motor Symptoms. *J Neurol Neurosurg Psychiatry* 2016; **online fir**: jnnp-2016-314408.
- 192 Evans RW, Evans RE. A survey of neurologists on the likeability of headaches and other neurological disorders. *Headache* 2010; **50**: 1126–9.
- 193 Ahern L, Stone J, Sharpe MC. Attitudes of neuroscience nurses toward patients with conversion symptoms. *Psychosomatics* 2009; **50**: 336–9.
- 194 O’Cathain A, Thomas KJ, Drabble SJ, Rudolph A, Hewison J. What can qualitative research do for randomised controlled trials? A systematic mapping review. *BMJ Open* 2013; **3**: 1–15.
- 195 Nettleton S, Watt I, O’Malley L, Duffey P. Understanding the narratives of people who live with medically unexplained illness. *Patient Educ Couns* 2005; **56**: 205–10.
- 196 Rawlings GH, Reuber M. What patients say about living with psychogenic nonepileptic seizures: A systematic synthesis of qualitative studies. *Seizure*. 2016; **41**: 100–11.
- 197 Calman L, Brunton L, Molassiotis A. Developing longitudinal qualitative designs: lessons learned and recommendations for health services research. *BMC Med Res Methodol* 2013; **13**: 14.
- 198 Ritchie J, Lewis J, Elam G. Designing and Selecting Samples. In: Ritchie J, Lewis J, eds. *Qualitative Research Practice: A guide for social science students and researchers*. London: SAGE Publications, 2003: 78–108.
- 199 Braun V, Clarke V. Using thematic analysis in psychology. *Qual Res Psychol* 2006; **3**: 77–101.
- 200 Parees I, Saifee TA, Kassavetis P, *et al*. Believing is perceiving: mismatch between self-report and actigraphy in psychogenic tremor. *Brain* 2012; **135**: 117–23.
- 201 Ricciardi L, Demartini B, Morgante F, Parees I, Nielsen G, Edwards MJ. Symptom severity in patients with functional motor symptoms: Patient’s perception and doctor’s clinical assessment. *Parkinsonism Relat Disord* 2015; **21**: 529–32.
- 202 Engel GL. The need for a new medical model: a challenge for biomedicine. *Science* 1977; **196**: 129–36.
- 203 Anderson KE, Gruber-Baldini AL, Vaughan CG, *et al*. Impact of psychogenic movement disorders versus Parkinson’s on disability, quality of life, and psychopathology. *Mov Disord* 2007; **22**: 2204–9.
- 204 Binzer M, Eisemann M, Kullgren G. Illness behavior in the acute phase of motor disability in neurological disease and in conversion disorder: A comparative study. *J Psychosom Res* 1998; **44**: 657–66.
- 205 Kanaan RA, Armstrong D, Wessely SC. Neurologists’ understanding and management of conversion disorder. *J Neurol Neurosurg Psychiatry* 2011; **82**: 961–6.
- 206 Sifneos PE. The prevalence of ‘Alexithymic’ characteristics in psychosomatic patients. 1973; **22**: 255–62.

- 207 Gulpek D, Kelemence Kaplan F, Kesebir S, Bora O. Alexithymia in patients with conversion disorder. *Nord J Psychiatry* 2014; **68**: 300–5.
- 208 Karterud HN, Risør MB, Haavet OR. The impact of conveying the diagnosis when using a biopsychosocial approach: A qualitative study among adolescents and young adults with NES (non-epileptic seizures). *Seizure* 2015; **24**: 107–13.
- 209 Turner J, Holtzman S, Mancl L. Mediators, moderators, and predictors of therapeutic change in cognitive–behavioral therapy for chronic pain. *Pain* 2007; **127**: 276–86.
- 210 Chalder T, Goldsmith KA, White PD, Sharpe M, Pickles AR. Rehabilitative therapies for chronic fatigue syndrome: A secondary mediation analysis of the PACE trial. *The Lancet Psychiatry* 2015; **2**: 141–52.
- 211 Cockerham WC. Medical sociology. *Int Rev Mod Sociol* 1981; **11**: 231–50.
- 212 Moss-Morris R, Weinman J, Petrie KJ, Horne R, Cameron LD, Buick D. The revised illness perception questionnaire (IPQ-R). *Psychol Heal* 2002; **16**: 1–16.
- 213 Ferreira PH, Ferreira ML, Maher CG, Refshauge KM, Latimer J, Adams RD. The therapeutic alliance between clinicians and patients predicts outcome in chronic low back pain. *Phys Ther* 2013; **93**: 470–8.
- 214 Health Improvement Scotland. Stepped care for functional neurological symptoms. 2012. http://www.healthcareimprovementscotland.org/our_work/long_term_conditions/neurological_health_services/neurological_symptoms_report.aspx (accessed March 6, 2017).
- 215 Hurley DA, Murphy LC, Hayes D, *et al.* Using intervention mapping to develop a theory-driven, group-based complex intervention to support self-management of osteoarthritis and low back pain (SOLAS). *Implement Sci* 2015; **11**: 56.
- 216 Bartholomew Eldredge LK, Markham CM, Ruitter RA, Fernández ME, Kok G, Parcel GS. Planning health promotion programs : an intervention mapping approach, 4th edn. San Francisco: Jossey-Bass, 2016.
- 217 Guo B, Moga C, Harstall C, Schopflocher D. A principal component analysis is conducted for a case series quality appraisal checklist. *J Clin Epidemiol* 2016; **69**: 199–207.
- 218 Boger E, Ellis J, Latter S, *et al.* Self-Management and Self-Management Support Outcomes: A Systematic Review and Mixed Research Synthesis of Stakeholder Views. *PLoS One* 2015; **10**: e0130990.