



THE UNIVERSITY *of* EDINBURGH

This thesis has been submitted in fulfilment of the requirements for a postgraduate degree (e.g. PhD, MPhil, DClinPsychol) at the University of Edinburgh. Please note the following terms and conditions of use:

- This work is protected by copyright and other intellectual property rights, which are retained by the thesis author, unless otherwise stated.
- A copy can be downloaded for personal non-commercial research or study, without prior permission or charge.
- This thesis cannot be reproduced or quoted extensively from without first obtaining permission in writing from the author.
- The content must not be changed in any way or sold commercially in any format or medium without the formal permission of the author.
- When referring to this work, full bibliographic details including the author, title, awarding institution and date of the thesis must be given.



**Post-Stroke Depression (PSD) and Post-Stroke Emotional Lability (PSEL):
A systematic review of the problems with the evidence for non-pharmacological
interventions for PSD, and a qualitative study of specialist professionals'
conceptualisations of PSEL**

Hannah Kathryn Picton

*Submitted in part fulfilment of the degree of
Doctorate in Clinical Psychology
at the University of Edinburgh*

August 2013

Word Count: 25, 667

Table of Contents

DECLARATION OF OWN WORK	5
EPIGRAPH	6
ACKNOWLEDGEMENTS	7
OVERVIEW OF THESIS PORTFOLIO	8
PORTFOLIO THESIS ABSTRACT	9
CHAPTER ONE: INTRODUCTION TO THESIS	10
CHAPTER TWO: SYSTEMATIC REVIEW	14
SYSTEMATIC REVIEW ABSTRACT	15
BACKGROUND	16
METHOD	22
RESULTS	28
DISCUSSION	43
SYSTEMATIC REVIEW REFERENCES	50
CHAPTER THREE: JOURNAL ARTICLE	53
JOURNAL ARTICLE ABSTRACT	56
INTRODUCTION	57
STUDY AIMS	61
METHOD	60
RESULTS	64
DISCUSSION	87
JOURNAL ARTICLE REFERENCES	92
CHAPTER FOUR: EXTENDED METHOD	97
DESIGN	97
PROCEDURE	98
DATA COLLECTION AND ANALYSIS	99
EVALUATING GROUNDED THEORY	109
DATA MANAGEMENT	111
ETHICAL REVIEW	112
THESIS REFERENCES	113

List of Tables and Figures

TABLE 1: OVERVIEW OF EXCLUDED STUDIES	28
TABLE 2: OVERVIEW OF SELECTED STUDIES	30-31
TABLE 3: OVERVIEW OF METHODOLOGICAL QUALITY RATINGS	36
TABLE 4: PARTICIPANT INFORMATION	63
FIGURE 1: FLOW CHART OF ARTICLE SELECTION PROCESS	25
FIGURE 2: 'HOW STAFF THINK ABOUT PSEL' (DIAGRAMMATIC REPRESENTATION)	67
FIGURE 3: OVERVIEW OF CONCEPTUAL CATEGORIES	68

List of Appendices

APPENDIX 1: AUTHOR GUIDELINES FOR THE JOURNAL	124
APPENDIX 2: SYSTEMATIC REVIEW PROTOCOL	130
APPENDIX 3: SYSTEMATIC REVIEW DATA EXTRACTION FORM	133
APPENDIX 4: QUALITY RATING TOOL FOR SYSTEMATIC REVIEW	135
APPENDIX 5: PARTICIPANT INFORMATION SHEET	142
APPENDIX 6: INTERVIEW QUESTIONS	146
APPENDIX 7: CONSENT FORM	147
APPENDIX 8: SUPPLEMENTARY PARTICIPANT INFORMATION	148
APPENDIX 9: ETHICAL APPROVAL	149
APPENDIX 10: R&D AMENDMENT APPROVAL	150
APPENDIX 11: NHS-TO-NHS ACCESS FOR RESEARCH APPROVAL	151

D.CLIN. PSYCHOL.
UNIVERSITY OF EDINBURGH / NHS (SCOTLAND)
TRAINING PROGRAMME



Front sheet / Title Page for Submitted Academic Work

NAME: Hannah Picton

TITLE: Post-Stroke Depression (PSD) and Post-Stroke Emotional Lability (PSEL): A systematic review of non-pharmacological interventions for PSD, and a qualitative study of specialist professionals' conceptualisations of PSEL

COURSE SUBMITTED FOR (please tick relevant box):

	CP1	CP2
Case study conceptualisation (CP1 and CP2)	<input type="checkbox"/>	<input type="checkbox"/>
	3	4
Case study (only for those starting pre 2009)	<input type="checkbox"/>	<input type="checkbox"/>
	OA/Neuro	Child
Essay questions (only for those starting pre 2009)	<input type="checkbox"/>	<input type="checkbox"/>
Research proposal (R1)		<input type="checkbox"/>
Small scale research project (R2)		<input type="checkbox"/>
Small scale research project 2 (only for those starting pre 2009)		<input type="checkbox"/>
Thesis		<input checked="" type="checkbox"/>

Submitted in part fulfilment of the degree of doctorate in Clinical Psychology at the University of Edinburgh

DATE SUBMITTED:

For small scale research projects, case studies and case study conceptualisations:

I certify that this report is a fair and accurate account of the work carried out:

Trainee Signature: _____

Supervisor Name: Dr Ethel Quayle/Dr David Gillespie

Supervisor Signature: _____



**D. CLIN. PSYCHOL.
UNIVERSITY OF EDINBURGH / NHS (SCOTLAND) TRAINING PROGRAMME**

Declaration of Own Work

NAME: Hannah Picton

ASSESSED WORK: Doctoral thesis

TITLE OF WORK: Post-Stroke Depression (PSD) and Post-Stroke Emotional Lability (PSEL):
A systematic review of non-pharmacological interventions for PSD, and
a qualitative study of specialist professionals' conceptualisations of
PSEL

I confirm that all this work is my own except where indicated, and that I have:

- Read and understood the Plagiarism Rules and Regulations
- Composed and undertaken the work myself
- Clearly referenced/listed all sources as appropriate
- Referenced and put in inverted commas any quoted text of more than three words (from books, web, etc)
- Given the sources of all pictures, data etc. that are not my own
- Not made undue use of essay(s) of any other student(s) either past or present (or where used, this has been referenced appropriately)
- Not sought or used the help of any external professional agencies for the work (or where used, this has been referenced appropriately)
- Not submitted the work for any other degree or professional qualification except as specified
- Acknowledged in appropriate places any help that I have received from others (e.g. fellow students, technicians, statisticians, external sources)
- Complied with other plagiarism criteria specified in the Programme Handbook
- I understand that any false claim for this work will be penalised in accordance with the University regulations

Signature: _____

Date: 07/08/2013

Epigraph

(Frances McGill, 1980)

"I have been aware while it was happening that I was not as upset or as sad as my crying would imply, nor as uproariously amused as my uncontrollable laughter would indicate. Such episodes of laughter and tears may have slight connection with my actual frame of mind or the feeling which is actually mine at the time. In fact, I usually become very frustrated and angry at my inability to put a halt to such ridiculous behavior!

I begin to smile, but the smile becomes an exaggerated grin, which attaches itself, fixedly, to my face, and I have to use all my powers of concentration to remove the embarrassing grimace. If I yield to the impulse, it becomes the onset to equally uncontrollable giggles, which in turn so embarrass me, that I become angry, humiliated, and subject to uncontrollable tears—it is a vicious, see-sawing circle!

...I am mortally afraid of squealing bawls. They destroy me—they weaken and crumble me...those deep debilitating agonizing episodes. You have no idea how terrible it is when the crying is fully triggered and takes hold like a seizure. I cannot control any of it. I simply disintegrate and it is not only emotionally horrible with me, it is physically painful and debilitating"

ACKNOWLEDGEMENTS

First and foremost, I would like to thank the professionals who participated in the emotionalism study, whose insight, thoughtfulness, experience, and expertise are the essence of the journal article.

Secondly, I owe my deepest gratitude to Dr David Gillespie, for going so far over and above the call of duty in his role as my clinical supervisor, and for always pushing me further than I think I can go. In the absence of his enthusiasm, patience, endless generosity, and expert guidance, this thesis would never have been possible. A huge thanks to Dr Ethel Quayle, for her kindness, salutary qualitative research advice, and containment! Thanks to Dr Niall Broomfield for his support with recruitment, and to Fleur-Michelle Coiffait, Amy Cadden, and Nick Earley for their support and friendship, kindness, and input into the systematic review and journal article. I would also like to thank Dr Rosalind Evans, for providing me with a 'secure base' over the last four years!

I would like to say a big thanks to Dee, Leanne, Anna, and all my other friends and colleagues, for their support and encouragement throughout the training process. Also, a huge thanks to my best friends, Lindsay and Vicki especially, for being there for me from a distance, and for always trying so hard to understand how hard this process has been.

I would like to extend a very special thanks to Marlene, for kindly taking me into your home and rescuing me from the local resident avian robots!

I am dedicating this thesis to Mum, Dad, and Ste. I would never have got this far without your endless support, love, and unwavering faith in my ability to do anything I put my mind to. You have always believed in me, especially when I didn't believe in myself.

And last but certainly not least, a heartfelt thanks to Nic. For your infinite patience, support, and understanding at the busiest and most stressful time in my career to date. I feel very lucky to have you in my life, and you have been a welcome ray of sunshine in the midst of the thesis cloud! Thank you.

OVERVIEW OF THESIS PORTFOLIO

A main abstract provides an overall summary of the thesis portfolio aims, findings, and implications. Chapter One is an introduction outlining the background to the thesis.

Chapter Two consists of a systematic review of non-pharmacological interventions for post-stroke depression (PSD). This review focuses specifically on some of the issues highlighted in previous studies when designing and evaluating interventions for PSD, and was prepared for submission to the Journal *Rehabilitation Psychology*.

Chapter Three is the empirical research study written in the form of a journal article, using Grounded Theory to explore specialist stroke professionals' conceptualisations of post-stroke emotional lability (PSEL). This journal article was also prepared for submission to the Journal *Rehabilitation Psychology*.

Chapter Four provides a detailed outline of the methodology adopted for the qualitative study reported in the journal article. It was more practical to include a detailed account of the methodology in a separate chapter rather than attempting to include this within the methodology section of the journal article.

The thesis portfolio adopts the British Psychological Society's editorial style (BPS, 2004) throughout. The systematic review and journal article are exceptions, where American Psychological Association (2006) referencing style will be used in line with requirements for the Journal *Rehabilitation Psychology*.

PORTFOLIO THESIS ABSTRACT

Aims: A review of the literature on non-pharmacological interventions for post-stroke depression (PSD) was completed with the aim of examining issues regarding the design and methodology of trials for non-pharmacological interventions for PSD. The empirical research study used a constructivist Grounded Theory approach to explore specialist stroke professionals' conceptualisations of post-stroke emotional lability (PSEL). The author also aimed to examine how staff experienced, understood, and identified PSEL in clinical practice, and particularly how they differentiated it from PSD.

Method: A systematic review of RCT and non-RCT studies was conducted to address the first aim. In relation to the second aim, a qualitative investigation of specialist professionals' conceptualisations of PSEL was conducted using Charmaz's (2006) version of constructivist Grounded Theory.

Results: The systematic review highlighted the importance of appropriate sampling methods, multiple treatment design, rigorous data collection, the implementation and monitoring of interventions, assessment of participant suitability for the intervention, and issues related to long-term sustainability (follow-up) when designing and evaluating non-pharmacological interventions for PSD. The qualitative investigation yielded a conceptual model of how specialist professionals conceptualise and identify PSEL in their clinical practice.

Conclusions and implications: The findings from the systematic review and the qualitative investigation highlighted a range of issues for specialist stroke services. The systematic review emphasised the importance of further evaluation and consideration of carefully planned randomised controlled trials for investigating non-pharmacological interventions for PSD. The qualitative study indicated a need for further guidance on the assessment and identification of PSEL, training for staff, and further examination of the causes of PSEL.

Keywords: *post-stroke depression, emotional lability, identification, randomised controlled trials*

CHAPTER ONE: INTRODUCTION TO THESIS

Stroke can be a frightening and challenging condition. The onset can be sudden and traumatic, and individuals are faced with the threat of death and disability (Lincoln, Kneebone, Macniven & Morris, 2012). In this context, it is not surprising that emotional problems are so common. Emotional problems after stroke include clinical mood disorders such as depression and anxiety, and also those disorders related to organic brain damage, such as post-stroke emotional lability (PSEL), anger or aggression, and mania (Lincoln *et al.*, 2012).

Post-stroke depression (PSD)

PSD is the most common post-stroke mood disorder, affecting approximately one in three stroke survivors (Hackett, Yapa, Parag & Anderson, 2005; Robinson, 1997). PSD is characterised by low mood, loss of interest and pleasure in activity, suicidal thoughts, poor motivation, changes in sleep and appetite, problems with thinking and concentration, and feelings of guilt, worthlessness, or hopelessness (American Psychiatric Association, 1994).

It is well known that depression after stroke is a particularly important factor determining long-term outcome, including increased risk of subsequent mortality (House, Knapp, Bamford & Vail, 2001; Townend, Whyte, Desborough, Crimmins, Markus *et al.*, 2007), quality of life (Bays, 2001), functional recovery and level of disability (Pohjasvarra, Vataja, Leppavouri, Kaste, & Erkinjuntti, 2001).

Post-stroke emotional lability (PSEL)

PSEL is a disorder of emotional behaviour control that occurs as a result of neurological damage, where an individual is unable to control crying or laughing. It is one of the most common post-stroke behavioural syndromes, and occurs as a result of brain lesions and diseases of the central nervous system (Feinstein, Feinstein, Gray & O'Connor, 1999). PSEL has been labelled using a variety of different terms, including 'pseudobulbar affect', 'pseudobulbar palsy', 'emotional incontinence', and 'emotionalism', and the terminology

used to describe PSEL is used inconsistently across the literature (Allman, Hope & Fairburn, 1990). The essential feature of PSEL is an increase in and lack of control over emotional behaviour (Hackett, Yang, Anderson, Horrocks & House, 2010).

Prevalence figures for PSEL remain somewhat unclear, and although the general consensus is that PSEL affects one in four stroke survivors (House *et al.*, 2008), prevalence rates have been found to vary between 15 and 50 per cent (House *et al.*, 1989; Kim & Choi-Kwon, 2000). Symptoms of PSEL tend to reduce over time, however it continues to affect one in 10 patients a year after stroke (House *et al.*, 2010), and for some, the disorder can be pervasive.

Rationale for this Thesis

A systematic review of interventions for PSD has revealed some evidence to support the use of pharmacotherapy, however, as is often the case with drug treatments, trials have also found evidence of associated adverse effects such as central nervous system and gastrointestinal problems (Hackett, Anderson, House & Xia, 2008). There have been no studies to date that have confirmed the efficacy of non-pharmacological interventions (Hackett *et al.*, 2008b; Knapp, Young, House & Forster, 2000; Kneebone & Dunmore, 2000). This is despite evidence to suggest the applicability of certain non-pharmacological interventions for PSD, in particular, cognitive behavioural interventions (Broomfield, Laidlaw, Hickabottom, Murray, Gillespie *et al.*, 2011; Kneebone & Dunmore, 2000; Lincoln & Flannaghan, 2003). PSD is a complex disorder (Broomfield *et al.*, 2010), thus interventions for it are also likely to be complex. Furthermore, complex interventions are difficult to develop and evaluate (Craig, Dieppe, Macintyre, Michie, Petticrew *et al.*, 2008). Craig *et al.* (2008) outline what makes an intervention complex, and they refer to the number of interactions between components within control and experimental interventions, the difficulty and number of behaviours required by those delivering or receiving the intervention, and the variability and number of possible outcomes.

Individuals who experience PSEL often constrict their social lives because of embarrassment, and unsurprisingly it is not uncommon for sufferers to experience depression and/or anxiety in addition to, and as a result of, their PSEL (Anderson,

Vestergaard, Ingeman-Nielsen & Lauritzen, 1995; Calvert, Knapp & House, 1998; Carota, Berney, Aybeck, Iaria, Staub *et al.*, 2005; Hackett *et al.*, 2010). The well established association between PSEL and PSD, in addition to an inconsistent use of terminology when referring to PSEL, and confusion between disorders of emotional expression and clinical mood disorders, have been some of the main sources of problems for professionals in attempting to identify PSEL in clinical practice (Cummings, Arciniegas, Brooks, Herndon, Lauterbach *et al.*, 2006). These issues related to identification of PSEL may also explain some of the discrepancies in prevalence figures.

Gillespie and Cadden (in preparation) underlined a number of important issues regarding the conceptualisation and experience of PSEL amongst specialist stroke service staff. In their study, they discovered that many members of specialist stroke staff reported that the number of individuals that they saw with PSEL in their day-to-day practice was less than prevalence rates would indicate. Some professionals said that they rarely or never saw it, and others reported not knowing how to manage it when they did see it. Similar to the reported findings of Cummings *et al.* (2006), there also appeared to be some confusion between PSEL and PSD, and staff did not report feeling confident in distinguishing one from the other (Gillespie & Cadden, in preparation).

Miller, Pratt and Schiffer (2011) reported that although the understanding of the aetiology of PSEL has advanced in recent years, it continues to be poorly understood, under-diagnosed, and under-treated by professionals. In addition, guidelines as to its classification and management (*cf.* Champion, 2006; Intercollegiate Stroke Working Party, 2012) are not directly derived from an evidence base (Hackett *et al.*, 2012; SIGN, 2010). Stroke guidelines (Intercollegiate Stroke Working Party, 2012; SIGN, 2010) currently refer to education, psychological support, and advice for patients with PSEL and their families (SIGN, 2010, p. 45).

In light of evidence of poor understanding of PSEL (Cummins *et al.*, 2006; Miller *et al.*, 2011) and guidelines advocating for education, support and advice, it is imperative to know exactly how specialist professionals conceptualise and understand PSEL, so that services can be sure that patients and families are receiving appropriate and accurate

information. Different and conflicting information and advice from staff is likely to result in negative psychological outcomes such as confusion and anxiety for both patients and their families. Consequently, a qualitative investigation of specialist professionals' conceptualisations of PSEL was undertaken by the author and constitutes part of this portfolio thesis.

CHAPTER TWO: SYSTEMATIC REVIEW

A systematic review of the problems with the evidence for non-pharmacological interventions for depression following stroke: Lessons learned.

This chapter includes a systematic review of the research literature exploring non-pharmacological interventions for depression following stroke (also referred to as post-stroke depression, abbreviated to PSD). An abstract summarises the findings, followed by a background summary of the evidence for interventions for PSD and consideration of how this review adds to those that have already been undertaken.

A systematic literature search was conducted, and details of the review method and criteria for selection that were used are presented, followed by the results of the search. The implications of the findings of this systematic review are then presented in a summary of findings and synthesis of the results, followed by a discussion and conclusions.

The systematic review was prepared for submission to the Journal *Rehabilitation Psychology*. As specified by this journal, formatting and references follow APA (2009) style (see Appendix 1 for these guidelines in full).

Systematic Review Abstract

Background: Previous systematic reviews have not found support for non-pharmacological interventions for post-stroke depression (PSD), despite evidence to suggest their feasibility and applicability. Reviews have tended to exclude non-RCT studies, although consideration of both the experimental and non-experimental literature is recommended when developing and evaluating complex interventions for specific health conditions (Craig, Deippe, McIntyre, Michie, Nazareth *et al*, 2008).

Objectives: This review systematically examined experimental and non-experimental studies of the non-pharmacological treatment of PSD. It focused on the reasons why studies did not produce favourable results, with the aim of informing future research trials into the non-pharmacological treatment of PSD.

Method: Online electronic databases and research trial databases were searched, in addition to hand searches of reference lists. Nine papers were deemed eligible for review and were assessed according to predetermined criteria.

Results: The nine studies reviewed were assessed as being of fair to good quality. There was some evidence of support for the feasibility and applicability of non-pharmacological interventions for PSD, however, no statistically significant improvements in symptoms were reported. Issues related to sampling, multiple treatment design, data collection, the implementation and monitoring of the intervention, assessment of participant suitability for the intervention, and long-term sustainability (follow-up) were identified and discussed.

Conclusions: It is imperative that further RCTs are conducted into the efficacy of non-pharmacological interventions for PSD. These trials need to take into account the issues that have arisen for researchers in the past.

KEYWORDS: *stroke, depression, non-pharmacological interventions, cognitive-behaviour therapy, randomised-controlled trials*

Background

Post stroke depression (PSD) is the most common post-stroke mood disorder, affecting approximately one in three stroke survivors (Hackett, Yapa, Parag & Anderson, 2005; Robinson, 1997). PSD is characterised by low mood, loss of interest and pleasure in activity, suicidal thoughts, poor motivation, changes in sleep and appetite, problems with thinking and concentration, and feelings of guilt, worthlessness, or hopelessness (American Psychiatric Association, 1994).

Evidence for the treatment of PSD is limited, and it has been highlighted that despite the suggested amenability for certain treatments, research trials have not been successful at finding an approach that can be recommended for use with patients (Hackett, Anderson, House & Xia, 2008b). Craig, Dieppe, Macintyre, Michie, Petticrew *et al.* (2008) produced guidance for the development and evaluation of complex interventions in general healthcare settings, and this guidance is of particular relevance to this review.

Hackett *et al.* (2008b) undertook a systematic review for the Cochrane Collaboration that included all randomized controlled trials (RCTs) of pharmaceutical agents, psychological therapies and electroconvulsive therapy (ECT) for the treatment of depression after stroke. Thirteen pharmacological interventions were compared with placebo treatment across a total of 12 trials (n=1121). Among the psychotherapy trials, 445 participants across four trials were compared with standard care and attention-control.

Pharmacological interventions for PSD

Hackett *et al.* (2008b) concluded that pharmacotherapy treatment of depression after stroke can reduce depressive symptoms. However, Hackett *et al.* (2008b) also found that the use of pharmacotherapy can result in a range of adverse effects such as central nervous system and gastrointestinal complications (Hackett *et al.*, 2008b). Substantial levels of heterogeneity also emerged across individual studies (Hackett *et al.*, 2008b). This of course has implications for the use of medication in clinical practice, and due to the adverse effects of pharmacotherapy, alternative non-pharmacological approaches such as psychotherapy, should ideally be considered.

Hackett *et al.* (2008b) reported that since their first review on this area in 2004, four new trials of pharmacotherapy had become available. These additional studies added further evidence supporting the use of pharmacotherapy for the treatment of PSD. However, they also revealed stronger evidence of more adverse events for those receiving anti-depressants (Hackett *et al.*, 2008b).

Hackett *et al.* (2008b) explored the range of methodological limitations of the trials. They reported on issues with the variation in the types of trial participants, lack of a measurable end point, the short duration of many of the interventions, variation in tools and procedures used for the diagnosis of depression, variation in the duration between stroke onset and entry to the trial, and finally, generally poor design, outcome assessment, analysis, and interpretation of results. In light of the limitations of the studies included in the Hackett review (Hackett *et al.*, 2008b), it is only possible to draw tentative conclusions regarding pharmacotherapy treatment for PSD.

Another review of RCTs in this area that examined interventions for psychological problems in chronic stages of stroke (between six and 12 months) found that repetitive transcranial magnetic stimulation (rTMS) was the most effective treatment for depressed mood, followed by drug treatment (Mehta, Pereira, Janzen, McIntyre, McClure *et al.*, 2012). However, rTMS is a relatively novel treatment and thus it has not been possible to assess longer-term treatment impact.

Non-pharmacological interventions for PSD

Research involving brain-injured populations, particularly into the psychological treatment of mood disorders, is inherently difficult, which may account for the current paucity of research in this area (Broomfield, Laidlaw, Hickabottom, Murray, Gillespie *et al.*, 2011). Kneebone and Dunmore (2000) refer to a need to draw upon research and practice from related areas. Cognitive and behavioural treatments (CBT) in particular have been found to offer benefits to those with other neurological conditions, including multiple sclerosis and comorbid depression (Mohr & Goodkin, 1999) and those encountering emotional distress after brain injury (Bradbury, Christensen, Lau, Ruttan, Green *et al.*, 2008). In

addition, cognitive and behavioural treatments have been used successfully to ameliorate psychological difficulties in the context of other physical health disorders, such as diabetes (Lustman, Griffith, Freedland, Kissel & Clouse, 1998).

Behavioural and cognitive-behavioural therapies have been found to be as effective as medication in the treatment of depression among older people, a population more likely to experience stroke and/or other physical health conditions (e.g. Fraser, Christensen & Griffiths, 2005). Further to this, the nature of depression and the evidence surrounding the role of cognition and behaviour in PSD, points to the potential utility of psychosocial - particularly cognitive behavioural - treatments for those experiencing PSD. Indeed, Broomfield *et al.* (2010) made a strong case for augmented CBT for PSD, which included specific components from motivational interviewing and grief/adjustment work in order to address the particular pattern of psychological issues associated with PSD.

Knapp, Young, House and Forster (2000) were the first researchers to review non-drug treatment of psychosocial difficulties after stroke, such as clinical depression, anxiety and general psychological distress. Despite the evidence base relating to depression in other illnesses being indicative of its potential utility in PSD, Knapp *et al.* (2000) were not able to establish that non-drug interventions were effective for psychosocial problems after stroke. However, they considered possible reasons as to why support for these interventions may not have been revealed. The paucity of research in this area was identified as one reason, in addition to problems with methodological weaknesses and variation in timing, intensity and type of intervention (Knapp *et al.*, 2000). The need to focus more attention on the quality of the design and implementation of future trials was also highlighted, including the process of randomization to control for biases, calculation of power for sample size, the use of outcome measures, and the utility of a control group (Knapp *et al.*, 2000).

A review by Kneebone and Dunmore (2000) stated that it was not possible to draw firm conclusions regarding the efficacy of psychological approaches to the treatment of PSD due to the paucity and limitations of available studies. This particular review identified a range of issues that require consideration when evaluating the use of psychological

treatment of PSD. In addition to controlled trials, case studies and uncontrolled trials were also examined and the authors highlighted the importance of the use of formal assessment and measurement of depression (Kneebone & Dunmore, 2000). Use of psychometric questionnaires that have been standardised with stroke populations and that are suitable for those with communication and cognitive issues was recommended, in addition to quantifying the level of experience of and supervision received by therapists. The importance of having a clearly defined population for which the treatment was designed and tested was also emphasised (Kneebone & Dunmore, 2000).

Van het Hoofd, Roelse, Kwakkel, Schepers and van de Port (2011) reviewed the effect of exercise therapy on depressive symptoms after stroke. They reported that the evidence available did not allow for the conclusion that exercise had a favourable effect on PSD (van het Hoofd *et al.*, 2011). They highlighted that exercise studies had mainly focussed on physical outcomes and tended to examine depression symptoms as a secondary measure, which of course made it difficult to draw any firm conclusions regarding depression (van het Hoofd *et al.*, 2011). They concluded that future trials should be sufficiently powered and focus specifically on patients with a depression diagnosis (van het Hoofd *et al.*, 2011).

The Randomised Controlled Trial (RCT)

Chambless and Hollen (1998) concluded that a treatment should be evaluated in an RCT before it can be considered to be empirically supported. The main strength of the RCT is that they are rigorous, and allow researchers to control for unknown factors. However, RCTs do not always reveal specifically why or how an intervention worked (or indeed did not work). The answers to these questions often lie in the wider evidence base, such as the single case study and case series design (Craig, *et al.*, 2008; Kneebone & Dunmore, 2000).

Rationale for this review

PSD is a complex problem and this has implications for the development of treatment interventions. Stroke survivors present with a myriad of difficulties, including physical and

neuropsychological disability, as well as emotional issues such as depression, grief, and loss. In addition to this, treatment research trials involving populations who have a brain injury are inherently difficult, which may account for the lack of available evidence and positive findings for the non-pharmacological treatment of PTSD. Kneebone and Dunmore (2000) highlighted that historically, research has neglected to take into account the specific nature of PTSD and implications of this for the design of trials evaluating treatment interventions.

The Medical Research Council (Craig *et al.*, 2008) reported that a lack of impact in some complex intervention studies reflects 'implementation failures' rather than genuine ineffectiveness and they discuss the need for a thorough 'methodology process evaluation' (Craig *et al.*, 2008) following an intervention trial in order to identify implementation problems. This guidance also underlines the importance of considering non-experimental methods and evidence outside of the health service as part of this evaluation process (Craig *et al.*, 2008). Furthermore, this guidance highlights a number of issues relating to the standardization of the design and delivery of interventions and the challenges associated with applying these within service and policy contexts (Craig *et al.*, 2008). The role of the local and organizational context is important to consider, as well as presenting problems in the population under study, and how these might influence the standardization of the design, delivery of the intervention, and subsequent conclusions (Craig *et al.*, 2008).

In summary, a high quality systematic review of treatments for PTSD has been undertaken (Hackett *et al.*, 2008a). However, this review was limited to RCT evidence only. Other reviews have concluded that there is no clear evidence that non-pharmacological - particularly psychological - treatments are effective (Hackett *et al.*, 2008b; Knapp *et al.*, 2000; Kneebone & Dunmore, 2000). This is despite indirect evidence that is indicative of the feasibility and applicability of these interventions (e.g. Lustman *et al.*, 1998; Mohr & Goodkin, 1999; Bradbury *et al.*, 2008). Furthermore, there has been no review to date that has considered exactly why these trials may not have produced positive findings (i.e. what was missing/what was overlooked). Neither has any review to date examined other

types of non-RCT evidence such as case series and uncontrolled studies, which may assist with these answers and allow thoughtful and rigorous planning for future RCTs.

Aims of this review

Building on the recommendations from Knapp *et al.* (2000) and Kneebone and Dunmore (2000), this review aims to provide a useful overview of specific issues arising from both RCTs and non-experimental evidence that would be important to consider when designing future research trials into non-pharmacological interventions for PSD. Craig *et al.* (2008) identified that although some aspects of best practice for designing and evaluating complex interventions are available generally, there are currently no good practice guidelines for research trials for interventions for specific conditions such as PSD, or indeed other psychological sequelae of stroke. Consequently, this review identified and critically appraised the wider research literature on non-pharmacological treatment of PSD (beyond RCTs) as suggested by Craig *et al.* (2008) in order to inform the development and evaluation of future trials of non-pharmacological interventions for PSD.

Method

Protocol

A study selection protocol based on the University of York Centre for Reviews and Dissemination (CRD, 2009) was developed prior to undertaking the literature searches with the aim of minimising bias and facilitating transparency. This protocol is provided in Appendix 2. The search was conducted between 13th January and 15th February 2013.

As a first step, leading researchers in the area of PSD were contacted in order to find out whether any other similar reviews were currently taking place in order to avoid duplication. The Cochrane Database of Abstracts and Reviews of Effects (DARE) was also searched in order to confirm that no similar review had already taken place. The DARE search identified two Cochrane review articles that were somewhat related to the current review. The first was a review of RCTs examining interventions for treating PSD and has already been summarised (Hackett *et al.*, 2008b). The second, also a Cochrane publication, reviewed interventions designed to proactively prevent depression following stroke (Hackett, Anderson, House & Halteh, 2008a). These reviews were limited to RCTs and did not include studies from the wider non-RCT literature.

Eligibility

Inclusion criteria

Studies were retained for review if they met the following inclusion criteria:

- English language publication
- Full-text version available online or via the British Library
- Intervention studies
- Adult population (over 18 years of age)
- Participants diagnosed with either ischaemic or hemorrhagic stroke, and PSD
- All publication date ranges

Exclusion criteria

Studies were excluded from the review if they met one or more of the following criteria:

- Study not specific to stroke *and* depression

- Studies that did not use one of the validated assessment measures of depression (Lincoln, Kneebone, Macniven & Morris, 2012).
- Dissertation and thesis abstracts
- Medical procedural or pharmaceutical interventions
- Qualitative, observational, and prevention studies
- Conference or magazine publications
- Combination treatment studies (pharmacotherapy and psychotherapy)
- Duplicate studies

Information sources

All databases were searched from their inception. MEDLINE (1946 to January 2013), and PsycINFO (1974 to January 2013), CINAHL Plus (Cumulative Index of Nursing and Allied Health Literature; 1982 to January 2013), the Psychology and Behavioural Sciences Collection (1945 to January 2013), and Embase (1947 to February 2013) were all searched for relevant titles. The searches identified a total of 16,185 titles, which were screened for relevance. Search results were managed and saved using RefWorks and electronic database folders. Duplicates were removed automatically where possible.

Trials registers websites (www.ClinicalTrials.gov and www.who.int/trialsearch/) were also searched to identify any further relevant studies that may have been in progress or in press as these studies may not have been detected by the online databases. No further titles were identified as a result of this search. The reference lists of the final papers selected for the review were hand searched in order to identify any papers that could have been missed. Reference lists of the two Cochrane reviews were searched (Hackett *et al.*, 2008a, and Hackett *et al.*, 2008b), as well as five other relevant systematic reviews looking at treatment of PSD identified during electronic journal searches (de Man-van Ginkel, Gooskens, Schuurmans, Linderman *et al.*, 2010; Knapp, *et al.*, 2000; Kneebone & Dunmore, 2000; Mehta *et al.*, 2012; van het Hoofd *et al.*, 2011) in an attempt to capture any further studies that may have been missed in electronic journal searches.

Search strategy

The PICOS (Participants, Interventions, Comparison, Outcomes, Study Design) framework (CRD, 2009) was used when designing the parameters of the search. The use of the Medical Subject Headings (MeSH) database (United States National Library of Medicine) was also used to select appropriate search terms. The MeSH thesaurus identified ten different search terms for stroke pathology and two terms for depression. Therefore, the terms used in the search were 'stroke', 'apoplexy', 'CVA', 'cerebrovascular accident', 'haemorrhage', 'hemorrhage', 'intracranial bleed', 'vascular accident', 'SAH', and 'subarachnoid'. Each of these terms were searched in combination with 'depressi* AND treatment', 'depressi* AND therapy', 'low mood AND treatment', and 'low mood AND therapy'.

Authors and publishers of any papers that were identified as potentially relevant, but which could not be obtained electronically or via the British Library, were contacted where an email address was available. None of the authors contacted responded. There were many studies identified that may have been relevant, but were not available in English language format. There were also various titles identified that may have been relevant but full-text versions were not available online or through the British Library.

Study selection

Using the eligibility criteria outlined above, the abstracts were initially reviewed in order to determine suitability for inclusion in the full-text review. Full-text articles were then reviewed at a second stage of screening and a total of nine papers were selected for final methodological review and appraisal. A flowchart based on the PRISMA statement (Moher, Liberati, Tetzlaff & Altma, 2009) provides an overview of the study selection process and details each stage of selection (Figure 1).

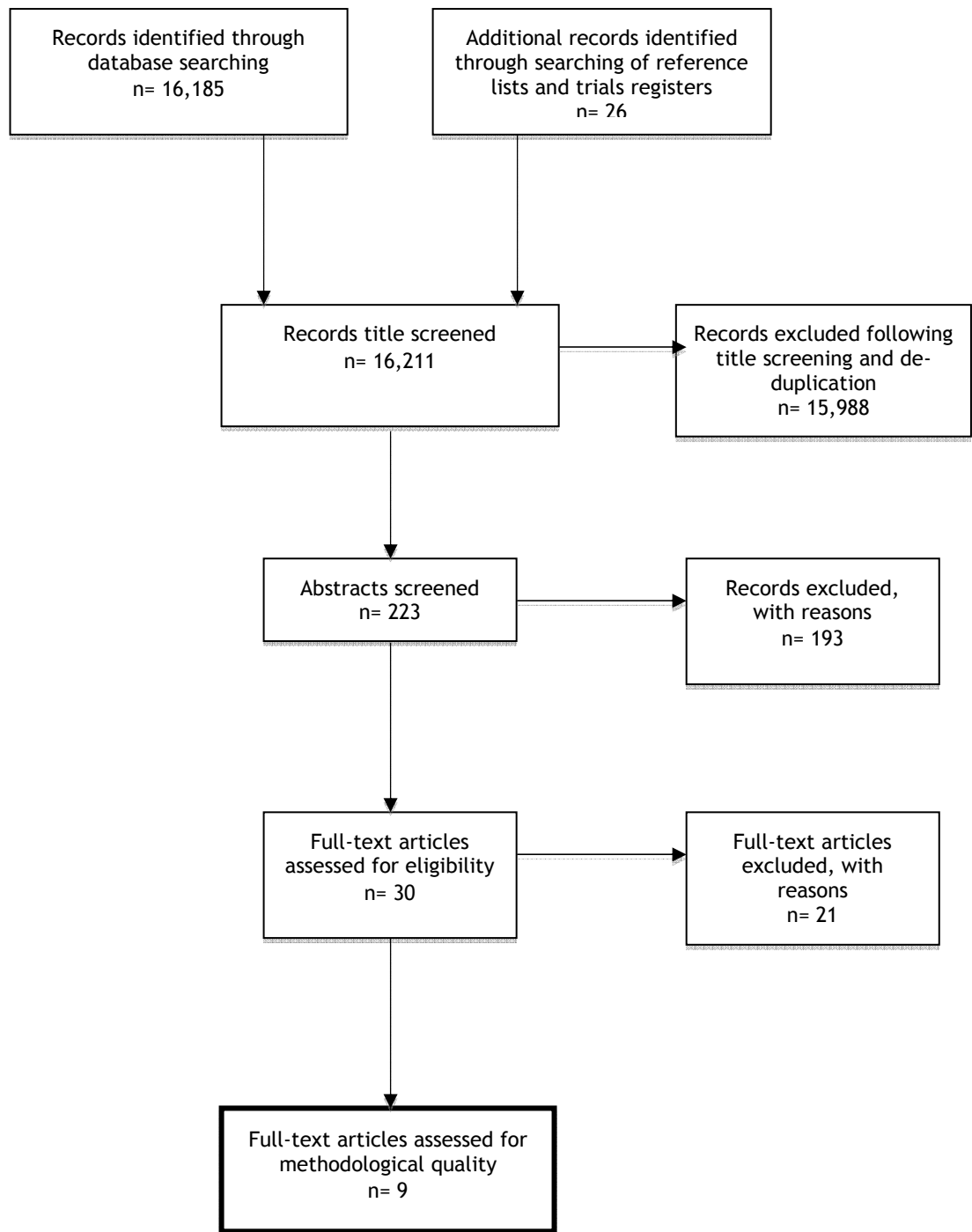


Figure 1: Flow chart of article selection (based on Moher *et al.*, 2009)

Data collection

Information was collated for each of the selected studies using the SIGN 50 checklists and study design algorithm (SIGN 2008). This included study characteristics, participant characteristics, intervention and setting, and outcome data/results. A standardised data extraction form was used (see Appendix 3), and an overview of study characteristics and findings is presented for each study reviewed in Table 2.

Quality appraisal tool

A quality assessment tool was developed for the purpose of assessing and appraising the methodological quality of the studies that met the inclusion criteria of the review (see Appendix 4). This was based on the Scottish Intercollegiate Guidelines Network guidance on systematic literature reviews (SIGN, 2008) and the University of York's Centre for Reviews and Dissemination guidance for undertaking reviews in healthcare (CRD, 2009).

Summary measures

Studies were rated using nine quality criteria items across seven different dimensions: research questions and objectives; sampling; design and method; statistical analysis; quality of reporting; generalisability; and overall quality. Numerical ratings were assigned to each element of quality criteria domain: 1= well covered; 2= adequately addressed; 3= poorly addressed; 0= not addressed/not reported/not applicable. The overall quality ratings assigned were: 3= good to excellent; 2= adequate to good; 1= poor to adequate; 0= poor. Five of the studies included in the methodological appraisal were rated by two academics, working independently of one another. The second rater was blinded to the title, author, and journal of the papers. Inter-rater reliability scores were calculated, and the overall percentage agreement was high, at 81%.

Total numerical scores were calculated for each study, and converted to percentages. For items rated as not applicable, percentage calculations were adjusted to reflect the number of applicable items. Finally, percentages were categorised after all articles had been reviewed in order to provide an overall descriptive quality rating for each study

(Good $\geq 70\%$; Fair $\geq 50\%$; Weak $< 50\%$). A detailed breakdown of ratings for each study is provided in Table 3.

Results

Study selection

As shown in Figure 1, a total of 16,211 records were identified from the literature search. Out of these, 15,988 records were removed following title screening and automatic de-duplication. The abstracts of the remaining 223 records were then screened for eligibility, and a further 214 records were excluded for a range of reasons. Table 1 provides an overview of reasons for exclusion at this stage.

Table 1: Overview of excluded studies following screening and automatic de-duplication

Number of studies excluded	Reasons for exclusion n= 214
35	Duplicate record
31	Study not specific to stroke and depression
28	Not an intervention study
27	No full-text available in English
23	Title not directly relevant
21	Dissertation abstract or conference presentation
18	Drug or medical treatment study/combination study
12	Qualitative study
11	Title not accessible
8	Participants not diagnosed with depression

Included studies

Nine quantitative studies undertaken in five different countries between 1989-2012 were selected for appraisal and review of their methodological quality (Alexopoulos, Wilkins, Marino, Kanellopoulos, *et al.*, 2012; Hibbard, Grober, Stein & Gordon, 1992; Joo, Lee, Chung & Shin, 2010; Lincoln, Flannaghan, Sutcliffe, & Rother, 1997; Lincoln & Flannaghan, 2003; Rasquin, van de Sande, Praamastra & van Heughten, 2009; Sims, Galea, Taylor, Dodd, *et al.*, 2009; Thomas, Walker, Macniven, Haworth & Lincoln, 2012; Towle, Lincoln & Mayfield, 1989). These included a total of 377 patients with a diagnosis of PSD. All selected studies examined the efficacy and applicability of a range of non-pharmacological

interventions for depression after stroke. Table 2 provides an overview of the information extracted from these nine studies.

Table 2: Overview of selected studies

<i>Study (RCTs)</i>	<i>Country</i>	<i>Sample</i>	<i>Assessment of Depression</i>	<i>Intervention</i>	<i>Main Findings</i>	<i>Limitations</i>
<i>Lincoln & Flannaghan (2003)</i>	UK	N= 123	Beck Depression Inventory (BDI), Wakefield Depression Inventory (WDI)	10 one hour sessions of cognitive behavioural psychotherapy by the same research CPN over 3 months	No significant differences in mood between the three groups (cognitive behaviour therapy, attention control, and standard care)	Small sample size, problems with method of recruitment and assessment for suitability for therapy, issues regarding use of therapists, and the quality, duration, and supervision of therapy
<i>Sims, et al. (2009)</i>	Australia	N= 45	Patient Health Questionnaire (PHQ-9), Centre for Epidemiologic Studies for Depression Scale (CES-D)	10 week, community based progressive resistance training (PRT) program	Progressive resistance training (PRT) was found to reduce symptoms of PSD, although this was not found to be statistically significant after adjusting for baseline scores. Reduction in PSD symptoms were also found to be maintained at 6-month follow up, although this was not found to be statistically significant	Small sample size, issues with method of recruitment, short term follow-up only, comparison groups received more attention than usual which may have been therapeutic, possibility of social interaction effects
<i>Thomas, et al. (2012)</i>	UK	N= 105	Stroke Aphasic Depression Questionnaire, Visual Analogue Mood Scales 'sad' item	Three month duration behaviour therapy intervention, up to 20 one- hour session	A 3-month behavioural therapy intervention significantly improved symptoms of PSD in aphasic patients, and improvements were maintained at 6-month follow-up	Small sample size, difficulties defining precise intervention, under-representation of patients with severe aphasia and mood problems
<i>Towle, et al. (1989)</i>	UK	N= 44	Wakefield Depression Inventory (WDI), General Health Questionnaire (GHQ)	16 week social work intervention	Social work involvement had no detectable effect on level of depression one year after stroke	Small sample size, and reliance on questionnaires only for diagnosis of depression

<i>Study (non-RCTs)</i>	<i>Country</i>	<i>Design</i>	<i>Sample</i>	<i>Assessment of Depression</i>	<i>Intervention</i>	<i>Main Findings</i>	<i>Limitations</i>
<i>Alexopoulos, et al. (2012)</i>	USA	Uncontrolled trial	N= 24	Patient Health Questionnaire (PHQ-9), Hamilton Depression Rating Scale (HAM-D)	Twelve weekly 45 minute sessions of Ecosystem Focussed Therapy (EFT)	EFT is more efficacious than Education on Stroke and Depression (ESD) in reducing depressive symptoms and signs, and in leading to a higher remission rate, and in ameliorating disability in PSD	Small sample size, no control group, raters could not be blinded to the treatment condition, same therapists used to deliver EFT and ESD
<i>Hibbard, et al. (1992)</i>	USA	Clinical case study	N= 1	Beck Depression Inventory (BDI), Hamilton Rating Scale for Depression (HAM-D)	Individually tailored cognitive behavioural psychotherapy	The patient's symptoms of depression were found to have improved at 6-month follow up. These improvements were maintained at 1-year follow up	Case study, no comparison or control, issues with generalisability
<i>Joo, et al. (2010)</i>	Korea	Clinical case series	N= 11	Beck Depression Inventory (BDI)	Eight weeks of weekly 2.5 hour long sessions of Mindfulness-Based Stress Reduction (MBSR)	The MBSR program was found to reduce depressive symptoms after aneurismal subarachnoid haemorrhage	Small sample size, and no control group. No long term follow up
<i>Lincoln, et al. (1997)</i>	UK	Series of AB single case experimental designs	N= 19	Beck Depression Inventory (BDI), Hospital Anxiety and Depression Scale (HADS), Wakefield Depression Inventory (WDI)	Three months of cognitive behavioural treatment delivered by a CPN and an assistant psychologist	Cognitive behavioural therapy is an appropriate and feasible treatment for some patients, and some patients benefitted from the treatment	Small sample size, no control group, problems with recruitment and data collection, issues with generalisability, and no long term follow-up
<i>Rasquin, et al. (2009)</i>	Netherlands	Single-subject quasi experimental design (SSED)	N= 5	Beck Depression Inventory (BDI), Symptom Checklist Depression Scale (SCL-90-D), Check-List for Cognitive and Emotional Consequences of Stroke (CLCE-24)	Eight weeks of weekly one-hour sessions of cognitive behavioural intervention with a psychologist	Cognitive behaviour therapy is a feasible intervention for reducing symptoms of PSD	Small sample size, issues with generalisability, difficulties with controlling for confounding effects, short follow-up, no control group

Methodological quality appraisal

A summary of each paper's methodological ratings on each domain is provided in Table 3, in addition to total scores, percentage scores and corresponding methodological quality categories.

Research question and objectives

All nine studies addressed an appropriate and clearly focused question, drawn from a theoretical model or previous research on the non-pharmacological treatment of PSD. Lincoln and Flannaghan (2003), Lincoln *et al.* (1997), Hibbard *et al.* (1992), Rasquin *et al.* (2009), and Thomas *et al.* (2012) all considered behavioural and cognitive-behavioural theory in relation to PSD. Lincoln *et al.* (1997) was a pilot study in preparation for the Lincoln and Flannaghan (2003) trial.

All studies highlighted the prevalence and impact of PSD (although this varied), and how there is a lack of good quality evidence to guide treatment. Some of the studies also highlighted how PSD lends itself to certain types of non-pharmacological treatment approaches, and specifically to cognitive behavioural therapy (Lincoln *et al.*, 1997; Lincoln & Flannaghan, 2003; Thomas *et al.*, 2012). Hibbard *et al.* (1992) also reported the increasing demand for support for individuals experiencing psychological consequences of stroke due to demographic population change and advances in the medical treatment of stroke. Some of the studies also discussed the impact of low mood on rehabilitation, and Sims *et al.* (2009) examined this as one of the aims of their study.

For the four RCTs (Lincoln & Flannaghan, 2003; Sims *et al.*, 2009; Thomas *et al.*, 2012; Towle *et al.*, 1989), the specific areas of randomization, concealment of treatment method, similarity between treatment and control participants, standardization, validity and reliability of outcome measurement, intention to treat analysis, and comparison of multicentre data were assessed, as recommended by SIGN (2008). The highest scoring study on this dimension was the Thomas *et al.* (2012) trial, followed by Lincoln and

Flannaghan (2003). These two studies also gave the most comprehensive qualitative evaluation of their intervention.

In terms of the non-RCT studies (Alexopoulos *et al.*, 2012; Hibbard *et al.*, 1992; Joo *et al.*, 2010; Lincoln *et al.*, 1997; Rasquin *et al.*, 2009), each article was assessed on how well the study addressed an appropriate and clearly focussed question and whether it was drawn from a theoretical model or previous research. Rasquin *et al.* (2009) considered the feasibility of a CBT intervention for this population as one of the primary aims of the study, and used a standardised feasibility questionnaire.

Sampling

The representativeness of the samples selected for each study varied. The sample in the Thomas *et al.* (2012) study was predominantly male, and some of the studies did not specify the male to female ratio or socio-economic status (Lincoln & Flannaghan, 2003, Towle *et al.*, 1989). Three of the RCTs (Lincoln & Flannaghan, 2003, Sims *et al.*, 2009, and Towle *et al.*, 1989) excluded participants with communication and cognitive problems. Conversely, Thomas *et al.* (2012) specifically recruited individuals with aphasia and low mood, and tailored their intervention to this group.

The studies varied in terms of the quantity of information provided regarding numbers invited to participate, numbers who actually participated, and numbers who declined or dropped out. Participants were recruited from hospital and rehabilitation settings and from stroke registers in six of the nine studies, (Joo *et al.*, 2010, Lincoln *et al.*, 1997, Lincoln & Flannaghan, 2003, Rasquin *et al.*, 2009, Thomas *et al.*, 2012, Towle *et al.*, 1989). Sims *et al.* (2009) recruited a proportion of their participants via newspaper articles and adverts in GP practices.

Design and method

Four of the studies were randomised controlled clinical trials (Lincoln & Flannaghan, 2003; Sims *et al.*, 2009; Thomas *et al.*, 2012; Towle *et al.*, 1989), one was an uncontrolled trial (Alexopoulos *et al.*, 2012), one was a clinical case study (Hibbard *et al.*, 1992), one a clinical case series (Joo *et al.*, 2010), one was a series of AB single case experimental designs (Lincoln *et al.*, 1997), and one was a single-subject quasi-experimental design (SSED) used with an AB design (Rasquin *et al.*, 2009). Alexopoulos *et al.* (2012) and Lincoln *et al.* (1997) were both pilot studies.

All studies used self-report questionnaires for the purpose of diagnosing depression, monitoring symptoms and measuring intervention outcome. Joo *et al.*, (2010) did not supplement any of their depression self-report questionnaires with data from another source for diagnosing depression or confirming eligibility for the study, such as a clinical interview. In one study, participants were telephoned for an assessment appointment to confirm diagnosis (Sims *et al.*, 2009). Hibbard *et al.* (1992) interviewed both the participant and his wife, in addition to completing outcome measures to confirm diagnosis of depression. One study (Thomas *et al.*, 2012) also included observational measures of mood completed by a carer or relative and the clinician delivering the intervention.

The time period between intervention and administration of follow-up measurement of depression symptoms varied across studies. Alexopoulos *et al.* (2012), Joo *et al.* (2010), and Lincoln *et al.* (1997) completed pre- and post-intervention measures only. Rasquin *et al.* (2009) obtained follow-up at three months. Sims *et al.* (2009) obtained follow-up at six months. Lincoln and Flannaghan (2003) obtained follow-up at three and six months, as did Thomas *et al.* (2012). Hibbard *et al.* (1992) measured the progress of the patient included in their case report at 12 months.

Statistical analysis

The statistical analyses employed across the nine studies was assessed to be of fair or good quality. A range of statistical analyses methods were employed where appropriate in the studies reviewed. Analyses in all of the studies were appropriately reported and justified, with some providing more extensive detail and depth of exploration than others.

Quality of reporting

Similarly, the quality of reporting across all of the studies reviewed was judged to be fair or good. Some studies provided more detail than others, but overall this seemed to be appropriate in relation to the length of the article, design of the study, and issues highlighted. It is likely that studies had been limited to journal guidelines or word limits, which would have had an impact on the amount of detail they were able to provide.

Generalisability

The generalisability ratings for the papers ranged from poor to adequate. Three of the non-RCT studies were rated as poor (Hibbard *et al.*, 1992; Joo *et al.*, 2010; Rasquin *et al.*, 2009). Two of the non-RCT studies were rated as adequate (Alexopoulos *et al.*, 2012; Lincoln *et al.*, 1997). The four RCT studies were rated as adequate in terms of generalisability (Lincoln & Flannaghan, 2003; Sims *et al.*, 2009; Thomas *et al.*, 2012; Towle *et al.* 1989). The main factors that undermined the generalisability of the studies were small sample sizes and issues related to recruitment. None of the RCT studies were suitably powered to produce reliable, valid and generalisable results. Thomas *et al.* (2012) reported a multi-centre study, although the study was restricted to aphasic patients. They also identified that their study criteria excluded participants with severe aphasia (Thomas *et al.*, 2012).

Table 3: Overview of methodological quality ratings

RCT studies:	<i>Thomas, et al. (2012)</i>	<i>Lincoln & Flannaghan (2003)</i>	<i>Sims, et al. (2009)</i>	<i>Towle, et al. (1989)</i>	Non-RCT studies:	<i>Rasquin, et al. (2009)</i>	<i>Lincoln, et al. (1997)</i>	<i>Alexopolous, et al. (2012)</i>	<i>Hibbard, et al. (1992)</i>	<i>Joo, et al. (2010)</i>
Percentage and overall descriptive category	85% Good	83% Good	78% Good	59% Fair	Percentage and overall descriptive category	86% Good	76% Good	67% Fair	60% Fair	50% Fair
Total score (max= 54)	46	45	42	32	Total score (max= 30)	26	23	20	18	15
Research question and objectives (max= 27)	27	23	21	15	Research question and objectives (max= 3)	3	3	2	2	2
Sampling (max= 6)	5	6	5	3	Sampling (max= 6)	6	5	4	2	2
Design and method (max= 9)	7	7	7	6	Design and method (max= 9)	8	6	5	7	4
Statistical analysis (max= 3)	2	3	3	2	Statistical analysis (max= 3)	2	3	2	2	2
Quality of reporting (max= 3)	3	2	2	2	Quality of reporting (max= 3)	3	2	3	2	2
Generalisability (max= 3)	2	2	2	2	Generalisability (max= 3)	1	2	2	1	1
Overall assessment of study (max= 3)	3	2	2	2	Overall assessment of study (max= 3)	3	2	2	2	2

Synthesis of results

The designs of the studies included in this review each had their own individual strengths and limitations. Thomas *et al.* (2012) and Lincoln and Flannaghan (2003) were the most rigorous in terms of design and methodology. They both achieved high quality ratings for clearly defined and operationalised variables, adequate validity and reliability of variable measurement, and also accounted for the influence of confounding variables where possible (Thomas *et al.*, 2012; Lincoln & Flannaghan, 2003). As discussed earlier, RCTs are rigorous and can control for unknown factors, but they do not always offer the qualitative detail that non-experimental designs can in relation to reasons why an intervention has not proven effective despite evidence to suggest feasibility (Craig *et al.*, 2008).

As with most 'small *n*' and non-experimental designs, the main limitations of the non-RCT studies were lack of control of confounding effects (medication, other therapies/rehabilitation, life events, etc.) and generalisability. Alexopoulos *et al.* (2012), Hibbard *et al.* (1992), Joo *et al.* (2010), Lincoln *et al.* (1997), and Rasquin *et al.* (2009) did not have comparison or attention control groups, thus it is not possible to conclude that confounding effects were controlled for and managed in any of these studies. As outlined by Craig *et al.* (2008), these designs may not contribute directly to our knowledge regarding the efficacy of PSD interventions, nevertheless qualitatively, these studies offer relevant contributions to the evidence base in relation to assisting researchers to understand the detailed components of the interventions and illuminating potential problems with the intervention implementation.

Sampling

Two of the most significant issues reported in the RCT studies included in this review were problems related to sample size and generalisability. None of the studies included in this review achieved statistical power and many of them did not provide a power calculation, which meant that it was not possible to draw firm conclusions regarding the

efficacy of the treatment and reliability of the results. These findings illustrate the importance of the use of power calculations and effect sizes in large trials (Cohen, 1988).

Six of the studies reported problems with recruitment (Alexopoulos *et al.*, 2012; Lincoln *et al.*, 1997; Lincoln & Flannaghan, 2003; Sims *et al.*, 2009; Thomas *et al.*, 2012; Towle *et al.*, 1989). Four of the studies used opt-in return postal questionnaires as their recruitment method (Alexopoulos *et al.*, 2012; Lincoln *et al.*, 1997; Lincoln & Flannaghan, 2003; Towle *et al.*, 1989). Lincoln and Flannaghan (2003) considered their sample to possibly be under-representative of those with severe symptoms, as eligible participants who were more severely depressed may not have completed and returned questionnaires as a direct result of their depressive symptoms. Similarly, Sims *et al.* (2009) recruited a proportion of their participants via newspaper articles and adverts in GP practices, which could have had implications for the reliability and validity of the results in the sense that it is possible their sample only captured individuals motivated and well enough to contact researchers to express their interest.

The Thomas *et al.* (2012) study was carried out and results compared across multi-centre sites, which contributes to the external validity, reliability and generalisability of the results. Friedman, Furberg and DeMets (1998) referred to the benefits of multi-centre trials and urged consideration of uniformly agreed and executed protocols, confidence in data integrity, and protection of participant rights when designing multi-centre trials.

Multiple treatment designs

Inclusion of a control group is essential to experimental design and methodology and enables the influence of confounding effects to be controlled for in trials where experimental conditions are complex. Two of the studies included in this review argued that even an attention-placebo control group can have some therapeutic effect, which can obscure the potential positive outcomes of an 'active' intervention (Alexopoulos *et*

al., 2010; Lincoln *et al.*, 1997). In addition to the attention-placebo group usually included in RCTs, Lincoln and Flannaghan (2003) used a third 'no intervention' group. The use of this additional no intervention group enabled the researchers to be more confident about the reliability and validity of their findings by eliminating the possibility of the therapeutic effect of an attention-placebo control group, although blinding was an issue for this group. Multiple treatment designs like this also allow for further exploration of other relative interventions, and permit the calculation of the absolute effectiveness of each intervention, compared with the effectiveness of no intervention at all (Chambless & Hollon, 1998; Keppel & Wickens, 1982).

Data collection

Studies that did not use psychometric measures of depression that had been validated for stroke populations were excluded from this review. Kneebone and Dunmore (2000) highlighted the requirement for formal assessment measures in intervention studies for PSD in order to be able to make empirically based conclusions. Lincoln *et al.* (2012) provided a comprehensive list of measures that have been validated for stroke populations and use of psychometric measures is always recommended in addition to a clinical interview (Lincoln *et al.*, 2012).

It is well known that sole reliance on psychometric measures for participant selection and symptom monitoring in either research or clinical work should be avoided (British Psychological Society, 2011; Chambless & Hollon, 1998). This systematic review has further highlighted the importance of completing more extensive assessment of depressive symptoms for both participant selection and for the purpose of tracking symptom change over time. Two of the nine studies included in this review (Joo *et al.*, 2010, Towle *et al.*, 1989) used self-report measures to confirm symptoms of depression for the purpose of recruiting participants to their study and also to monitor change over time. Additionally, Lincoln *et al.* (2012) recommended the use of psychometric measures as well as a clinical interview when undertaking research in this population. Hibbard *et*

al. (1997) obtained measures and information from both the participant's partner and clinician and these data indicated that the participant may have been minimising his emotional distress, emphasising the utility of comprehensive data collection.

The frequency and accuracy of symptom measurement is also of relevance. Rasquin *et al.* (2000) utilized session-by-session outcome measures, as is now advised as good practice in the context of mental health and wellbeing (IAPT, 2011). They also used the Reliable Change Index (RCI) to define clinically significant change over time (Jacobson & Truax, 1991). Both of these processes added to the reliability and validity of the data from this study. Regular mood measurement also contributed to the reliability of these studies and is a key strength. Recent good practice guidance on recording and monitoring outcomes in mental health and wellbeing advise that in order to reliably measure change, at least weekly/session-by-session measures should be taken (IAPT, 2011).

Descriptions of the severity of depression symptoms, as well as severity and type of stroke and resulting disability varied across the studies. Towle *et al.* (1989) considered location of stroke lesion, and in addition to Lincoln and Flannaghan (2003), included details of length of time since the participant's stroke and the number of strokes they had experienced. Lincoln and Flannaghan (2003) considered this information during randomization to treatment groups and analysis of results. The non-RCT studies were able to examine these variables in considerably more detail and discussed these in relation to proposed implications for the intervention and outcome. Rasquin *et al.* (2009) did this particularly well, monitoring symptom severity on a session-by-session basis. Sims *et al.* (2009) used the Present State Exam (PSE) depression module (Wing, Cooper & Sartorius, 1974), completed by a psychiatrist to ascertain depression severity and this was considered when analysing the data.

Implementation and monitoring of intervention

Chambless and Hollen (1998) and Kneebone and Dunmore (2000) refer to the importance of treatment manualisation in intervention studies. Chambless and Hollon (1998) argue that unless an intervention is very simple, studies that do not follow a manualised treatment are of limited value as it is not possible to know exactly what was tested, nor repeat the intervention with fidelity. One of the studies in this review utilised a treatment manual (Lincoln & Flannaghan, 2003) that had been produced in a pilot study (Lincoln *et al.*, 1997). Alexoloulos *et al.* (2012) make reference to manuals used for training staff in the delivery of therapy, but not explicitly in the delivery of the intervention under investigation. Alexopoulos *et al.* (2012) did however use audio recordings of therapy sessions and examined treatment fidelity using a fidelity scale to enable evaluation of how faithfully therapists were adhering to the treatment approach, which was a strength of the study.

The importance of the training, experience, and supervision of therapists delivering an intervention has been highlighted by Chambless and Hollon (1998), Kneebone and Dunmore (2000), and Lincoln and Flannaghan (2003). In the Thomas *et al.* (2012) study, an assistant psychologist delivered the intervention, supervised by a qualified clinical psychologist. It has been argued in the past that there are times when these issues are of little importance (Christensen & Jacobson, 1994), for example, if the intervention being delivered is relatively straightforward as in the case in the Thomas *et al.* (2012) study. However, the experience of the therapist is likely to be of greater importance when delivering complex interventions requiring a higher level of skill.

Lincoln and Flannaghan (2003) also referred to the duration and intensity of the intervention, and how this might have contributed to the lack of positive outcome in their study i.e. they did not offer enough of the intervention. The pilot study (Lincoln *et al.*, 1997) found that on average, improvement in mood was observed at 8.4 sessions.

However, other studies using similar populations and interventions identified gains with higher intensity interventions of longer duration (Koder, Brodaty, & Anstey, 1996).

Assessment of patient suitability for intervention

In addition to assessment of treatment feasibility (Rasquin *et al.*, 2009), and to the intensity and duration of the intervention, the studies also highlighted the importance of participant suitability for the intervention. Hibbard *et al.* (1992), Lincoln *et al.* (1997), Lincoln and Flannaghan (2003), and Rasquin *et al.* (2009) all emphasised the importance of emotional/psychological insight to an individual's ability to engage in and benefit from an intervention.

Assessment of sustainability of the intervention

Information regarding long-term effects of interventions is important to consider when examining the quality of complex interventions. It is impossible to be confident that an effect of an intervention is sustained over time without long-term follow-up (Chambless & Hollon, 1998). Mitchell, Teri, Ceith, Buzatis, Tirschwell, *et al.* (2008) highlighted the need for long-term (at least 12 month) follow-up in relation to treatment outcome, even for short interventions. Guidance and protocol for follow-up data differ depending on the nature of the disorder, but guidance on interventions for complex mental health disorders state that longer follow-up assessment is desirable when developing complex interventions (Jacobson & Christensen, 1996).

Discussion

Previous systematic reviews of the non-pharmacological treatment of PSD have concluded that there is no clear evidence that non-pharmacological - particularly psychological - treatments are effective (de Man-van Ginkel *et al.*, 2010; Hackett *et al.*, 2008a; Knapp *et al.*, 2000; Kneebone & Dunmore, 2000; Mehta *et al.*, 2012; van het Hoofd *et al.*, 2011), despite evidence to suggest the applicability and feasibility of these interventions for PSD (e.g. Lustman *et al.*, 1998; Mohr & Goodkin, 1999; Bradbury *et al.*, 2008), thus conclusions have been drawn based on a limited pool of studies and several limitations of sole reliance on RCTs have been outlined. These include the limited depth of information that can be gained from RCTs in terms of the processes and mechanisms underpinning interventions, what works for whom, and the reasons behind this.

RCT studies also often neglect consideration of the wide range of factors that may account for outcomes in intervention studies for PSD, whereas more detailed studies using other designs can capture this information. Furthermore, RCTs do not generally yield qualitative data in relation to the experiences of the patient, their families, and staff supporting them. This information is also important and may provide more detailed insight into quantitative outcomes and barriers to successful intervention.

The nine papers evaluated in this systematic review revealed a number of issues regarding the problems encountered in studies examining non-pharmacological interventions for PSD. Furthermore, it has been suggested that the outcomes of these studies may be due to intervention implementation failures and methodological issues, rather than genuine ineffectiveness of the interventions (Craig *et al.*, 2008). PSD is a complex multifaceted disorder, of which stroke professionals have a limited understanding, and interventions for the management of PSD are still being developed. As with any complex disorder, interventions for the management of PSD are also complex (Broomfield *et al.*, 2010; Lincoln *et al.*, 2012), and research trials require careful

consideration and planning (Craig *et al.*, 2008).

The synthesis of the findings from this review will contribute to the systematic review literature on non-pharmacological interventions for PSD, providing a useful overview of relevant issues from both experimental and non-experimental evidence to consider when designing future research trials evaluating the efficacy of non-pharmacological treatments of PSD.

Implications

This review explored a number of issues related to sampling and recruitment in studies of non-pharmacological interventions for PSD. Lincoln *et al.* (1997) outlined the reasons why patients did not opt into their study and discovered that the main reason was that they did not view themselves as depressed. This finding highlights how patient education and promotion of awareness of PSD might be indicated when offering patients the opportunity to participate in a research trial, in addition to educating them regarding the rationale of the study. Patel, Doku and Tennakoon (2003) referred to the potential problems with recruitment of research participants and suggested ways of managing these issues when designing trials, such as collecting preliminary data on issues that might prevent or deter patients from accessing research trials. Patel *et al.* (2003) also recommended consideration of the participants' perspectives regarding recruitment procedures and forming good alliances with collaborators.

A number of issues related to data collection have also been outlined in this systematic review, including incomplete datasets and the unreliability of self-report data. Lincoln *et al.* (1997) reported that patients tended to minimise their distress, and Hibbard *et al.* (1992) suggested that this is a common post-stroke pattern reflective of a patient's overall lack of awareness. The British Psychological Society (2011) research guidelines recommend that a broad-based approach to data collection is likely to control for some of these issues, and strengthens validity, reliability, and generalisability of data. IAPT

(2011) provided a framework on the importance of rigorous collection of outcome data in the context of mental health and wellbeing (the 'IAPT Data Standard'). Since implementing this framework, a significant increase in the completion of patient outcome data has been observed (IAPT, 2011). Integration of outcome measures into clinical sessions and demonstration of the relevance of their completion with patients has also been recommended (IAPT, 2011). This is based on the rationale that if patients can see the usefulness of outcome measures and can see their progress in therapy plotted on a graph, they are more likely to fill the measures in. The IAPT Data Standard (IAPT, 2011) also discussed the use of session-by-session outcome measures for progress monitoring purposes. This approach to data collection also means that even if a patient drops out of treatment, end of treatment data will always be available from their final session.

The role of therapist experience and supervision in relation to those delivering interventions has also been discussed in this systematic review. Chambless and Hollon (1998) and Lincoln and Flannaghan (2003) emphasised the importance of specialist training, experience and supervision of therapists when delivering complex interventions and working with more complex conditions. Broomfield *et al.* (2010) presented the case for augmented CBT for PSD, which included elements of motivational interviewing and grief work in addition to CBT. This sort of intervention is likely to require a high level of skill and experience, and provision of regular, ongoing clinical supervision for those delivering it to people with PSD.

The issues of manualisation, treatment fidelity and careful monitoring of interventions to control for variation that might undermine potential successful outcomes have also been raised as part of this systematic review. Lincoln and Flannaghan (2003) attempted to video record sessions as a way of monitoring their intervention, but were forced to discontinue with this due to patients feeling uncomfortable about being recorded in their homes. However, Alexopoulos *et al.* (2012) managed to maintain a reasonable stance by

using audio instead of video recording and using a fidelity scale. Craig *et al.* (2008) also reported that adhering to a strict fidelity protocol may be inappropriate, and more a more flexible approach to treatment delivery may be more favourable under certain circumstances. In addition to this, they recommended consideration of the context of the local setting (Craig *et al.*, 2008).

As discussed by Lincoln and Flannaghan (2003) and Rasquin *et al.* (2009), patient suitability for interventions needs to be taken into account. Chambless and Hollon (1998) referred to the importance of compliance with the intervention model and reported that patients could fail to benefit from interventions because they were unwilling or unable to adhere to their recommended regime. Lincoln and Flannaghan (2003) and Rasquin *et al.* (2009) made the valid point that in clinical practice, individuals are assessed for suitability before being allocated to an intervention. However, randomisation procedures can make replication of this process in research more complicated and can also introduce bias into a study.

This is the first systematic review guided by recommendations from the MRC (Craig *et al.*, 2008) that has considered both experimental and non-experimental research with a specific focus on attempting to examine the reasons why previous support has not been found for non-pharmacological - particularly psychological - interventions for PSD despite evidence to suggest their feasibility and applicability. The findings of this review thus offer advice and guidance for future researchers when designing research trials for non-pharmacological interventions for PSD.

Limitations of this review

Some of the points raised in this systematic review highlight the reality of the constraints of field research and how it is not always possible to fully exclude potential problems that may arise as a result of conducting field research, particularly with a vulnerable and complex clinical population. This systematic review was limited due to various resource

constraints. The reviewer did not have access to translation services, which meant that studies that were not published in English were excluded. The papers reviewed were therefore mainly from the UK, USA, and Australia, which automatically introduces a bias against research conducted in non-Western and non-English speaking countries. It is possible that PSD and interventions for its management might appear differently in different cultures. During the search process, the researcher noted a number of studies emerging from Eastern and Asian cultures on the use of acupuncture for the treatment of PSD, but was unable to access the articles to review them. Also, a full meta-analysis of the included studies would have been desirable to inform a more thorough process evaluation. However, this was outwith resources and beyond the scope of this review.

This review excluded qualitative studies in order to maintain consistency of methodological appraisal. However, following consideration of the findings, particularly regarding stroke patients' perceptions of depression after stroke highlighted by Lincoln *et al.* (1997), it is likely to have been informative to explore patient experiences of non-pharmacological interventions for PSD, as well as their ideas and conceptualisations of the condition itself.

Conclusions

This review has systematically examined the existing literature evaluating non-pharmacological interventions for PSD. The key findings of the nine studies reviewed have been summarised, based on the outcomes of both experimental (RCTs) and non-experimental evidence (such as case series and small *n* designs). This was with the aim of facilitating researchers' understanding of the 'teething problems' (Craig *et al.*, 2008) encountered by researchers in this specialist area when developing and evaluating non-pharmacological interventions for PSD.

Methodological issues in this area of research have been considered in detail and the nine studies reviewed have been appraised in terms of their methodological quality.

Furthermore, a number of issues regarding the implementation of non-pharmacological interventions for PSD have been explored. Issues identified in the nine studies reviewed concerned sampling, multiple treatment design, data collection, problems with the implementation and monitoring of interventions, assessment of participant suitability for intervention, and issues related to long-term sustainability (follow-up). The issues with implementation of trials may be responsible for the reported lack of impact of non-pharmacological PSD interventions, rather than genuine ineffectiveness given the support from the wider evidence base.

More generally, this review has drawn further attention to the fact that despite a number of research trials examining the treatment (both pharmacological, and non-pharmacological) of PSD, researchers have yet to find a treatment that has been proven safe and effective (Hackett *et al.*, 2008b). This of course presents a problem for both patients with PSD, as well as for professionals whose jobs it is to attempt to treat and care for these patients. It is obvious that further research is required in this area in order to inform treatment and practice guidelines, and this review has further highlighted the issues to consider when planning and organising future research, which should include both experimental and non-experimental designs.

It is imperative that further trials are conducted into the non-pharmacological treatment of PSD. Psychological - particularly cognitive behavioural - interventions are worthy of further exploration as they lend themselves well to PSD (Broomfield *et al.*, 2010; Lincoln *et al.*, 2012). However, future trials need to take into account the issues that have arisen for researchers in the past. Consideration of the findings of this review will contribute to the evidence base for designing and evaluating complex interventions, and while some aspects of good practice are relatively clear, methods for developing, evaluating, and implementing complex interventions are still under investigation themselves (Craig *et al.*, 2008). Mayo-Wilson, Grant, Hopewell, Macdonald & Montgomery (2013) recently produced a commentary on developing a reporting guideline for social and psychological

intervention trials, however, at the time of writing, there is no accepted consensus on what is considered best practice specifically in relation to trials of non-pharmacological interventions for PSD. However, this review has provided an overview of key issues that need to be considered and it is hoped that such guidance may be developed in the future.

For many years now, the RCT has long been considered the ‘gold standard’ for the collation of evidence to inform treatment and practice guidelines in healthcare. This systematic review, along with guidance from Craig *et al.* (2009) and Mayo-Wilson *et al.* (2013) has highlighted the potential benefits of the use of a range of methodologies, including naturalistic and observational research designs. Hotopf (2002) referred to some of the issues with RCTs and their problems with external validity, and conversely, the problems with observational and naturalistic studies lacking internal validity. Hotopf (2002) discussed how the ‘pragmatic RCT’ aims to bridge the gap between internal and external validity when designing and evaluating treatment algorithms for specific health conditions. The key aim of the pragmatic RCT is to address heterogeneity of patients typically encountered in clinical practice, so these trials tend not to be as preoccupied as traditional RCTs with narrow diagnostic labels and categories, thus, allowing for more naturalistic study conditions. This systematic review provides support for the pragmatic RCT, and highlights how it might be a good compromise for future studies evaluating the efficacy of the non-pharmacological treatment of PSD.

Declaration of Interests

The author conducted this systematic review as part of a portfolio thesis for doctoral training in clinical psychology (DClinPsychol) undertaken at the University of Edinburgh. This training place was jointly funded by NHS Education for Scotland and NHS Lothian health board. No other sponsorship, funding or support were received.

Systematic Review References

- *Alexopoulos, G. S., Wilkins, V. M., Marino, P., Kellanopouluos, D., Reding, M., Kiossis, D. N., *et al.* (2012). Ecosystem focused family therapy in poststroke depression: a preliminary study. *International Journal of Geriatric Society*, 27, 1053-1060.
- American Psychiatric Association (1994). *Diagnostic and Statistical Manual of Mental Disorders*, 4th ed. Washington, DC: American Psychiatric Association.
- American Psychological Association (2009). *Publication manual of the American Psychological Association (6th Ed.)*. Washington, DC: Author.
- Bradbury, C. L., Christensen, B. K., Lau, M. A., Ruttan, L. A., Arundine, A. L., & Green, R. E. (2008). The efficacy of cognitive behaviour therapy in the treatment of emotional distress after acquired brain injury. *Archives of Physical Medicine and Rehabilitation*, 89(12), 61-68.
- British Psychological Society (2011). *The Use of Psychological Tests in Healthcare Research*. The British Psychological Society: Leicester.
- Broomfield, N., Laidlaw, K., Hickabottom, E., Murray, M. F., Pendrey, R., Gillespie, D. C. (2010). Post-stroke depression: The case for augmented, individually tailored cognitive behavioural therapy. *Clinical Psychology and Psychotherapy*, 8(3), 202-217.
- Centre for Reviews and Dissemination (2009). *Systematic reviews: CRD's guidance for undertaking reviews in healthcare*. York: CRD, University of York.
- Chambless, D. L., & Hollen, S. D. (1998). Defining empirically supported therapies. *Journal of Consulting and Clinical Psychology*, 66(1), 7-18.
- Christensen, A., & Jacobson, N. S. (1994). Who (or what) can do psychotherapy: The status and challenge of nonprofessional therapies. *Psychological Science*, 5, 8-14.
- Cohen, J. (1988). *Statistical Power Analysis for the Behavioral Sciences* (2nd ed.). Hillsdale, NJ: Earlbaum.
- Craig, P., Dieppe, P., Macntyre, S., Michie, S., Nazareth, I., & Petticrew, M. (2008). *Developing and evaluating complex interventions: new guidance*. Medical Research Council. doi: <http://dx.doi.org/10.1136/bmj.a1655>

- de Man-van Ginkel, Gooskens, F., Schuumans, M. J., Lindeman, E., Hafsteinsdottir, T. B. (2010). A systematic review of therapeutic interventions for poststroke depression and the role of nurses. *Journal of Clinical Nursing*, *19*, 3274-3290.
- Fraser, C. J., Christensen, H., & Griffiths, K. M. (2005). Effectiveness of treatments for depression in older people. *Medical Journal of Australia*, *182*, 627-632.
- Friedman, L. M., Furberg, C., & DeMets, D. (1998). Multicenter Trials. In: *Fundamentals of Clinical Trials*. New York: Springer-Verlag; 345-356.
- Hackett, M. L., Anderson, C. S., House, A. O., & Halteh, C. (2008). Interventions for preventing depression after stroke. *Cochrane Database of Systematic Reviews*, (2008)3.
- Hackett, M. L., Anderson, C. S., House, C. S., & Xia, J. (2008a). Interventions for treating depression after stroke. *Cochrane Database of Systematic Reviews*, (2008)4.
- Hackett, M. L., Yapa, C., Parag, V., & Anderson, C. S. (2005). Frequency of depression after stroke: A systematic review of observational studies. *Stroke*, *36*, 1330-1340.
- *Hibbard, M., Grober, S. E., Stein, P. N., & Gordon, W. A. (1992). Poststroke depression. In: A. Freeman & F. Dattilio (Eds.), *Comprehensive Casebook of Cognitive Therapy* (pp. 303-310). New York: Plenum Press.
- Hotopf, M. (2002). The pragmatic randomised controlled trial. *Advances in Psychiatric Treatment*, *8*, 326-333.
- IAPT (2011). *Increasing Access to Psychological Therapies Data Handbook: Guidance on recording and monitoring outcomes to support local evidence-based practice*. IAPT: London. Retrieved 2 June 2013 from <http://www.iapt.nhs.uk/silo/files/iapt-data-handbook-v2.pdf>
- Jacobson, N. S., & Christensen, A. (1996). Studying the effectiveness of psychotherapy: How well can clinical trials do the job? *American Psychologist*, *51*, 1030–1039.
- Jacobson, N. S., & Truax, P. (1991). Clinical significance: A statistical approach to defining meaningful change in psychotherapy research. *Journal of Consulting and Clinical Psychology*, *59*(1), 12-19.

- *Joo, H. M., Lee, S. J., Chung, Y. G., & Shin, Y. (2010). Effects of mindfulness based stress reduction program on depression, anxiety and stress in patients with aneurismal subarachnoid hemorrhage. *Journal of the Korean Neurosurgical Society*, 47, 345-351.
- Keppel, G., & Wickens, T. D. (1982). *Design and Analysis: A researcher's handbook*. Prentice Hall: NJ.
- Knapp, P., Young, J., House, A., & Forster, A. (2000). Non-drug strategies to resolve psycho-social difficulties after stroke. *Age and Aging*, 29, 23-30.
- Kneebone, I. I., & Dunmore, E. (2000). Psychological management of post-stroke depression. *British Journal of Clinical Psychology*, 39, 53-65.
- Koder, D-A., Brodaty, H., & Anstey, K. J. (1996). Cognitive therapy for depression in the elderly. *International Journal of Geriatric Psychiatry*, 11, 97-107.
- *Lincoln, N. B., & Flannaghan, T. (2003). Cognitive behavioural therapy for depression following stroke. *Stroke*, 34, 111-115.
- *Lincoln, N. B., Flannaghan, T., Sutcliffe, L., & Rother, L. (1997). Evaluation of cognitive behavioural treatment for depression after stroke: a pilot study. *Clinical Rehabilitation*, 11, 114-122.
- Lincoln, N. B., Kneebone, I. I., Macniven, J. A. B., & Morris, R. C. (2012). *Psychological Management of Stroke*. Chichester: John Wiley & Sons, Ltd.
- Lustman, P. J., Griffith, L. S., Freedland, K. E., Kissel, S. S., & Clouse, R. E. (1998). Cognitive behaviour therapy for depression in type 2 diabetes mellitus: A randomised, controlled trial. *Annals of Internal Medicine*, 129, 613-621.
- Mayo-Wilson, E., Grant, S., Hopewell, S., Macdonald., G., Moher, D., & Montgomery, P. (2013). Developing a reporting guideline for social and psychological intervention trials. *Trials*, 14, 242-257.
- Mehta, S., Pereira, S., Janzen, S., McIntyre, McClure, A., Teasall, R. W. (2012). Effectiveness of psychological interventions in chronic stage of stroke: A systematic review. *Topics in Stroke Rehabilitation*, 19(6), 536-544.
- Mitchell, P. H., Veith, R. C., Becker, K. J., Buzaitis, Cain, K.C, Tirschwell, D., & Teri, L. (2009). Brief psychosocial-behavioural intervention with antidepressant reduces

- poststroke depression significantly more than usual care with antidepressant. *Stroke*, 40, 3073-3078.
- Moher, D., Liberati, A., Tetzlaff, J., & Altman, D. G. (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. *PLoS Medicine*, 6(6). Retrieved 19 January 2013 from <http://www.prisma-statement.org/usage.htm>
- Mohr, D. C., & Goodkin, D. E. (1999). Treatment of depression in multiple sclerosis: review and meta-analysis. *Clinical Psychology: Science and Practice*, 6, 1-9.
- Patel, M. X., Doku, V. & Tennakoon, L. (2003). Challenges in recruitment of research participants. *Advances in Psychiatric Treatment*, 9, 229-238.
plosmedicine.org/article/info%3Adoi%2F10.1371%2Fjournal.pmed.1000097
- *Rasquin, S. M. C., van de Sande, P., Praamstra, A. J., & van Heughten, C. M. (2009). Cognitive-behavioural evaluation for depression after stroke: Five single case studies on effects and feasibility. *Neuropsychological Rehabilitation*, 19(2), 208-222.
- Robinson, R. G. (1997). Neuropsychiatric consequences of stroke. *Annual Review of Medicine*, 48, 217-299.
- SIGN (2008). *SIGN 50: A guideline developer's handbook. Revised Edition, November 2011*. Retrieved 19 February 2013 from <http://www.sign.ac.uk/guidelines/fulltext/50/>
- *Sims, J., Galea, M., Taylor, N., Dodd, K., Jespersen, S., Joubert, L., & Joubert, J. (2009). Regenerate: assessing the feasibility of a strength-training program to enhance the physical and mental health of chronic post stroke patients with depression. *International Journal of Geriatric Psychiatry*, 24, 76-83.
- *Thomas, S. A., Walker, M. F., Macniven, J. A., Haworth, H., & Lincoln, N. B. (2012). Communication and Low Mood (CALM): a randomized controlled trial of behavioural therapy for stroke patients with aphasia. *Clinical Rehabilitation*, 0(0), 1-11.
- *Towle, D., Lincoln, N. B., & Mayfield, L. M. (1989). Evaluation of social work on depression after stroke. *Clinical Rehabilitation*, 3, 89-96.

- van het Hoofd, M. G. V., Roelse, H., Kwakkel, G., Schepers, V. P. M., & van de Port, I. G. L. (2011). Systematic Review: Does exercise therapy affect depression or depressive symptoms after stroke? *Physical Therapy Review*, 16(3) 191-200.
- Wing, J., Cooper, J., & Sartorius, N. (1974). *Measurement and Classification of Psychiatric Symptoms: Instruction Manual for the PSE*. Cambridge University Press: London.

*references marked with asterisks refer to the articles selected for methodological appraisal in the systematic literature review.

CHAPTER THREE: JOURNAL ARTICLE

The conceptualisation of post-stroke emotional lability (PSEL): A qualitative investigation with specialist stroke staff.

This chapter summarises the background, aims, method, results and findings of the research study in the format of a journal article. A discussion of the implications of these findings with regard to clinical practice and future research follows. The author guidelines of the Journal *Rehabilitation Psychology* (APA, 2010) are adopted.

Journal Article Abstract

Objective: To examine the ways in which specialist stroke staff conceptualise and identify post-stroke emotional lability (PSEL) in their clinical practice.

Background: Previous research has identified that PSEL is a prevalent and distressing condition. It has been found to have a significant impact on stroke survivors' rehabilitation and psychological wellbeing, and is known to be a source of distress for families and carers. However, the aetiology of the condition is not well understood, and guidance on the identification, treatment, and management of PSEL is scarce.

Method: Seven interviews were conducted with experienced specialist stroke professionals working clinically in post-acute rehabilitation settings in Scotland. Grounded Theory informed the collection and analysis of data.

Results: This study provides a tentative theoretical model for 'how staff think about PSEL' in relation to their clinical practice. The model captures the way in which specialist experienced staff conceptualise PSEL. The model also captures some of the problems staff encounter with the definition and identification of PSEL, and how they have noticed different patients' responses to it.

Conclusions and implications: Specialist stroke staff conceptualise PSEL as a neurobiological condition. PSEL can be difficult to identify and manage, especially when it is severe and appears in combination with other issues and complexities such as post-stroke depression (PSD), significant behavioural disturbance, anosognosia, and aphasia. Further research on the aetiology of PSEL is required in order to be able to provide appropriate information and support to patients and their families. It is also important to explore training needs for staff in relation to PSEL.

KEYWORDS: *stroke, emotional lability, conceptualisation, Grounded Theory*

Introduction

It has long been known that changes in emotional behaviour can follow brain damage. For example, in 1872, Darwin wrote “...certain brain diseases such as hemiplegia, brain wasting, and senile decay, have a special tendency to induce weeping.” (Allman, Hope & Fairburn, 1990, p.1). These changes in emotional behaviour have been found in a variety of disorders of the central nervous system (House, Dennis, Molyneux, Warlow & Hawton, 1989).

Disturbances of emotional behaviour such as difficulty controlling crying or, less commonly, laughing, have been reported to be common after stroke (House *et al.*, 1989). Poeck (1969) distinguished two main types of disturbed emotionality associated with brain lesions: one he called ‘pathological crying and laughing’ (PCL), and the other emotional lability (EL). Hackett, Yang, Anderson, Horrocks and House (2010) stated that the main differences in Poeck’s (1969) dichotomy were that in PCL, emotional outbursts were provoked by non-emotive or incongruent stimuli, and the response appeared socially inappropriate, whilst in EL, the emotional outburst was more socially familiar and congruent, and provoked by appropriate emotive stimuli (Hackett *et al.*, 2010, p. 2). However, the evidence to support Poeck’s (1969) dichotomy is tenuous (Hackett *et al.*, 2010), and much more emphasis is now placed on the frequency and intensity of emotional behaviour (Allman *et al.*, 1990). Individuals are often classified within ‘subtypes’ of disturbed emotionality, depending on a variety of clinical features, organic disease pathology, and individual differences, and the issue is that patients subjectively have little control over their emotional behaviour (Allman *et al.*, 1990; Hackett *et al.*, 2010).

The aetiology of this problem with control over emotional behaviour remains uncertain, and in most cases cerebral pathology is widespread, bi-lateral, and involves the cortico-bulbar tract (white matter pathway connecting the cerebral cortex to the brainstem),

however it may also result from isolated unilateral lesions (Feinstein, Feinstein, Gray & O'Connor, 1999; Wilson, 1924). There is a statistical association between increased emotional behaviour and left frontal and large stroke lesions (House *et al.*, 1989). However, more recently, Parvizi, Anderson, Martin, Damasio, and Damasio (2001) hypothesized that PSEL is linked with the cerebellum, following more recent knowledge regarding the neurobiology of cognition, emotion and feeling. Parvizi *et al.* (2001) highlighted that although their view on PSEL is based on more recent knowledge, a more in depth understanding of the neurobiology of emotion is required, and only further neurophysiological, neuroanatomical, and neuroimaging studies have the scope to provide this understanding. Miller, Pratt and Schiffer (2011) refer to a number of studies that have found PSEL to be more common in patients with cerebellar damage that has been recognized previously. Miller *et al.* (2011) discuss how the different cell types (e.g. golgi, basket, Lugaro, Purkinje, granulae) and levels (white matter, granular, and molecular) and their functional interactions within the microcircuitry of the cerebellum could perform a gate-control function for the processing of cognitive and emotional information. The neurotransmitters and neuromodulators involved in PSEL pathophysiology could potentially include and that play a role in emotion and its expression, however, serotonin and glutamate appear to be particularly relevant. This is assumed based on their participation in circuits thought to underlie PSEL pathophysiology, as well as their role in the types of pharmacotherapy (serotonergic or antiserotonergic) used to treat PSEL (Wortzel, Oster, Anderson & Archinegas, 2008).

The phenomenon of poor control over emotional behaviour following brain lesions and diseases of the central nervous system has been labelled using a variety of different terms, including 'pseudobulbar affect', 'pseudobulbar palsy', 'emotional incontinence', and 'emotionalism', and the terminology used to describe PSEL is used inconsistently across the literature (Allman *et al.*, 1990). The essential feature of PSEL is an increase in and lack of control of emotional behaviour (Hackett *et al.*, 2010). Disturbance in emotional behaviour following stroke (referred to for the purpose of this study as 'post-

stroke emotional lability', or PSEL) is one of the most common post-stroke behavioural syndromes.

Prevalence figures for PSEL remain somewhat unclear, and although the general consensus is that PSEL affects one in four stroke survivors (House *et al.*, 2008), prevalence rates have been found to vary between 15 and 50 per cent (House *et al.*, 1989; Kim & Choi-Kwon, 2000). PSEL continues to affect one in 10 patients a year after stroke (House *et al.*, 2008).

PSEL has been found to have a particularly significant impact on relationships, activities of daily living, and work participation (Calvert, Knapp & House, 1998), and for patients and carers, it is a potent source of distress (Leiberman & Benson, 1977). It has also been found that PSEL can make a major contribution to participation or social functioning, especially when the episodes interfere with communication or rehabilitation (Leiberman & Benson, 1977).

Individuals who experience PSEL often constrict their social lives because of embarrassment, and unsurprisingly it is not uncommon for sufferers to experience depression and/or anxiety in addition to, and as a result of, their PSEL (Anderson, Vestergaard, Ingeman-Nielsen & Lauritzen, 1995; Calvert *et al.*, 1998; Carota, Berney, Aybeck, Iaria, Staub *et al.*, 2005; Hackett *et al.*, 2010). Professionals have encountered problems in attempting to identify PSEL in clinical practice (Cummings, Arciniegas, Brooks, Herndon, Lauterbach *et al.*, 2006). These problems are mainly due to the association between PSEL and post-stroke depression (PSD), an inconsistent use of terminology when referring to PSEL, and confusion between disorders of emotional expression and clinical mood disorders (Cummings *et al.*, 2006). These issues related to the identification of PSEL may also contribute to an explanation of the discrepancies in prevalence figures.

Gillespie and Cadden (in preparation) further highlighted a number of important issues regarding the conceptualisation and experience of PSEL amongst specialist stroke service staff. Gillespie and Cadden (in preparation) discovered that many members of specialist stroke staff reported that the number of individuals that they saw with PSEL in their day-to-day practice was less than prevalence rates would indicate, and some professionals said that they rarely or never saw it. Others reported not knowing how to manage it when they did see it. Similar to findings by Cummings *et al.* (2006), there also appeared to be some confusion between PSEL and PSD, and staff did not report feeling confident in distinguishing one from the other.

Miller *et al.* (2011) reported that although the understanding of the aetiology of PSEL has been advancing in recent years, it continues to be poorly understood, under-diagnosed, and under-treated by professionals. In addition, guidelines as to its classification and management (Champion, 2006; Intercollegiate Stroke Working Party, 2012) are not directly derived from an evidence base (Hackett *et al.*, 2012; SIGN, 2010). Stroke guidelines (Intercollegiate Stroke Working Party, 2012; SIGN 2010) refer to education, psychological support, and advice for patients with PSEL and their families (SIGN 2010, p. 45). In light of evidence of poor understanding of PSEL and guidelines advocating for education, support and advice, it is imperative to know exactly how specialist professionals conceptualise and understand PSEL, so that services can be sure that patients and families are receiving appropriate and accurate information. Different and conflicting information and advice from staff is likely to result in confusion and anxiety for patients and families.

Study Aims

PSEL has been identified as an area in need of further clarification and exploration in terms of aetiology and presentation, identification, and treatment (Hackett *et al.*, 2010; SIGN, 2010). The association between PSEL and PSD, inconsistent use of terminology, and confusion between disorders of emotional expression and clinical mood disorders, raise questions regarding the relationship between problems with the identification of PSEL and discrepancies in prevalence figures.

Stroke guidelines (Intercollegiate Stroke Working Party, 2012; SIGN, 2010) refer to education, psychological support, and advice for patients with PSEL and their families (SIGN 2010, p. 45). In light of evidence of poor understanding of PSEL (Cummings *et al.*, 2006; Miller *et al.*, 2011) and guidelines advocating for education, support, and advice, it is imperative to know exactly how professionals conceptualise and understand PSEL, so that services can be sure that patients and families are receiving appropriate and accurate information.

This study aims to use a Grounded Theory approach to investigate specialist stroke professionals' experiences of PSEL in their clinical practice, how they conceptualise and understand it, how they identify it in their patients, and how they distinguish it from PSD. This will help gauge the need for further exploration of PSEL as outlined by Hackett *et al.* (2010) and SIGN (2010), as well as possible training and support for staff with regards to knowledge and identification in order to be able to provide appropriate support for patients and their families.

Method

Design

Qualitative research can be a useful tool for learning about a person's experience with a phenomenon, uncovering what lies behind it, and gaining fresh insights into something about which little is known (Strauss & Corbin, 1990). Charmaz's (2006) constructivist Grounded Theory approach was used to explore the conceptualisations of PSEL with experienced stroke specialist professionals in Scotland.

Grounded Theory research begins with an area of study, and discovers the concepts within it, and develops and verifies them through the systematic collection and analysis of data to create a theory (Strauss & Corbin, 1990). Grounded Theory is unique in the way it moves back and forth between gathering data, coding and analysis before returning to gather more data to answer further queries until "saturation" (when no new phenomena are reported in the data) is achieved. Engaging in the Grounded Theory method enables the researcher to retain control of the research process and gives greater analytic power to the results (Charmaz, 2006).

Recruitment

Participants were approached by Specialist Stroke Service Managers via the Lothian Stroke Managed Clinical Network (MCN) in the first instance. The Lothian Stroke MCN is a co-ordinated group of professionals from primary, secondary, and tertiary stroke services working together to ensure quality, equitable, and effective care for patients and families affected by stroke illness. Once participants expressed an interest to managers and agreed to take part, the researcher contacted them directly to arrange interviews. Interviews took place during participants' working hours, and this was negotiated with senior staff and managers.

Eligibility

Those eligible to participate were experienced qualified clinical rehabilitation staff working in post-acute specialist stroke services in Scotland. Professionals were recruited from the medical, occupational therapy, speech and language therapy, physiotherapy, nursing, and clinical psychology disciplines. Participants were required to have a minimum of two years experience of working in specialist stroke services.

Participants

A total of seven participants were recruited from specialist stroke services across Edinburgh (n= 5) and Glasgow (n= 2). Participants were individuals from the main professions that comprise the typical community stroke rehabilitation team, namely: nursing (n=2), medicine (n=1), speech and language therapy (n=1), physiotherapy (n=1), occupational therapy (n=1), and psychology (n=1). See Table 4 for a breakdown of participant information.

Table 4: Participant information

Participant No.	Profession	Gender	Age	Years of stroke experience	Approximate no. of patients seen per month with PSEL
1	Consultant Physiotherapist	M	53	25	<1
2	Community Stroke Nurse	M	35	9	10
3	Community Stroke Nurse	F	38	6	5
4	Specialist Speech and Language Therapist	F	36	7	1-2
5	Specialist Occupational Therapist	F	50	15	2-3
6	Professor of Clinical Pharmacology	M	41	18	8-10
7	Clinical Psychologist	F	29	2	16

In Grounded Theory, the researcher ceases data collection when it has been deemed that categories have been “saturated”. “Saturation” has been achieved when collecting new data no longer creates fresh ideas or theoretical insights, or adds anything new to core theoretical categories (Charmaz, 2006). In this research, “saturation” of categories was

reached after the sixth interview. The seventh interview assisted with consolidation of existing categories, but did not add anything new to the dataset.

Data Collection

Single session individual interviews were conducted and recorded. Interviews ranged in length from 52 - 72 minutes (mean 62 minutes). The interview questions were open-ended and non-judgemental, and designed to gather rich data as well as striving to avoid imposing pre-conceived ideas onto the participant (Charmaz, 2006). Interviews were transcribed verbatim, analysed, and new questions formulated before the next interview.

Memoing is a technique used in Grounded Theory to help the researcher shape the results and monitor and reflect upon their own thoughts, feelings and ideas about the research process, interviews, and resulting analysis. Memos help the researcher stay closely involved with the data, prompting them to scrutinise data, codes, and emerging categories constantly throughout the research process from the early stages, capturing the researcher's thoughts, and comparisons and connections made (Charmaz, 2006). Memos also help highlight gaps in the analysis, and assist with the formulation of codes and raising focussed codes to theoretical categories.

Analysis

Transcripts were coded using initial line-by-line coding, followed by focussed and theoretical coding. Three transcripts were also given to a colleague to code, and ideas about codes and emerging theoretical categories were compared in order to reduce researcher bias.

For validity purposes, findings were also clarified and discussed with two participants, and their feedback was assimilated into the results. This is a strategy commonly used in qualitative research to strengthen reliability and validity of results (Willig, 2008).

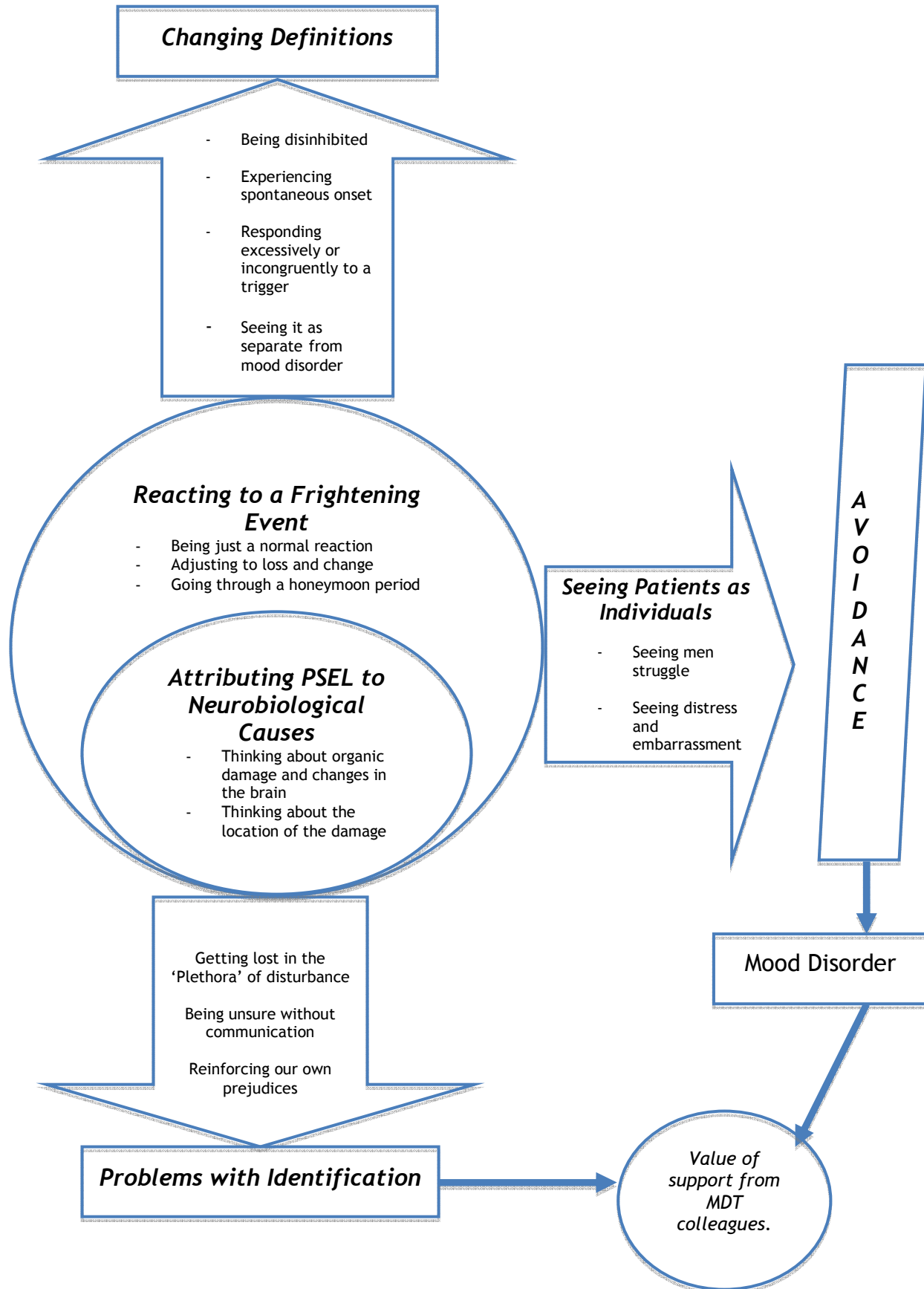
Please see the Chapter four for a more detailed account of the methodology.

Results

This study proposes a model of how experienced specialist stroke professionals think about and experience PSEL in their clinical work (figure 3). Participants discussed their attributions of PSEL, as well as thoughts about seeing patients reacting to a traumatic and frightening event and the impact this has on the maintenance of PSEL. The model also proposes professionals' thoughts about the definition of PSEL, their experiences of problems with the identification of it, and how PSEL affects patients differently depending on gender, roles, beliefs, and values.

The model also refers to the distress that professionals observe patients experiencing as a result of PSEL, and how this is often what individuals require more intensive and specialist input for from the multi-disciplinary team (MDT).

Figure 2: How staff think about PSEL (Diagrammatic representation)



How Staff think about PSEL

The overall theoretical category was ‘How Staff think about PSEL’. This category captured the way in which the participants conceptualised and understood PSEL and its causes, in relation to their clinical work. Five subcategories identified were ‘Attributing it to Neurobiological Factors’, ‘Reacting to a Frightening Event’, ‘Changing Definitions’, ‘Experiencing Problems with Identification’, and ‘Seeing Patients as Individuals’ (figure 2).

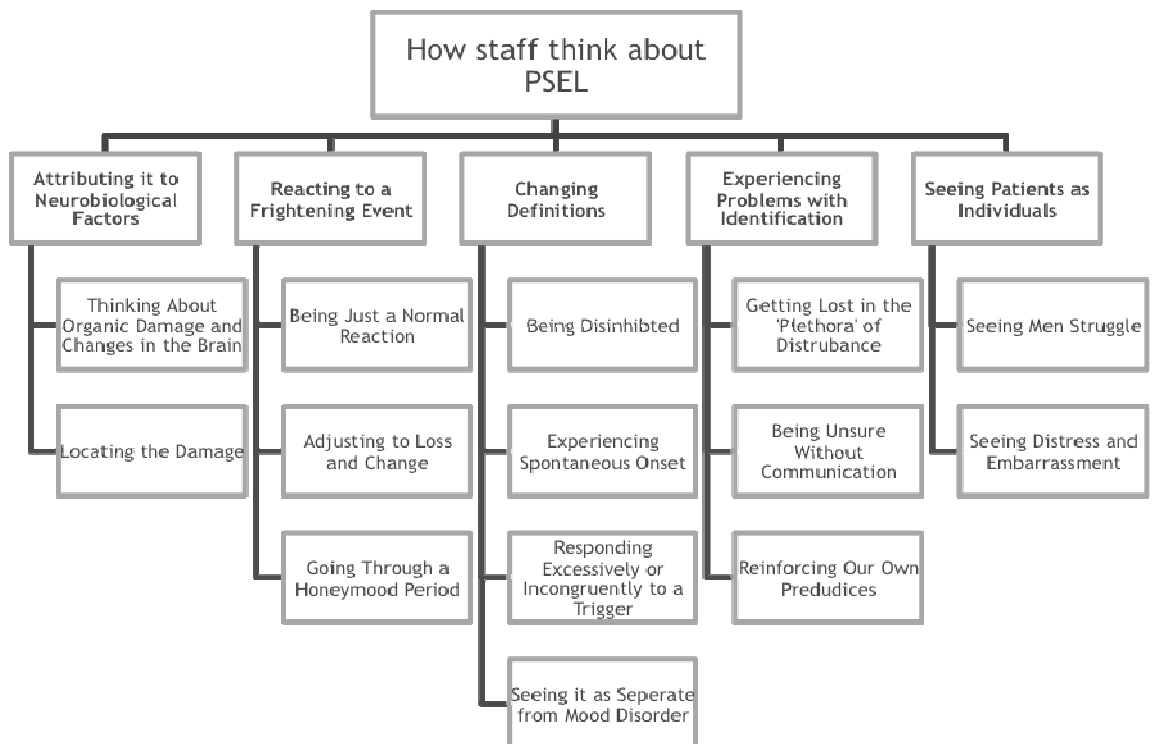


Figure 3: Overview of conceptual categories

Subcategory 1: Attributing it to Neurobiological Factors

All seven participants attributed the cause of PSEL to various changes to an individual's neurobiology after stroke. Participants referred to organic changes and damage to tissues in the brain as a result of the stroke. Some participants were specific in their thoughts about damage and changes in the brain, and some referred to stroke lesion location and how they thought this might contribute to the aetiology of PSEL.

Thinking About Organic Damage and Changes in the Brain

Six of the participants attributed PSEL to organic cortical damage and changes in the brain.

if there has been a disruption to the blood supply to the brain temporarily... then it wouldn't surprise me that your emotions, that your brain controls, will be in some way affected (Participant 3)

Participants' attributions of emotional changes in neuropathology are similar to the early hypothesis of Wilson's (1924) theory on the neural correlates of pathological laughing and crying. Wilson's hypothesis remains the most influential today, although it is not supported by definitive evidence. Wilson (1924) hypothesised that PSEL occurs as a result of disconnect between the motor and limbic systems due to damage to the corticobulbar motor tracts. Similarly more recently, Miller *et al.* (2011) and Parvizi *et al.* (2001) hypothesised about the role of the disconnection of the cortico-pontine cerebellar pathways in PSEL.

Not all participants made explicit reference to organic brain damage, but they talked about the impact of the stroke on the tissues and activity in the brain. One participant discussed the role of neurotransmission, particularly serotonin (5-HT), in relation to causality and neurobiology.

I guess related to the aetiology and pathogenesis of post stroke emotionalism, I guess, the treatment we use, it is likely that there is

something related to neurotransmission, particularly 5-HT as a, as a kind of neurobiological basis of it (Participant 6)

This is congruent with the hypothesis regarding serotonergic neurotransmission (Wortzel *et al.* 2008) and the involvement of the raphe nucleus (the main site of serotonin synthesis), and supported by selective serotonin reuptake inhibitor (SSRI) studies (Derex, Oswestry & Nighoghossian, 1997; Feldman, Meyer & Qunezer, 1997).

Locating the Damage

Three participants discussed the location of damage in the brain, and how they thought this could play a role in the causality of PSEL. Specifically, they considered the link between organic damage and changes to centres in the brain responsible for emotional functioning, and this was tentatively discussed. This is similar to Feinstein *et al.* (1999), and Wilson (1929).

I guess changes in the brain, for one, one cause, emotional centres being dysregulated...depending on where the damage is" (Participant 7)

Five participants frequently referred to damage to the frontal cortex. Again, this would be consistent with Feinstein *et al.* (1999) and Wilson (1924).

...potentially frontal lobe has been affected and that erm, somebody's behaviour is disinhibited to the extent that they can't control their emotions and therefore the neurology, if you like behind that is absolutely key here (Participant 5)

Participant six made reference to the most severe cases of PSEL being related to frontal lobe infarcts.

...from an anatomical perspective, so, from the ones I've seen and the bad ones I've seen, generally have had frontal lobe infarction...the really severe cases are almost always frontal lobe injuries, bilateral frontal injury (Participant 6)

One participant made reference to the role of the tracts and white matter pathways in PSEL. The role of the cortico-spinal tracts in PSEL is strongly supported by a number of

case reports (Mouton, Remy, & Cambon, 1994; Tei & Sakamoto, 1997) where PSEL was significantly associated with basis pontis stroke (the basilar part of the pons, located in the brain stem).

I think anything that's in the tracts, sort of the midbrain area kind of, the amygdala and the basal ganglia, the connections between the two, so white matter pathways, I imagine that damage to all those things can cause issues (Participant 7)

In further support of this, a correlation has been found between PSEL and lenticulocapsular lesions (Kim, 2002). The lenticular nucleus is well known for its role in conveying sensory information and influencing motor activity of cranial nerves (Giroud, Lemesie, Madinier, Billiar & Dumas, 1997).

Three participants referred to the role of the frontal lobes in executive functioning, and the relationship between frontal lobe damage, executive functioning, and PSEL. Similarly, House *et al.* (1989) proposed that frontal lesions may result in disinhibition of emotion control in the same way that frontal lobe damage produces disinhibition of other social behaviour.

I'd wonder in terms of self-monitoring as well potentially, again its frontal, its and executive, executive abilities, in being able to sort of regulate yourself (Participant 7)

Three of the participants highlighted that they typically see PSEL in combination with other sorts of impairments and behavioural disturbances associated with frontal brain regions suggestive of issues with regulation of other behaviours. It is of course well established that the frontal lobes play a central role in the governing of behaviour; Champion (2006) refers to the frontal lobes as the parts of the brain that 'put the brakes on' (p. 108).

I can also think of individuals who have quite extensive frontal lobe involvement, so they present with lots of executive dysfunction, and perhaps are emotionally labile as well so perhaps that erm, that whole, inhibition (Participant 5)

Kim, Choi, Kwon and Seo (2002) found relationships between frontal lesions, disinhibition, and inability to control anger and aggression (ICAA). Interestingly, they also found that ICAA was closely related to dysarthria, motor dysfunction, PSEL, and lesions affecting frontal, lenticulocapsular pontine base areas.

we've definitely seen people who've got the very typical Phineas Gage type, typical problems with bilateral frontal, you know, I've had some very memorable patients who've exhibited extreme forms of that sort of behaviour, really distressing things actually... very bizarre behaviour, as I recall [one patient] was trying to have sex with the washing machine... and he was sexually disinhibited to an extreme degree...he was also emotionally labile, aggressive, and had the full degradation (Participant 6)

Not only did participants make attributions about organic damage and changes in the brain, they also hypothesised about the location of the damage. However, they did not draw on specific models to illustrate their thinking, so it is possible that they have a conceptual understanding of PSEL that comes from wider knowledge of behaviour, emotion, and the brain. Further to this, participants were tentative in the expression of their ideas about the aetiology of PSEL, and this could be reflective of the uncertainty and confusion that continues to surround PSEL. Participants may also be making educated assumptions about the aetiology of PSEL based on their experiences of working with individuals with neurological damage and the common issues they see clinically, i.e. that individuals with damage to frontal regions of the brain often experience problems with disinhibition and behavioural control.

Subcategory 2: Reacting to a Frightening Event

Although participants attributed the cause of PSEL to neurobiological changes, they also associated the condition with a psychological reaction to a sudden, traumatic, and frightening event. This subcategory relates to how participants have reflected on their experiences as clinicians working with patients who have encountered a frightening and traumatic event. They were thoughtful about how the experience of having a stroke must

have felt for their patients, and imagined how they might feel under the same circumstances.

This way of thinking draws parallels with the literature on PSEL as a manifestation of problems with the control of re-experiencing of 'emotionally charged mental events' in the context of having encountered a life-threatening experience (Calvert *et al.*, 1998). This is similar to the way in which individuals can experience symptoms (such as the reliving of memories) of post-traumatic stress disorder (PTSD) following a traumatic event. Although Calvert *et al.* (1998) distinguish clearly between PTSD and PSEL (and reiterate that they are inherently different in aetiology and presentation) they advocate that PSEL should not be thought of as a 'psychologically meaningless accompaniment of brain injury' (p. 2). Lincoln, Kneebone, Macniven, and Morris (2012) more recently highlighted the need for further exploration of the psychological components of PSEL, and they too outlined the contribution of PSEL to distress and resulting problems with low mood and anxiety.

Being Just a Normal Reaction

Five participants conceptualised PSEL as a 'normal' reaction to the experience of having had an unexpected, frightening experience of threat to life.

its just like an emotional response to more of the sort trauma, or the, I guess sort of the getting to terms with what's kind of happened to them (Participant 2)

for many people I'm sure is a pretty normal reaction to you know, you've had a stroke, you feel a bit down, upset, confused, and, you know, then you're kind of knocked off in a very kind of global sense...it's a sort of normal reactive process (Participant 6)

Participants reflected on and empathised with patients' experiences, feelings of fear and confusion in relation to their situation, and how these might result in an increase in emotional behaviour. Other research has demonstrated that individuals with PSEL experience higher levels of distress than those without (Calvert *et al.*, 1998; House *et*

al., 1989), so one might expect a increase in emotional expression due to a higher intensity of distress.

you're in hospital and you're in a sudden, you know, change of scenery, you've no idea maybe why you're there or what's happened to you, what's caused it, you're gonna be frightened, you're gonna be scared, a bit all over the place, not sure how long you're gonna be there for, or if another ones gonna happen again so...its no wonder that someone cries unexpectedly (Participant 7)

Adjusting to Loss and Change

Four participants referred to PSEL in relation to the process of adjusting to loss and change following stroke. This is similar to the literature on how individuals experience distress and emotional disturbance in association with adjustment after stroke (e.g. Lincoln *et al.*, 2012).

I see it as like an emotional response to a trigger, really something, like I said the stroke is kind of, you know they haven't had that kind of mental time to prepare for the sort of the news (Participant 2)

I guess adjustment issues as well, erm, it's a change, erm, also can be quite traumatic for people as well, in terms of what's happened with them (Participant 7)

Two participants also talked about PSEL in the context of change and loss, and associated cognitive appraisal of a new situation. This resonates with reports of heightened levels of distress experienced by stroke survivors (Calvert *et al.*, 1998; House *et al.*, 1989).

they've lost their role, and kind of...I suppose it's the decision making, in the relationships kind of thing where as I think people, you know they feel quite worthless and things...it's a response to something that's kind of happened, and its uncontrollable (Participant 2)

I wonder whether it might be more because of the circumstances that they are in, you know just being more reactive to the change and loss you know that they've encountered (Participant 7)

Going Through a Honeymoon Period

It is known that discharge from hospital to home can be a happy but difficult time for patients, particularly following treatment for life-threatening illness, and when patients have resulting disabilities (Perrins, King & Collins, 1998). Four participants mentioned the 'honeymoon period', where a patient experiences excitement and positive feelings about returning home. They prepare themselves for being reunited with their families and becoming reconnected with their lives again, and they often have an expectation that things will be fine once they are home.

...rehab goals [in hospital] have been about preparing for discharge and immediate kind of self care issues in the home, so however, if they're lucky they've maybe had a spell in the rehab hospital, for a couple of months...then they're back home, and there's this erm, period, honeymoon period I call it, where everything is great (Participant 5)

However, participants illustrated how returning home forces the individual with stroke to come face-to-face with loss and change, from which the hospital environment has protected them. Four participants commented on how discharge is often a crucial time for patients in terms of emotional and psychological adjustment. Patients can find controlling their emotions more difficult at these times due to being exposed to life outside of hospital, and a higher intensity and frequency of emotional stimuli, so PSEL might start to become more noticeable following discharge.

you know, when you are in hospital...a lot of the real emotional issues that going home throws up you might not necessarily have to deal with in hospital...whereas when you go home, you are back in the midst of your family, there is no escape in a sense from those emotional interfaces that go on. I think for the person, I guess that's the time...when people erm, remember how things were (Participant 1)

PSEL is known to present early (within the week following stroke), with symptoms having resolved between three and six months in 90 per cent of cases (House *et al.*, 2008).

Although the participants' narratives offer some explanations as to why PSEL symptoms seem more obvious following discharge, this finding raises questions regarding how the condition is conceptualised, defined, and identified in acute settings. Gillespie and

Cadden (in preparation) found that stroke specialist acute care staff did not always recognise PSEL clinically. It might be that emotional outbursts are not considered pathological in hospital, but perhaps more abnormal or pathological in the community, when the stroke survivor's role changes, and they are no longer a 'patient'. In support of this, one participant noted that PSEL might be more noticeable following discharge, as the focus at home is different to that in hospital i.e. the preoccupation with hospital routine and intensive functional rehabilitation is not present at home.

within more acute settings...its one of those things that maybe in some people doesn't even present until discharge anyway and for some people its one of things that is really low down on the list of priorities to manage cause people might have like a, a swallowing problem, so eating is an issue, they are needing to learn to eat again, walk again, move again, talk again, so emotional lability is way down on the priority list for people [in the acute phase] (Participant 6)

Subcategory 3: Changing Definitions

PSEL has been defined in many different ways over the years, and the terminology associated with naming the condition has been unclear and confusing (Miller *et al.*, 2011). However, within the present study all participants defined PSEL in a similar way. They said that it was defined by a lack of control or disinhibition, and the outbursts were either incongruent with, or an excessive response to a trigger. This is similar to the definition used by Miller *et al.* (2011), who define PSEL as an emotional expression disorder, characterised by spontaneous and uncontrollable outbursts of crying or laughing that occur in response to an inappropriate environmental trigger and may not be reflective of the underlying emotional state. All participants also differentiated PSEL from disorders of mood, but reported that it can be linked to PSD and anxiety due to avoidance of social situations, distress, and negative cognitions.

Being Disinhibited

Participants frequently referred to the 'lack of control' or 'disinhibition' of emotional behaviour following stroke. Cummings *et al.* (2006) refer to 'emotional disinhibition

syndrome', and use the term 'involuntary emotional expression disorder'. Participants highlighted this lack of control/disinhibition of emotional behaviour as the most important point in relation to the definition of PSEL.

well in the first instance, one tends to think of a disinhibition of normal emotional responses or generation of abnormal emotional responses (Participant 1)

most importantly more fluent expression of emotion, erm and, in my experience, that's been both about tears, uncontrolled emotional expression in a negative way, but also in a positive way so, sort of unexplained giggling, that sort of thing (Participant 4)

Experiencing Spontaneous Onset

Five participants also made reference to the onset of the episodes and that they were often sudden or 'spontaneous', taking patients and those around them by surprise.

they seem fine...and then all of a sudden, bang something kind of happens and there doesn't seem to be a kind of particularly emotional trigger, it could be anything really, you know its just that kind of spontaneous kind of thing really that happens (Participant 2)

One participant also discussed a return to a normal, or 'pre-outburst' level.

it seems excessive and it doesn't appear to have a real erm, so somebody potentially is having an emotional outburst whether it's tears or a euphoria, which then returns to some pre, pre-outburst state (Participant 4)

This is similar to how PSEL is often described in the literature as 'sudden outbursts' of emotional expression that are not necessarily indicative of emotional distress (Miller *et al.*, 2011).

Responding Excessively or Incongruently to a Trigger

PSEL can occur without a clear relationship to a stimulus, and patients do not necessarily experience any congruent change in feeling (Tang, Chen, Lu, Mok, Xiang *et al.*, 2009).

Participants referred to PSEL as an incongruent emotional reaction to a benign trigger or stimulus.

[PSEL is] inappropriate emotional expression, be that laughter or crying, more often in my experience the latter in response to what would have previously been regarded as benign or neutral stimuli, so you could just say hello to someone, and they burst into tears or start laughing (Participant 6)

They also referred to instances where some degree of emotion or feeling might be expected, but the expression of the emotion is excessive.

they might be out in the street and somebody asks them you know...how are they and that just triggers this kind of overly emotional...response (Participant 2)

These descriptions are similar to Poeck's dichotomy (Poeck, 1969). This distinguishes between two main types of PSEL. Pathological crying and laughing (PCL) is provoked by non-emotive or incongruent stimuli, and emotional lability (EL), where uncontrollable crying or laughter seems related to emotionally congruent stimuli and to correspondent emotional internal feelings, but is excessive in degree.

Although participants discussed PSEL in these two different ways interchangeably in their narratives, no participant made this distinction clearly. This might again be related to a conceptual understanding of PSEL based on clinical experience, and knowledge of behaviour, emotion, and the brain as opposed to an understanding that is derived directly from a theoretical model.

Seeing it as Separate from Mood Disorder

All participants were generally clear that the expression of the emotion is often not reflective of the patient's true emotional experience.

its a bit like phantom limb pain, almost like phantom emotion...that is a really interesting way of describing it, can you tell me more?...because in phantom limb pain, you might not have the limb...so there is no damage to tissues but...you experience pain. And I guess the emotion is...in tact, pre-stroke and then the effect

of the stroke is...an inappropriate level of emotion, so I just say phantom emotion in the sense that it may not be really what that person is feeling (Participant 1)

they just can be watching something, on TV and all of a sudden the emotion just comes over them and they just start crying, they're not necessarily feeling sad, they just cry (Participant 3)

Although participants referred to psychological factors and distress associated with PSEL, they were clear that PSEL is a disorder of emotional expression, and a separate phenomenon from a mood disorder like PSD.

when they [the patient] appeared to be on a chronic basis being more withdrawn and erm, expressing...thoughts of unhappiness or...crying which was maybe I guess more of a sustained, and almost seemingly appropriate kind of emotional response, I would be thinking then that there was perhaps erm, a depression going on as opposed to a fluctuating emotional lability (Participant 1)

Participants did refer to times where this distinction might be unclear (which is explored in the next subcategory), and this is related to discussions in the literature regarding the common confusion between mood, affect, and emotional behaviour (Cummings *et al.*, 2006). Six participants said that they did not see themselves as 'experts' in emotional problems after stroke, and that that they would seek support from multi-disciplinary colleagues if they felt unsure, particularly if they were concerned about a mood problem. This finding is important in terms of the model of how staff conceptualise PSEL, and it demonstrates how MDT colleagues draw upon knowledge and support from one another.

I would discuss it with the team, I mean I wouldn't, I wouldn't er, profess to be able to make that decision singlehandedly (Participant 1)

The first five participants (non-psychology/medical professionals) said that they would be inclined to seek support particularly from either psychology or medical colleagues, and that they saw professionals from those disciplines as the 'experts' in PSEL.

...occasionally we have people who are so low in mood, you know that we would... engage other people within the team as appropriate, whether it's the GP, and ask them to screen this person to speak to them about everything from medication to

maybe talking therapies, and then we've got the psychologists as well and we would be thinking about a referral to a neuropsychologist, to get a more expert opinion (Participant 5).

It was clear that Participant 6 and Participant 7 (medic and psychologist, respectively) had further in depth knowledge of both PSEL and PSD, however, neither participant 6 or 7 viewed themselves as experts in the area either. If highly specialist and experienced staff do not see themselves as experts in the area, less experienced and non-specialist staff working with stroke patients are unlikely to feel knowledgeable or confident about PSEL. This is again likely to be reflective of the uncertainty and confusion that surrounds PSEL (Miller *et al.*, 2011). One participant highlighted this as a particular issue that she had observed.

I have observed some staff who, maybe more junior staff, qualified staff who struggle with it because, I suppose they don't have an understanding that it may be a symptom of emotionalism, or that they've not been trained enough perhaps to deal with someone who maybe has low mood either (Participant 5)

Subcategory 4: Experiencing Problems with Identification

Participants consistently referred to some of the difficulties they can have with the identification of PSEL in practice.

Getting Lost in the 'Plethora' of Disturbance

All participants referred to the way PSEL can often present in combination with a 'plethora' of other sometimes quite severe disturbance after stroke.

its like a graphic equalizer, adjusting, that kind of like a music system, there's a whole load of different things going on at the same time, it may be that, erm, inappropriate crying for example or inappropriate laughing, there may be peaks that make you take notice of that particular presenting feature more in the context of everything else (Participant 1)

When PSEL appears alongside other issues, staff can often experience difficulties with classifying and identifying it. It can be particularly unclear when issues associated with insight (anosognosia), communication (aphasia), and severe behavioural disturbance are

present. These sorts of disorders can often act as barriers to gathering information from the patient themselves (Cummings *et al.*, 2006), as well as affecting the individual's ability to participate in therapy. Further to this, when PSEL appears alongside other more disturbing or challenging issues such as anosognosia, aphasia, or severe behavioural disturbance, it does not usually take priority in terms of management (Cummings *et al.*, 2006; Prigatano, 2010).

I think the people who have bilateral frontal lobe injury present with this plethora of you know, profoundly disturbed different behaviour, have emotional lability too, but it's at the bottom of a long list of problems to deal with (Participant 6)

Two participants discussed how when PSEL presents with anosognosia (a deficit of self-awareness due to brain damage), it can be even more difficult to identify, and consequently treat.

basically its impossible [to treat]... if they aren't able to acknowledge that there is a problem, and...they are emotionally labile too, it's a bad combination (Participant 6)

This finding highlights the some of the complexities encountered when working with individuals with brain damage. In individuals with anosognosia, it is difficult to orientate the individual to the reality of their situation. Identification of PSEL often relies on the patient's report of the reality of their experience of emotion, which is a necessity for the identification of PSEL in that the patient needs to be able to express and communicate their true feelings in some way. If individuals have issues with insight, they may not give an accurate report of how they feel (Prigatano, 2010).

if you're presenting with anosagnosia, you might not actually have any insight into the fact that you are affected at all, so it might just be a big old laugh, erm, and then it's difficult to focus people on the reality of their situation (Participant 1)

Being Unsure Without Communication

Participants also discussed feeling unsure about when they are seeing PSEL in patients with communication problems.

when things become complex, and...confusing, or when we are not sure as professionals whether we are seeing emotional lability or whether we're not, and that's why I asked about aphasic patients particularly people who are receptively struggling as well, because...everything gets lost in translation with speech problems, so I guess there are times when maybe the tears may be due to frustration in association with the communication problems or whether it really is emotional lability, it really is difficult to know (Participant 5)

One participant illustrated how assessing for emotional problems in patients with aphasia is inherently difficult. In relation to this, there are currently no standardised screening measures available to assess for PSEL in patients with aphasia.

...the Broca's aphasia patients...are very emotional. Whether that's pathological or whether that's just the kind of chronic frustration of being unable to express themselves, its hard to tell... again its hard to precisely to nail down but you know you see folk with Broca's aphasia who are really tearful (Participant 6)

This is similar to what the literature says about the experience of aphasic patients, and how they are often very emotional and tearful in relation to the frustration they experience as a result of being unable to communicate effectively (Raffii & Hillis, 2006). Goldstein (1939) coined the term 'catastrophic reaction', which is characterised by an extreme emotional reaction expressed by a patient when they are confronted with things that they are no longer able to do as a result of disabilities due to their illness. This phenomenon might appear quite similar to PSEL in practice; patients may not be able to communicate what is going on for them, which often makes it difficult to judge whether tearfulness in aphasic patients is PSEL or simply an expression of the patient's frustration.

Reinforcing Our Own Prejudices

Five of the seven participants reported seeing smaller numbers of individuals with PSEL in practice than the numbers reflected in the prevalence figures. It was suggested by participant 6 that this might be due to professionals having certain expectations to see the disorder in certain cases i.e. the literature on PSEL states that it is most common in those with frontal lesions, so it might be that professionals 'go hunting' for it patients with this profile.

this is complex because whether its that we look for it more in patients where we expect it may be present, whether to an extent we are sort of erm, you know we go hunting for it in certain sorts of patients and therefore we are just reinforcing our own prejudices (Participant 6)

Three participants also talked about how PSEL in its crying form as it is generally more likely to be detected as it is more often associated with distress. Social learning theory demonstrates how we learn from a young age to respond and behave in specific ways in certain social situations (Bandura, 1977). Expression of emotion is a form of non-verbal communication (Axtell, 1991). As human beings, when we see someone crying, it evokes feelings of sympathy and concern, and implies that the individual exhibiting that behaviour is experiencing pain or emotional distress. It is also unusual to see crying in social situations. In the same way a parent attends to a crying baby, we would perhaps attempt to discover what is causing the individual's distress and offer them support. Social learning theory also encourages us to respond to laughter in a certain way, i.e. usually with reciprocal humour. Participants thought that PSEL in its laughing form might appear a little unusual at times, but we might not automatically think that the person is experiencing PSEL as laughing is a more socially common and acceptable behaviour than crying.

Again, its much easier to pick out, if someone is weeping and crying and obviously distressed, that is, a big alarm...but do you know if somebody's happy and laughing, I'm not convinced that it would be picked up...[because laughing is] much more socially acceptable than it is to cry, so...that person might not get referred to the [relevant] professionals, so it does worry me a little bit, we're not very good at dealing with that (Participant 5).

Subcategory 5: Seeing Patients as Individuals

All participants referred to some extent to the individual differences they observe in their patients, and how these differences contribute to the patients' experience of PSEL, and whether they encounter significant distress as a result of the condition, or not. Participants seemed to be saying that thoughts, beliefs, and values play a significant role in whether a person does or does not experience distress in response to PSEL.

Seeing Men Struggle

Although PSEL is no more common in males than females (Kim, 2002; Morris, Robinson & Raphael, 1993), participants reported that clinically, they had observed males struggling more with PSEL.

men of a certain age, or era will think you know 'I must not show any weakness' and therefore...suddenly crying and you know not having any control over that must feel horrendous for them"(Participant 1)

This finding shares similarities with the concept and theory of masculinity. Masculinity is socially, historically and culturally shaped (Beynon, 2002). It embodies all of the things that are traditionally perceived as what being a man is about. Masculinity traditionally and stereotypically implies strength, bravery, and physical strength (Shamir & Travis, 2002), and it is weaved tightly into human experience in many cultures. From a young age, children learn about the concept of masculinity through their experiences of social interactions with others, and therefore it plays an integral role in the development of values, beliefs, and identity in young males (Beynon, 2002). Shamir and Travis (2002) illustrate how historical stereotypes associated with the role of masculinity can cause men to experience shame and embarrassment in relation to expression of emotion and tearfulness as they can often be seen as a sign of weakness or inferiority.

The tough guys are always erm interesting because I kind of think its just not something that they are used to, to kind of seeing really and things so I think they kind of find it quite a struggle to kind of accept it as well (Participant 2)

I think they've often been sittin worrying about it, especially men, I find...you know strong grown men are sittin watchin something on TV and all of a sudden they're cryin, I think they really struggle with that...they don't really want to bring it up...the kind of shame and embarrassment of it (Participant 3)

In light of this, it is not surprising that men particularly struggle with PSEL, as a change in expression of emotion is likely to challenge a man's ideas about his identity, especially if tearfulness was rare for him before the stroke (Beynon, 2002; Shamir & Travis, 2002).

Seeing Distress and Embarrassment

Similarly, participants also noted witnessing the distress and embarrassment that their patients often experience as a result of PSEL more generally due to their cognitions, personal beliefs and values, and the impact that this distress can then have on their mood and engagement in rehabilitation and social activities.

it depends on the persons own...belief systems as well...what do they think if they're crying all the time, have they got...failure related thoughts, hopelessness about the future and those sorts of things and the fact that they're now losing control does...that then perpetuate...a depression then evolving over time (Participant 7)

This finding draws parallels with the 'rules' that govern the display of emotion (Axtell, 1991), and perhaps the level of importance that is attached to these rules for an individual (values). If an individual has a value or belief system where frequent and uncontrollable displays of tearfulness are not acceptable or do not fit (in the same way that masculinity related beliefs and values in men do not fit with PSEL), this is likely to cause that individual to experience negative intrusive cognitions and associated distress (Kasprisin, 2004).

In support of this theory, Eccles, House and Knapp (1999) found that individuals with PSEL had more negative and intrusive thoughts, and utilised more avoidant coping strategies than those without PSEL. Five participants referred to how they observe patients with PSEL avoiding social situations due to distress and embarrassment.

Emotionalism...can be really quite disabling for some people, people often avoid social contact, they will avoid going out and engaging in a whole array of activities because they are too embarrassed, and they know they wont be able to control their emotions (Participant 5)

It is well known in the cognitive-behavioural paradigm that avoidance of activity can act as a maintenance factor for both depression and anxiety. Avoidance can also contribute to depression and low mood due to a decrease in engagement in pleasurable and satisfying activities (Beck, 1979). This finding illustrates how individuals are more likely to experience anxiety and depression as a secondary consequence of PSEL.

Discussion

This study provides a theoretical model of how specialist stroke professionals conceptualise PSEL. Participants tentatively identified the neurobiological causes of PSEL, and none of them professed to feel confident about their knowledge of these causes. Participants' thoughts about the causes of PSEL appeared to reflect their clinical experience, and perhaps a more general conceptual knowledge and understanding of behaviour, emotion, and the brain, as opposed to knowledge and ideas being based on specific theoretical models. Participants reflected on the importance of support from neuropsychology and medical colleagues in the identification and management of PSEL. Participants also discussed the role of psychological and cognitive factors such as associated distress due to individual's beliefs and values, and problems with adjustment to change and loss in the maintenance and exacerbation of PSEL.

The findings of this study also highlight some incongruences regarding what being an 'experienced and specialist' stroke professional actually means, how 'experienced and specialist' professionals see themselves, and how they feel about the clinical work they do with patients, especially when they encounter issues about which little is known, like PSEL. Stroke guidelines and recommendations (e.g. Intercollegiate Stroke Working Party, 2012, SIGN, 2010) refer to the value of experienced and specialist staff, however, it is important to consider that although professionals may indeed be considered experienced and specialist, this does not mean to say that they do not continue to find certain particularly complex aspects of their clinical work, such as PSEL, difficult and confusing at times. The findings of this study demonstrates how ambivalence about diagnosis, aetiology, and treatment, is likely to be common for many health professionals working with a condition about which little is known, regardless of specialist training and experience.

The findings of this study illustrate how professionals experience problems with identifying PSEL. These problems appear to be caused by the inconsistent use of terminology, the confusion and relationship between PSEL and PSD (Cummings *et al.*, 2006; Miller *et al.*, 2011), and the ‘plethora’ of issues that can be experienced by stroke survivors, especially when sufferers have problems with insight and communication. These findings regarding problems with identification of PSEL may contribute to an explanation for the wide ranges observed in prevalence figures (House *et al.*, 1989; Kim & Choi-Kwon, 2000), and this idea is worthy of further exploration.

Implications of Findings

There do not appear to have been other studies to date examining how staff conceptualise PSEL. This study has demonstrated that highly specialist and experienced professionals have a tentative understanding of the condition. However, some of these professionals reported that they do not feel confident in their knowledge of the causes of PSEL, neither are they confident in how to identify it and treat it in clinical practice. Participants reported that they would seek advice or support from neuropsychology and medical colleagues when they felt unsure, particularly if they had concerns about low mood. These findings emphasise the importance for multi-disciplinary working in stroke.

Clinical guidelines advise that patients with PSEL should be ‘appropriately distracted from the provoking stimuli’ (Champion, 2006; Intercollegiate Stroke Working Party, 2012). However, it could potentially be detrimental for patients if this intervention were applied when a patient is presenting with a mood disorder, so it is imperative that professionals are making the distinction between PSEL and PSD correctly. Furthermore, Cummings *et al.* (2006) refer to the common confusion between mood, affect, and emotional behaviour, and advocate for the use of diagnostic classification systems like the Diagnostic and Statistical Manual, Fourth Edition (DSM-IV) and International Classification of Diseases, Tenth Edition (ICD-10) to provide clarity and assist with these issues. Cummings *et al.* (2006) refer to the use of the diagnostic term ‘involuntary

emotional expression disorder' (IEED) as a medically accurate and unifying term encompassing all disorders of emotional expression, including PSEL. Cummings *et al.* (2006) identified that the most important step in the differential diagnosis of IEED is distinguishing crying as part of IEED versus crying in the context of a depressive episode.

In line with the evidence and guidance on post-stroke mood disorders like anxiety and depression, this study highlights the need for a clear diagnostic system and the use of screening measures to assist with the identification and classification of PSEL where necessary. The Pathological Laughter and Crying Scale (PLACS; Robinson, Parikh, Lipsey, Starkstein & Price, 1993) is the only measure that has been validated for use with stroke populations, and may provide support for professionals when identifying and classifying PSEL. However, the use of this measure is only advised in combination with the use of other comprehensive evaluation by trained interviewers, and it is not possible to use this measure with individuals with aphasia (Lincoln *et al.*, 2012).

This study also raised the issue of confusion and limited knowledge of PSEL in junior or non-qualified/non-specialist staff. This adds further clarification to findings by Gillespie and Cadden (in preparation), who highlighted that PSEL was not consistently well understood or recognised by staff working in acute or non-specialist settings. As outlined in the aims and rationale for this study, guidelines on PSEL refer to psychological support, education and advice for patients and their families (Intercollegiate Stroke Working party, 2012; SIGN, 2010). If professionals do not have a clear understanding of PSEL, they are not likely to be able to offer accurate advice, education and support to patients and their families. This finding highlights a need for training and education for less experienced staff, and perhaps those working in non-specialist or acute hospital settings. This training may involve educating staff on the causes, maintenance and identification of PSEL, and may focus specifically on the use of screening measures, and differentiating between PSEL and PSD.

Limitations of the study

The major limitation of this study is the small sample size, and the implications for the generalisability of the results. Generalisability may be strengthened by the fact that participant recruitment was undertaken from specialist stroke services in both the East and West of Scotland, however, findings should be generalised with caution when considering populations outside of these two research sites. It is also important to note that participants were highly specialist, qualified, and experienced stroke clinicians, so again, findings should not be generalised outwith this population.

Future research directions and recommendations

Approaches to the treatment of PSEL are still under investigation, and recommendations and guidance (Intercollegiate Stroke Working Party, 2010; SIGN, 2010) currently advise on the use of pharmacological approaches and distraction strategies to manage it. However, the evidence base for the use of these approaches is tenuous (Champion, 2006; Hackett *et al.*, 2010). Gillespie and Cadden (in preparation) are currently investigating treatment approaches, and the report of their findings will follow.

In light of the findings regarding the emotional consequences of PSEL i.e. distress, embarrassment, shame, and negative thinking (see Subcategory 5), consideration of the use of a range of therapeutic models is indicated in terms of treatment of the emotional consequences of PSEL. Of course, the cognitive and behavioural model requires further thought (Champion, 2006), however, it may also be helpful to consider the application of other models such as mindfulness (Williams & Penman, 2011), compassion focussed (Gilbert, 2010), and acceptance-based (Hayes, Strosahl & Wilson, 2012) approaches.

This study has underlined many of the questions that academics and professionals in the fields of neurology and stroke have been asking for centuries. Some argue that being able to be sure of the aetiology of conditions like PSEL is simply a case of scientific curiosity, and that actually what matters for patients is how we identify and manage the condition.

However, in addition to the fact that guidelines emphasise the importance of information and education for patients and their families, it has long been known that individuals experience higher levels of psychological well being when they are able to ‘make meaning’ following personal trauma (Park & Ai, 2006).

What makes traumatic events so difficult is that they can often violate many of the beliefs and assumptions that individuals have about themselves and the world (Janoff-Bulman, 1992), which is similar to issues highlighted by participants in subcategory 5 ‘Seeing Patients as Individuals’. The work of Janoff-Bulman in the 1980’s with patients with breast cancer refers to the role of condition specific knowledge and understanding in ‘meaning making’, and the facilitation of psychological adjustment when faced with trauma, grief, and loss (Kernan & Lepore, 2009). Patients often search for the ‘why’ when faced with life threatening illness, and this research suggests that normalising, and providing education and knowledge to patients on PSEL may help them feel more in control and assist them with psychological adjustment. This argument supports the need for further investigation of the aetiology of PSEL. The use of new technologies such as sophisticated neuromaging equipment allow for the potential for some of the more general historical questions regarding the neurobiology of emotion and feeling to be answered (Parvizi *et al.*, 2001).

Declaration of Interest: The author conducted this systematic review as part of a portfolio thesis for doctoral training in clinical psychology (DClinPsychol) undertaken at the University of Edinburgh. This training place was jointly funded by NHS Education for Scotland and NHS Lothian health board. No other sponsorship, funding or support was received.

Journal Article References

- Allman, P., Hope, R. A., & Fairburn, C.G. (1990). Emotionalism following brain damage: a complex phenomenon. *Postgraduate Medical Journal*, 66, 818-821.
- American Psychological Association (2009). *Publication manual of the American Psychological Association (6th Ed.)*. Washington, DC: Author.
- Anderson, G., Vestergaard, K., Ingeman-Nielsen, M., & Lauritzen, L. (1995). Risk factors for post-stroke depression. *Acta Psychiatrica Scandinavia*, 92(3), 193-8.
- Axtell, R. E. (1991). *Gestures: the do's and taboos of body language around the world*. John Wiley & Sons: New York.
- Bandura, A. (1977). *Social Learning Theory*. Stanford University Press: California.
- Beck, A. T. (1979). *Cognitive Therapy of Depression*. The Guildford Press: New York.
- Beynon, J. (2002). *Masculinities and Culture*. Open University Press: Michigan.
- Brookshire, R. H. (1970). Control of involuntary crying behaviour emitted by a multiple sclerosis patient. *Journal of Communication Disorders*, 3, 171-176.
- Calvert, T., Knapp, P., & House, A. (1998). Psychological associations with emotionalism after stroke. *Journal of Neurology, Neurosurgery and Psychiatry*, 65, 928-929.
- Carota, A., Berney, A., Aybeck, S., Iaria, G., Staub, F., Ghika-Schmid, F., et al. (2005). A prospective study of predictors of post-stroke depression. *Neurology*, 64, 428-438.
- Champion, A. J. (2006). *Neuropsychological Rehabilitation: A Resource for Group-based Education and Intervention*. Wiley: Chichester.
- Charmaz, K. (2006). *Constructing Grounded Theory: A Practical Guide Through Qualitative Analysis*. Sage: London.
- Cummings, J. L., Arciniegas, D., Brooks, B. R., Herndon, R. M., Lauterbach, E. C., Piro, E.P., et al. (2006). Defining and diagnosing involuntary emotional expression disorder. *CNS Spectrums*, 11(6), 1-7.

- Dereux, L., Ostrowsky, K., & Nighoghossian, N. (1997). Severe pathological crying after left anterior choroidal artery infarct. Reversibility with paroxetine treatment. *Stroke*, 28, 1464-1466.
- Eccles, S., House, A., & Knapp, P. (1999). Psychological adjustment and self-reported coping in stroke survivors with and without emotionalism. *Journal of Neurology, Neurosurgery, and Psychiatry*, 67, 125-126.
- Feinstein, A., Feinstein, K., Gray, T., & O'Connor, P. (1999). Pathological laughing and crying in multiple sclerosis: a preliminary report suggesting a role for the prefrontal cortex. *Multiple Sclerosis Journal*, 5(2), 69-73.
- Feldman, R., Meyer, J., & Qunezer, L. (1997). *Principles of Neuropsychopharmacology*. Sunderland: Sinauer Associates.
- Gilbert, P. (2010). *The Compassionate Mind*. Constable & Robinson Ltd: London.
- Gillespie, D. C., & Cadden, A. P. (in preparation). Non-pharmacological treatment of post-stroke emotional lability in Scotland's specialist stroke services.
- Giroud, M., Lemesie, M., Madinier, G., Billiar, T., & Dumas, R. (1997). Unilateral lenticular infarcts: radiological and clinical syndromes, aetiology, and prognosis. *Journal of Neurology, Neurosurgery and Psychiatry*, 63, 611-615.
- Goldstein, K. (1939). *The organism: a holistic approach to biology derived from pathological data in man*. New York: American Book.
- Hackett, M. L., Yang, M., Anderson, C. S., Horrocks, J. A., & House, A. (2010). Pharmaceutical interventions for emotionalism after stroke (Review). *The Cochrane Database of Systematic Reviews* (2010)2.
- Hayes, S. C., Strosahl, K. D., & Wilson, K. G. (2012). *Acceptance and Commitment Therapy: The Process and Practice of Mindful Change*. The Guildford Press: New York and London.

- House, A. O., Hackett, M. L., Anderson, C. S., & Horrocks, J. A. (2008). Pharmaceutical interventions for emotionalism after stroke. *Cochrane Database of Systematic Reviews* (2008)2.
- House, A., Dennis, M., Molyneux, A., Warlow, C., & Hawton, K. (1989). Emotionalism after stroke. *BMJ*, *298*(3), 991-994.
- Intercollegiate Stroke Working Party (2012). *National Clinical Guidelines for Stroke*. (4th Ed.). Royal College of Physicians: Surrey.
- Janoff-Bulman, R. (1992). *Shattered assumptions: Towards a new psychology of trauma*. New York: Free Press.
- Kasprisin, A. (2004). Alternative cognitive therapy for emotional instability (pathologic laughing and crying). *Physical and Medical Rehabilitation Clinics of North America*, *15*, 883-917.
- Kernan, W. D. & Lepore, S. J. (2009). Searching for and making meaning after breast cancer: Prevalence, patterns, and negative affect. *Social Science & Medicine*. *68*, 1176-1182.
- Kim, J. S. (2002). Post-stroke emotional incontinence after small lenticulocapsular stroke: a correlation with lesion location. *Journal of Neurology*, *249*(7), 805-810.
- Kim, J. S. & Choi-Kwon, S. (2000). Post stroke depression and emotional incontinence: correlation with lesion location. *Neurology*, *54*, 1805-1810.
- Kim, J. S., Choi, R. N., Kwon, S. U., & Seo, Y. S. (2002). Inability to control anger of aggression after stroke. *Neurology*, *58*(7), 1106-1106.
- Leiberman, A. & Benson, F. (1977). Control of emotional expression in pseudobulbar palsy. *Archive of Neurology*, *34*, 717-729.
- Lincoln, N. B., Kneebone, I. I., Macniven, J. A. B., & Morris, R. C. (2012). *Psychological Management of Stroke*. Chichester: John Wiley & Sons Ltd.

- Mathers, C. D., Boerma, T., & Ma Fat, D. (2009). Global and regional causes of death. *British Medical Bulletin*, 92, 7-32.
- Miller, A., Pratt, H., & Schiffer, R. B. (2011). Pseudobulbar affect: the spectrum of clinical presentations, etiologies and treatments. *Expert Reviews in Neurotherapy*, 11(7), 1077-1088.
- Morris, P. L. P., Robinson, R. G., & Raphael, B. (1993) Emotional lability after stroke. *Australian and New Zealand Journal of Psychiatry*, 27(4), 601-605.
- Mouton, P., Remy, A., & Cambon, H. (1994). Spasmodic laughter caused by unilateral involvement of the brain stem. *Reviews in Neurology (Paris)*, 150, 302-303.
- Park, C. L. & Ai, A. L. (2006). Meaning making and growth: New directions in research on survivors of trauma. *Journal of Loss and Trauma: International Perspectives on Stress & Coping*, 11(5), 389-407.
- Parvizi, J., Anderson, S. W., Martin, C. O., Damasio, H., & Damasio, A. R. (2001). Pathological laughter and crying: a link to the cerebellum. *Brain*, 124, 1708-1719.
- Perrins, J., King, N., & Collins, J. (1998). Assessment of long-term psychological well-being following intensive care. *Intensive and Critical Care Nursing*, 14(3), 108-116.
- Poeck, K. (1969). Pathophysiology of emotional disorders associated with brain damage. In: Vinken, P. J. & Bruyn, G. W. (Eds.). *Handbook of Clinical Neurology*. (pp. 343-367). Holland: Holland Publishing Co.
- Prigatano, G. P. (2010). *The Study of Anosagnosia*. Oxford University Press: New York.
- Raffii, M. S. & Hillis, A. E. (2006). Compendium of cerebrovascular diseases. *International Review of Psychiatry*, 5, 395-407.
- Robinson, R. G., Parikh, R. M., Lipsey, J. R., Starkstein, S. E., & Price, T. R. (1993). Pathological laughter and crying following stroke; Validation of a measurement scale and a double-blind treatment study. *American Journal of Psychotherapy*, 150, 286-293.

- Shamir, M. & Travis, J. (2002). *Boys Don't Cry: Rethinking Narratives of Masculinity and Emotion in the U.S.* New York: Columbia University Press.
- SIGN (2010). *Management of Patients with Stroke: Rehabilitation, Prevention and Management of Complications and Discharge Planning (SIGN publication 118)*. Retrieved 10 January 2013 from <http://www.sign.ac.uk/pdf/sign118.pdf>
- Stone, P., Ream, E., Richardson, A., Thomas, H., Andrews, P., Campbell, P., *et al.* (2003). Cancer-related fatigue - a difference of opinion? Results of a survey of healthcare professionals, patients, and caregivers. *European Journal of Cancer Care*, 12(1), 20-27.
- Strauss, A. & Corbin, J. (1990). *Basics of Qualitative Research: Grounded Theory Procedures and Techniques*. London: Sage Publications.
- Tang, W. K., Chen, Y. K., Lu, J. Y., Mok, V. C., Xiang, Y. T., Ungvari, G. S., *et al.* (2009). Microbleeds and post-stroke emotional lability. *Journal of Neurology, Neurosurgery and Psychiatry*, 80(10), 1082-1086.
- Tei, H. & Sakamoto, Y. (1997). Pontine infarction due to basilar artery stenosis presenting as pathological laughter. *Neuroradiology*, 39, 190-191.
- Williams, M., & Penman, D. (2011). *Mindfulness: A practical guide to finding peace in a frantic world*. Paitkus: London.
- Willig, C. (2008). *Introducing Qualitative Research in Psychology*. New York: Open University Press.
- Wilson, S. A. K. (1924). Some problems in neurology - Pathological laughing and crying, *Journal of Neurology and Psychopathology*, 16, 299-333.
- Wortzel, H. S., Oster, T. J., Anderson, C. A., & Archiegas, D. B. (2008). Pathological laughing and crying: epidemiology, pathophysiology and treatment. *CNS Drugs*, 22(7): 531-545.

CHAPTER FOUR: EXTENDED METHOD

Design

Qualitative research can be a useful way of learning about a person's experiences, uncovering what lies behind a phenomenon, and gaining fresh insights into something which little is known about (Strauss & Corbin, 1990). Qualitative researchers are concerned with meaning, interpretation, and how people experience particular circumstances and make sense of the world around them (Willig, 2008).

Grounded Theory was originally developed by sociologists Glaser and Strauss (1967). The empirical research submitted as part of this thesis utilises a modern version of constructivist Grounded Theory methodology (Charmaz, 2006), and although this version has deviated somewhat from Glaser and Strauss' (1967) classical constructivist method, it maintains the basic qualities and guidelines of Grounded Theory e.g. the use of theoretical sampling for theory development, coding and memo writing, and the generation of conceptual categories (Charmaz, 2006).

The purpose of Grounded Theory is to create a theory which illustrates, and remains faithful to, the area which is being studied (Strauss & Corbin, 1990). Grounded Theory research begins with an area of study, discovers the concepts within it, and develops and verifies them through the systematic collection and analysis of data to create a theory (Strauss & Corbin, 1990). It is unique in the way it moves back and forth between gathering data, coding and analysis before returning to gather more data to answer further queries until "saturation" is reached (when no new phenomena are reported in the data). Through engaging in the processes of the Grounded Theory method, it enables the researcher to retain control of the research process and gives greater analytic power to the results (Charmaz, 2006).

In Charmaz's (2006) approach to Grounded Theory, the researcher's interpretation of the studied phenomenon is used to construct a theoretical model. The researcher's background, questions, and decisions are clearly acknowledged as contributing to the resulting theory in Charmaz's version of Grounded Theory (Willig, 2008). In light of this, it is important to remain aware of the researcher's preconceived ideas, and to be cautious about the impact these may have on the resulting theory. In this study, the researcher used a technique in Grounded Theory known as 'memoing' to monitor and minimise the impact of her own preconceived ideas on the data and resulting theory.

Procedure

Participants

Participants were approached by specialist stroke service managers via the Lothian Stroke Managed Clinical Network (MCN) in the first instance and were given the participant information sheet (Appendix 5). Once consent to participate had been obtained, the researcher contacted participants directly to arrange interviews and give participants an opportunity to ask any questions. Interviews took place during participants' working hours, and this was negotiated with senior staff and managers.

Eligibility

For the purpose of this study, a specialist stroke service was defined as any post-acute or rehabilitation service that designated itself a "stroke service", or any other treatment setting in which >50% of patients treated had a confirmed diagnosis of stroke.

Participants eligible to participate were qualified clinical rehabilitation staff working in specialist stroke services in Scotland (medical, occupational therapy, nursing, physiotherapy, speech and language therapy, and clinical psychology). In order to ensure that participants had gained enough clinical experience of PSEL, it was decided that participants were required to have worked in specialist stroke services for a minimum of

two years. Gillespie and Cadden (in preparation) highlighted a number of important issues regarding the conceptualisation and experience of PSEL amongst specialist stroke service staff. Gillespie and Cadden (in preparation) discovered that professionals working in acute settings reported seeing a lesser number of individuals with PSEL in their day-to-day practice than prevalence rates would indicate, and some less experienced professionals working in acute settings said that they rarely or never saw it. As highlighted in the aims of the study, these findings identified the need for further exploration of specialist professionals' conceptualisations of PSEL, and informed the development of the protocol for this study. In light of Gillespie and Cadden's (in preparation) findings, junior staff, and staff working in acute settings were excluded as they were not likely to have been able to provide the researcher with relevant data.

Recruitment numbers

A total of seven participants were recruited from specialist stroke services across Edinburgh (n= 5) and Glasgow (n= 2). Participants included professionals from community nursing (n=2), medicine (n=1), speech and language therapy (n=1), physiotherapy (n=1), occupational therapy (n=1), and clinical psychology (n=1).

Data Collection and Analysis

Grounded Theorists stress the importance of the simultaneous collection of data and analysis. The researcher is required to move back and forth between data collection and analysis in order to shape and inform the emerging theory (Charmaz, 2006).

Theoretical sampling

Theoretical sampling is a technique used to aid theoretical development (Charmaz, 2006). It works by selecting subsequent participants based on the information which emerges from the data already coded (Sarantakos, 2005). This process provides a means of ensuring that new data add value, and that they work with the concepts already

compiled through a measure of fit and relevance (Glaser, 1978). New data are confirmed and disconfirmed to ensure the emerging theory develops rigor and parsimony.

Theoretical sampling allows the researcher to seek out specific sources of data likely to assist with the development and elaboration of the emerging categories identified in early data. For this research, following from the first interview each subsequent participant was purposefully recruited following transcription and analysis of each interview, and the identification of categories. For example, it was apparent after the first and second interview that participants' ideas about the causes of PSEL required further exploration. To explore these ideas further, the researcher specifically asked participants what they thought the causes of PSEL were, which resulted in tentative ideas about the neurobiology of PSEL. Following this, the researcher recruited a participant with specialist medical training in neurology and pharmacology in order to explore this concept in further detail.

Saturation

In Grounded Theory, the researcher ceases data collection when it has been deemed that categories have been "saturated". "Saturation" has been achieved when collecting new data no longer creates fresh ideas or theoretical insights, or adds anything new to core theoretical categories (Charmaz, 2006). Therefore, it is always difficult to know exactly how many participants need to be recruited when beginning data collection. In this research, "saturation" of categories was reached after interview six; interview seven assisted with consolidation of existing categories, but did not add anything new.

Interviews

"Intensive interviewing" has been a method utilised for data gathering in various types of qualitative research (Charmaz, 2006). An intensive interview permits an in-depth exploration of a particular topic with a person who has the relevant experience (Charmaz, 2006). By using this method of data collection, the researcher was able to

exert a higher level of control over the construction of data than they would using other methods such as ethnography, which can often take a more structural than process oriented approach (Charmaz, 2006).

Individual interviews were conducted and recorded (using an encrypted audio recording device) with qualified specialist stroke professionals working in post-acute rehabilitation settings,. It was important to explore the emergence of participants' stories and narratives about their ideas and experiences. The interview questions were open-ended and non-judgemental, and designed to gather rich data as well as striving to avoid imposing pre-conceived ideas onto the participant (Charmaz, 2006). Examples of questions from the semi-structured interview include *"What does post-stroke emotional lability mean to you?"*, *"Can you tell me about your experiences of working with post-stroke emotional lability?"*, *"Why do you think people experience it?"* and *"What does it look like when you see it?"*. See Appendix 6 for a comprehensive overview of questions from the semi-structured interviews.

Interview questions were reviewed both during and after each interview, and new questions were formulated to assist with the exploration of emerging subjects and categories. Interviews were transcribed and analysed before the next interview was conducted to aid with the formulation of new questions to facilitate exploration of emerging conceptual categories and developing theory. It was necessary to conduct two of the participant interviews on the same day due to constraints on both the researcher and participants, leaving no time for transcription and analysis between interviews as per the usual protocol with Grounded Theory methodology. Interviewing in 'batches' is sometimes used in Grounded Theory for practical reasons (Strauss & Corbin, 1990). The same protocol was followed as with individual interviews regarding transcription and analysis for the interviews conducted in a 'batch'. Murphy, O'Shea, Cooney and Casey (2007) utilised a similar approach in their study on the quality of life of older people with a disability.

Interviews ranged in length from 52 - 72 minutes (mean 62 minutes), and were transcribed verbatim by the researcher using Microsoft Word.

Arranging interviews with participants

Initially, participants were approached by specialist service managers via the Stroke Managed Clinical Network (MCN) to see if they were interested in taking part. If they expressed an interest at that point, the manager then passed the participant's contact details to the researcher, and interviews were arranged. Interviews were arranged via NHS managers in three NHS hospitals in Edinburgh and Glasgow, and at a time convenient to the participant. Participants were given the participant information sheet (Appendix 5) to read before interviews (although they had received this prior to giving consent to their participation), and they had the opportunity to ask any questions before completing the consent form. Please see Appendix 7 for a copy of the consent form. The participants were also given the supplementary information sheet (Appendix 8) to fill in at this point. After interviews, participants were given further opportunity to ask questions, and they were also asked at this point if they would be happy to be contacted at a later date to discuss the findings.

In order to improve the quality of the data, the researcher met with participants 4 and 5 when data collection was complete to discuss the analysis. The feedback from these participants could then be incorporated into the theory. For example, participant four referred to support from multidisciplinary team colleagues, and the importance of this for working in stroke. This feedback contributed to an element of the resulting model (see Chapter Three for the details of this).

Memo writing and the Reflective Journal

Charmaz (2006) describes memo writing as a way of making steps from data collection to writing drafts of papers. Birks and Mills (2011) advocate that memo writing is crucial for

developing a theory. Memos help the researcher stay engaged with the data, prompting them to analyse data and codes constantly throughout the research process from the early stages, capturing the researcher's thoughts, comparisons and connections made (Charmaz, 2006). Memos also help highlight gaps in the analysis, and assist with the formulation of codes and raising focussed codes to theoretical categories.

Charmaz (2006) encourages the use of 'freewriting' to encourage the composition of fresh material, unlearn past immobilising habits, and to write in a natural voice, ignoring grammar, organisation, logic, evidence, and audience. The researcher used this technique to practice the art of memo writing and reflection throughout the research process. Charmaz (2006) suggested that freewriting can release and open our minds and imaginations, increasing our receptivity to the world.

Mays and Pope (1996) highlighted that the primary investigative tool in qualitative research is the 'person' of the researcher. In light of this, the researcher kept a reflective journal at all times during the research process, so that any thoughts that she had about the research could be recorded at the time they occurred to her whilst they were fresh. This journal contained the majority of the memos throughout the research process. The researcher recorded her own thoughts, feelings, observations, and ideas about all aspects of the research process in the journal. This strategy proved to be a vital aid to data analysis and theory formulation, and information from the memos were crucial in aiding the researcher's thinking.

Reflexivity

Reflexivity is defined as "an active process of systematically developing insight into your work as a researcher to guide your future actions" (Birks and Mills, 2011, p. 45). Being reflexive gives the researcher plentiful opportunity to think in detail about their experience as a researcher in relation to the field they are studying. It also encourages them to consider how their past experiences, thoughts, and feelings can impact upon

their work, making links between the emotional and personal on the one hand, and the methodical, logical, and stringent on the other (Lofland & Lofland, 1995, p. 15). Birks and Mills (2011) refer to how reflexive memo writing can also provide researchers with a way of monitoring methodical congruence when undertaking a Grounded Theory study.

Charmaz (2006) also maintains that reflexivity should be incorporated as a strategy in Grounded Theory research design, and emphasises its role in improving quality. Working reflexively improves validity in research as it encourages the researcher to be critical, thoughtful and sensitive to their own role and how they might influence the analysis and outcome as a result of their own prior assumptions and experience (Willig, 2008).

Strauss and Corbin (1994) refer to a concept known as 'reciprocal shaping', which highlights how the relationship between the researcher and participant facilitates the generation of the data. Meaning is co-constructed by the interactions and conversations between the researcher and participants. This is the essence of a constructivist approach (Charmaz, 2006). It is assumed that this process is implicit when using semi-structured interviews, and it should be considered mindfully throughout the research process. As with any relationship or interaction, there are likely to be dynamics between the researcher and participants, and the researcher thought it was important to be reflexive about these dynamics during the process. The researcher was a trainee clinical psychologist, relatively new to the speciality of stroke, participants were all mainly older and more experienced (some with in excess of 20 years experience) stroke clinicians, and the researcher noticed these dynamics during interviews and interactions with participants.

The age and inexperience of the researcher is a potential strength of the study in that other than what she had read in the literature in preparing the research proposal, she had very little knowledge or experience of working with PSEL, so she had had limited opportunity to develop preconceived ideas of the concept prior to conducting the

research. Therefore, the researcher was able to maintain a relatively neutral stance to data collection in the initial stages, although of course she developed further in depth knowledge and understanding later in the process as she became more engaged and immersed in data collection and analysis.

However, the inexperience of the researcher also created some potential issues for data collection, and this was captured in the reflexive memos. Some of the researcher's memos referred to how she felt about the dynamic between herself as a young inexperienced stroke clinician interviewing older experienced stroke clinicians; some of the more experienced participants were highly verbose, and one participant in particular often discussed topics at length that were not directly related to the questions asked by the researcher. Willig (2008) highlights the importance of keeping interviews focussed, and she states that it is the role of the researcher to draw participants back to the most relevant points and questions during interviews. One of the researcher's memos reflected on how she felt a presence of authority in the room with some of the participants during interviews due to their age and level of experience, and she discussed how she may have inhibited her wish to take more control of the interview process at those times due to this dynamic, and how this may have had an impact on the data.

Another memo also reflected on how these 'powerful' participants' narrative styles were often intellectual in nature, and at times the researcher wondered whether she resisted asking certain questions of these particular participants during interviews due to feeling overwhelmed by the wealth of knowledge and information provided in the narratives of these participants. However, the researcher's experience of working therapeutically with patients as a trainee clinical psychologist enabled a good awareness of these issues, which are known in the psychodynamic literature as transference and counter-transference (Racker, 2001). The researcher's prior experiences of these relational dynamics meant that she was able to notice them in the moment, reflect on them, and

use them to interpret and create meaning in the research process in a similar way she would do in her clinical work with patients.

Coding

Charmaz (2006) refers to Grounded Theory coding as the ‘bones’ of analysis, and these ‘bones’ need assembling into a working skeleton. Coding in Grounded Theory allows the researcher to stay close and familiar with the data, and to ask analytic questions. During coding, the researcher is able to define what is happening in the data, and begin to grapple with what it means (Charmaz, 2006).

In initial coding, Grounded Theorists remain open to exploring whatever theoretical possibilities they can discern in the data. At this stage, it is important to look closely at the actions and language in the narratives, and wherever possible, code data as actions (gerunds), and *in vivo* (Charmaz, 2006). Glaser (1978) demonstrated how using gerunds in coding assists you with sticking closely with the data, and helps with the detection of process. Using action based, *in vivo* codes keeps theory grounded, and prevents the researcher from making conceptual theoretical leaps without having done the necessary analytic work (Birks & Mills, 2011).

‘Initial codes are provisional, comparative, and grounded in the data’ (Charmaz, 2006, p. 48), and later focussed coding is used to begin synthesizing and explaining larger segments of data. Focussed coding compares and tests frequent earlier codes against other data, and ‘requires making decisions about which initial codes make the most analytic sense to categorise your data incisively and completely’ (Charmaz, 2006, p. 57). The final stage, known as theoretical coding, specifies possible relationships between categories developed in focussed coding (Charmaz, 2006).

Initial line-by-line coding was used in the study, which helped the researcher generate ideas to develop and explore further in subsequent interviews. Focussed coding was then

used to synthesize and explain larger amounts of data, and to determine the adequacy of the more significant and frequent earlier codes. Finally, theoretical coding was used to pull focussed codes together, substantiating how they related to one another as hypotheses to be integrated into a theory. Theoretical codes must earn their way into your grounded theory (Glaser, 1978). Coding is a dynamic process, which involves revisiting data and categories over and over as more data is obtained. An illustration of line-by-line and focussed coding of participant 5's interview is provided on the next page.

Participant Narrative	Line-by-line coding	Focussed coding
<p><u>Researcher</u>: so it, it interrupts your work with that person, its something that's uncontrollable, and that can happen</p> <p><u>Participant</u>: it can happen yes, but of course it takes more than that one session to determine if it really is lability, or whether that person perhaps is presenting with a mood disorder, until you get the opportunity to screen for that and get to know that person, I think that initial contact of, of deciding what the best strategy to cope with that, it takes a bit longer than that first session, so you have to give somebody a little bit more time until its, its really clear what the best strategy is</p> <p><u>Researcher</u>: and how would you, keeping in mind, being unsure when, when you first meet someone whether its emotional lability or whether it's a mood disorder, how would you go about deciding that?</p> <p><u>Participant</u>: well, on paper it sounds black and white and quite straightforward, but its never quite that straightforward. I mean when we, when somebody comes into our stroke service, we always screen for mood, and we use the Hospital Anxiety and Depression scale for that, erm, so sometimes that's helpful because if you get quite clear feedback from that then that leads into a discussion about how somebody has been feeling, is this score reflective of their mood</p>	<p><i>Reflecting on the process of assessment and distinguishing between PSEL and PSD</i></p> <p><i>Knowing that the patient might need time</i></p> <p><i>Reflecting on getting confused</i></p> <p><i>Using screening measures</i></p> <p><i>Getting feedback</i></p> <p><i>Having discussions with the patient about how they feel</i></p>	<p><i>Seeing it and identifying it</i></p> <p><i>Needing to be sure</i></p> <p><i>Feeling confused</i></p> <p><i>Being careful and thorough in assessment</i></p>

The researcher must remain faithful to coding principles in Grounded Theory so as not to contaminate the resulting theory with their own experience and ideas (Charmaz, 2006).

Interviews were listened to, and transcripts revisited a number of times during the research process, and three transcripts were coded by a social sciences colleague

familiar with Grounded Theory principles, and emerging codes and categories were compared in order to manage potential bias. Categories were also discussed with participants 4 and 5, and compared with relevant literature to strengthen the credibility of the resulting theory.

Evaluating Grounded Theory

Charmaz (2006) discusses how an audience will judge the usefulness of a method by the quality of the final product. She outlines how making sense of the research journey retrospectively helps the researcher imagine what the end product is going to look like, and that the researcher is able to do that because they have been immersed in the process from the outset. However, Charmaz (2006) highlights how the 'lines become blurred' for the researcher's audience. Charmaz (2006) outlines a set of criteria for the evaluation of Grounded Theory research (credibility, originality, resonance, and usefulness). She notes how a strong combination of originality and credibility gives a study greater resonance and usefulness, which makes it a more valuable contribution, and can give an audience confidence in the logic and rigor of the research process.

Credibility

Credibility is created by obtaining an 'intimate familiarity' with the studied concept or setting. Data should be broad and sufficient, and should have depth. The researcher is required to make rigorous comparisons between codes and categories, and categories should consider a wide range of empirical observations. There should be strong logical links between gathered data and resulting analysis, and enough evidence should be provided for the reader to form their own opinion in line with the researcher's opinion (Charmaz, 2006).

Although all coding and analysis in the study was completed by hand using large sheets of paper, pens and sticky notes (Charmaz, 2006), NVivo 10 (qualitative data analysis

computer software) was used to record and monitor the process i.e. conceptual categories were stored in 'nodes' in NVivo, and matched with excerpts from transcripts for process tracking and organisation purposes. The use of memos and the reflective journal were also useful strategies for the recording and monitoring of the above processes throughout the research journey. These techniques also act as an audit trail to add to the quality of the study. Birks and Mills (2011) refer to the importance of keeping an audit trail to maintain quality in Grounded Theory research.

Originality

Originality requires the categories to be fresh and offer new conceptual insights. The analysis should provide a new framework for the data, and the work should be of both social and theoretical significance. Charmaz (2006) stated that for research to be original, the Grounded Theory should 'challenge, extend, or refine current ideas, concepts, and practices'. The conceptualisation of PSEL is a relatively new area of research, and this was the first study examining professionals' conceptualisations of the condition. Previous research and guidelines have also identified this as a key area for further exploration.

Resonance

Resonance is achieved when the conceptual categories portray an overall picture of the studied phenomenon, which illuminates 'taken-for-granted' meanings and considers the influences and impact on people's lives. Resonance is also present when the results form links between larger collectives or institutions. The resulting theory should have meaning for the participants, but also for the individuals who experience the studied phenomenon personally. In this study, feedback on the findings was obtained from two participants to improve the validity of the theory, and their responses are implicit in the resulting theory. This process is known as participant validation (Mays & Pope, 1996; Willig, 2008).

Usefulness

The usefulness of Grounded Theory research is assessed on whether the resulting analysis offers interpretations that can be used in the world more generally. The Grounded Theory researcher should ask themselves if analytic categories suggest any generic processes that may have further reaching or more subtle implications, and whether the analysis can generate interest or ideas for research in other substantive or related areas. Charmaz (2006) also asks the researcher to consider ways in which the research contributes to the evidence base in a positive way. PSEL has been identified as a prevalent and distressing condition, about which little is known. This study is likely to directly inform staff training and knowledge, patient care, and the development of policy and clinical decision-making within stroke services in Scotland. The research has generated interest on both a local and national level.

Mays and Pope (1996) also refer to the idea that research of higher relevance and quality can be generalised beyond the setting in which it was conducted. This is implicit in that PSEL affects individuals with a range of neurological disorders, including stroke (Robinson, Parikh, Lipsey, Starkstein, & Price, 1993), amyotrophic lateral sclerosis (McCullagh, Moore, Gawel, & Feinstein, 1999), traumatic brain injury (Tateno, Jorge, & Robinson, 2004), multiple sclerosis (Feinstein, Feinstein, Gray, & O'Connor, 1997), and dementias such as Alzheimer's disease (Starkstein, Migliorelli, Teson, Petracca, Chemerinsky *et al.*, 1995).

Data Management

Confidentiality and anonymity

A number of measures were taken to protect the confidentiality and anonymity of participants. Participants were assigned a participant number, and recorded data and transcripts were labelled using these numbers. Any identifying features contained in the interviews were removed during transcription.

Data storage

Secure NHS email systems were used to arrange interviews with participants and interviews were recorded using an NHS encrypted recording device in line with Caldicott protocol, and stored on NHS and password protected computers. Any forms containing information that might reveal the identity of the participants such as consent forms, contact details, and supplementary data forms were stored securely and separately on NHS premises. NVivo 10 computer software was used to store, manage, and organise data and analysis. This software was installed on a University of Edinburgh password protected computer.

Modern Humanities Research Association Guidelines (MHRA) guidelines recommend that personal data e.g. consent forms should be archived for five years. Therefore, all data will be stored securely for five years before it is destroyed and disposed of in confidential waste. Electronic records will be also deleted after five years.

Ethical review

The researcher obtained Level 1 ethical approval from the University of Edinburgh School of Research Ethics Committee (see Appendix 9 for a copy of the approval letter).

This project was also reviewed and approved by the Lothian Research and Development (R&D) Office. The study was approved as an amendment to the Gillespie and Cadden (in preparation) study (see Appendix 10 for a copy of the confirmation letter). The NHS-to-NHS access for research approval arrangement was utilised for Greater Glasgow and Clyde (see Appendix 11 for a copy of the confirmation letter).

THESIS REFERENCES

- *Alexopoulos, G. S., Wilkins, V. M., Marino, P., Kellanopouluos, D., Reding, M., Kiossis, D. N., *et al.* (2012). Ecosystem focused family therapy in poststroke depression: a preliminary study. *International Journal of Geriatric Society*, 27, 1053-1060.
- Allman, P., Hope, R. A., & Fairburn, C.G. (1990). Emotionalism following brain damage: a complex phenomenon. *Postgraduate Medical Journal*, 66, 818-821.
- American Psychiatric Association (1994). *Diagnostic and Statistical Manual of Mental Disorders*, 4th ed. Washington, DC: American Psychiatric Association.
- Anderson, G., Vestergaard, K., Ingeman-Nielsen, M., & Lauritzen, L. (1995). Risk factors for post-stroke depression. *Acta Psychiatrica Scandinavia*, 92(3), 193-8.
- Axtell, R. E. (1991). *Gestures: the do's and taboos of body language around the world*. John Wiley & Sons: New York.
- Bandura, A. (1977). *Social Learning Theory*. Stanford University Press: California.
- Beck, A. T. (1979). *Cognitive Therapy of Depression*. The Guildford Press: New York.
- Beynon, J. (2002). *Masculinities and Culture*. Open University Press: Michigan.
- Birks, M., & Mills, J. (2011). *Grounded Theory*. London: Sage.
- Bradbury, C. L., Christensen, B. K., Lau, M. A., Ruttan, L. A., Arundine, A. L., & Green, R. E. (2008). The efficacy of cognitive behaviour therapy in the treatment of emotional distress after acquired brain injury. *Archives of Physical Medicine and Rehabilitation*, 89(12), 61-68.
- British Psychological Society (2011). *The Use of Psychological Tests in Healthcare Research*. The British Psychological Society: Leicester.
- British Psychological Society (2004). *Style Guide*. The British Psychological Society: Leicester.

- Brookshire, R. H. (1970). Control of involuntary crying behaviour emitted by a multiple sclerosis patient. *Journal of Communication Disorders*, 3, 171-176.
- Broomfield, N., Laidlaw, K., Hickabottom, E., Murray, M. F., Pendrey, R., Gillespie, D. C. (2010). Post-stroke depression: The case for augmented, individually tailored cognitive behavioural therapy. *Clinical Psychology and Psychotherapy*, 8(3), 202-217.
- Calvert, T., Knapp, P., & House, A. (1998). Psychological associations with emotionalism after stroke. *Journal of Neurology, Neurosurgery and Psychiatry*, 65, 928-929.
- Carota, A., Berney, A., Aybeck, S., Iaria, G., Staub, F., Ghika-Schmid, F., et al. (2005). A prospective study of predictors of post-stroke depression. *Neurology*, 64, 428-438.
- Centre for Reviews and Dissemination (2009). *Systematic reviews: CRD's guidance for undertaking reviews in healthcare*. York: CRD, University of York.
- Chambless, D. L., & Hollen, S. D. (1998). Defining empirically supported therapies. *Journal of Consulting and Clinical Psychology*, 66(1), 7-18.
- Champion, A. J. (2006). *Neuropsychological Rehabilitation: A Resource for Group-based Education and Intervention*. Wiley: Chichester.
- Charmaz, K. (2006). *Constructing Grounded Theory: A Practical Guide Through Qualitative Analysis*. Sage: London.
- Christensen, A., & Jacobson, N. S. (1994). Who (or what) can do psychotherapy: The status and challenge of nonprofessional therapies. *Psychological Science*, 5, 8-14.
- Cohen, J. (1988). *Statistical Power Analysis for the Behavioral Sciences* (2nd ed.). Hillsdale, NJ: Earlbaum.
- Craig, P., Dieppe, P., Macntyre, S., Michie, S., Nazareth, I., & Petticrew, M. (2008). *Developing and evaluating complex interventions: new guidance*. Medical Research Council. doi: <http://dx.doi.org/10.1136/bmj.a1655>
- Cummings, J. L., Arciniegas, D., Brooks, B. R., Herndon, R. M., Lauterbach, E. C., Piore, E.P., et al. (2006). Defining and diagnosing involuntary emotional expression disorder. *CNS Spectrums*, 11(6), 1-7.

- de Man-van Ginkel, Gooskens, F., Schuumans, M. J., Lindeman, E., Hafsteinsdottir, T. B. (2010). A systematic review of therapeutic interventions for poststroke depression and the role of nurses. *Journal of Clinical Nursing*, 19, 3274-3290.
- Derex, L., Ostrowsky, K., & Nighoghossian, N. (1997). Severe pathological crying after left anterior choroidal artery infarct. Reversibility with paroxetine treatment. *Stroke*, 28, 1464-1466.
- Eccles, S., House, A., & Knapp, P. (1999). Psychological adjustment and self-reported coping in stroke survivors with and without emotionalism. *Journal of Neurology, Neurosurgery, and Psychiatry*, 67, 125-126.
- Feinstein, A., Feinstein, K., Gray, T., & O'Connor, P. (1999). Pathological laughing and crying in multiple sclerosis: a preliminary report suggesting a role for the prefrontal cortex. *Multiple Sclerosis Journal*, 5(2), 69-73.
- Feinstein, A., Feinstein, K., Gray, T., & O'Connor, P. (1999). Pathological laughing and crying in multiple sclerosis: a preliminary report suggesting a role for the prefrontal cortex. *Multiple Sclerosis Journal*, 5(2), 69-73.
- Feldman, R., Meyer, J., & Qunezer, L. (1997). *Principles of Neuropsychopharmacology*. Sunderland: Sinauer Associates.
- Fraser, C. J., Christensen, H., & Griffiths, K. M. (2005). Effectiveness of treatments for depression in older people. *Medical Journal of Australia*, 182, 627-632.
- Friedman, L. M., Furberg, C., & DeMets, D. (1998). Multicenter Trials. In: *Fundamentals of Clinical Trials*. New York: Springer-Verlag; 345-356.
- Gilbert, P. (2010). *The Compassionate Mind*. Constable & Robinson Ltd: London.
- Gillespie, D. C., & Cadden, A. P. (in preparation). Non-pharmacological treatment of post-stroke emotional lability in Scotland's specialist stroke services.
- Giroud, M., Lemesie, M., Madinier, G., Billiar, T., & Dumas, R. (1997). Unilateral lenticular infarcts: radiological and clinical syndromes, aetiology, and prognosis. *Journal of Neurology, Neurosurgery and Psychiatry*, 63, 611-615.

- Glaser, B. G., & Strauss, A. L. (1967). *The discovery of grounded theory*. Chicago: Aldine.
- Glaser, B., G. (1978). *Theoretical Sensitivity*. Mill Valley, CA: The Sociology Press.
- Goldstein, K. (1939). *The organism: a holistic approach to biology derived from pathological data in man*. New York: American Book.
- Hackett, M. L., Anderson, C. S., House, A. O., & Halteh, C. (2008a). Interventions for preventing depression after stroke. *Cochrane Database of Systematic Reviews*, (2008)3.
- Hackett, M. L., Anderson, C. S., House, C. S., & Xia, J. (2008b). Interventions for treating depression after stroke. *Cochrane Database of Systematic Reviews*, (2008)4.
- Hackett, M. L., Yang, M., Anderson, C. S., Horrocks, J. A., & House, A. (2010). Pharmaceutical interventions for emotionalism after stroke (Review). *The Cochrane Database of Systematic Reviews* (2010)2.
- Hackett, M. L., Yapa, C., Parag, V., & Anderson, C. S. (2005). Frequency of depression after stroke: A systematic review of observational studies. *Stroke*, 36, 1330-1340.
- Hayes, S. C., Strosahl, K. D., & Wilson, K. G. (2012). *Acceptance and Commitment Therapy: The Process and Practice of Mindful Change*. The Guildford Press: New York and London.
- *Hibbard, M., Grober, S. E., Stein, P. N., & Gordon, W. A. (1992). Poststroke depression. In: A. Freeman & F. Dattilio (Eds.), *Comprehensive Casebook of Cognitive Therapy* (pp. 303-310). New York: Plenum Press.
- Hotopf, M. (2002). The pragmatic randomised controlled trial. *Advances in Psychiatric Treatment*, 8, 326-333.
- House, A. O., Hackett, M. L., Anderson, C. S., & Horrocks, J. A. (2008). Pharmaceutical interventions for emotionalism after stroke. *Cochrane Database of Systematic Reviews* (2008)2.

- House, A., Dennis, M., Molyneux, A., Warlow, C., & Hawton, K. (1989). Emotionalism after stroke. *BMJ*, 298(3), 991-994.
- IAPT (2011). *Increasing Access to Psychological Therapies Data Handbook: Guidance on recording and monitoring outcomes to support local evidence-based practice*. IAPT: London. Retrieved 2 June 2013 from <http://www.iapt.nhs.uk/silo/files/iapt-data-handbook-v2.pdf>
- Intercollegiate Stroke Working Party (2012). *National Clinical Guidelines for Stroke*. (4th Ed.). Royal College of Physicians: Surrey.
- Jacobson, N. S., & Christensen, A. (1996). Studying the effectiveness of psychotherapy: How well can clinical trials do the job? *American Psychologist*, 51, 1030–1039.
- Jacobson, N. S., & Truax, P. (1991). Clinical significance: A statistical approach to defining meaningful change in psychotherapy research. *Journal of Consulting and Clinical Psychology*, 59(1), 12-19.
- Janoff-Bulman, R. (1992). *Shattered assumptions: Towards a new psychology of trauma*. New York: Free Press.
- *Joo, H. M., Lee, S. J., Chung, Y. G., & Shin, Y. (2010). Effects of mindfulness based stress reduction program on depression, anxiety and stress in patients with aneurismal subarachnoid hemorrhage. *Journal of the Korean Neurosurgical Society*, 47, 345-351.
- Kasprisin, A. (2004). Alternative cognitive therapy for emotional instability (pathologic laughing and crying). *Physical and Medical Rehabilitation Clinics of North America*, 15, 883-917.
- Keppel, G., & Wickens, T. D. (1982). *Design and Analysis: A researcher's handbook*. Prentice Hall: NJ.
- Kernan, W. D. & Lepore, S. J. (2009). Searching for and making meaning after breast cancer: Prevalence, patterns, and negative affect. *Social Science & Medicine*. 68, 1176-1182.

- Kim, J. S. (2002). Post-stroke emotional incontinence after small lenticulocapsular stroke: a correlation with lesion location. *Journal of Neurology*, 249(7), 805-810.
- Kim, J. S. & Choi-Kwon, S. (2000). Post stroke depression and emotional incontinence: correlation with lesion location. *Neurology*, 54, 1805-1810.
- Kim, J. S., Choi, R. N., Kwon, S. U., & Seo, Y. S. (2002). Inability to control anger of aggression after stroke. *Neurology*, 58(7), 1106-1106.
- Knapp, P., Young, J., House, A., & Forster, A. (2000). Non-drug strategies to resolve psycho-social difficulties after stroke. *Age and Aging*, 29, 23-30.
- Kneebone, I. I., & Dunmore, E. (2000). Psychological management of post-stroke depression. *British Journal of Clinical Psychology*, 39, 53-65.
- Koder, D-A., Brodaty, H., & Anstey, K. J. (1996). Cognitive therapy for depression in the elderly. *International Journal of Geriatric Psychiatry*, 11, 97-107.
- Leiberman, A. & Benson, F. (1977). Control of emotional expression in pseudobulbar palsy. *Archive of Neurology*, 34, 717-729.
- *Lincoln, N. B., & Flannaghan, T. (2003). Cognitive behavioural therapy for depression following stroke. *Stroke*, 34, 111-115.
- *Lincoln, N. B., Flannaghan, T., Sutcliffe, L., & Rother, L. (1997). Evaluation of cognitive behavioural treatment for depression after stroke: a pilot study. *Clinical Rehabilitation*, 11, 114-122.
- Lincoln, N. B., Kneebone, I. I., Macniven, J. A. B., & Morris, R. C. (2012). *Psychological Management of Stroke*. Chichester: John Wiley & Sons, Ltd.
- Lincoln, N. B., Kneebone, I. I., Macniven, J. A. B., & Morris, R. C. (2012). *Psychological Management of Stroke*. Chichester: John Wily & Sons Ltd.
- Lofland, J., & Lofland, L. (1995). *Analyzing social settings: A guide to qualitative observation and analysis* (3rd ed.). Belmont, CA: Wadsworth.

- Lustman, P. J., Griffith, L. S., Freedland, K. E., Kissel, S. S., & Clouse, R. E. (1998). Cognitive behaviour therapy for depression in type 2 diabetes mellitus: A randomised, controlled trial. *Annals of Internal Medicine*, *129*, 613-621.
- Mathers, C. D., Boerma, T., & Ma Fat, D. (2009). Global and regional causes of death. *British Medical Bulletin*, *92*, 7-32.
- Mayo-Wilson, E., Grant, S., Hopewell, S., Macdonald., G., Moher, D., & Montgomery, P. (2013). Developing a reporting guideline for social and psychological intervention trials. *Trials*, *14*, 242-257.
- Mays, N., & Pope, C (Eds.). (1996). *Qualitative research in health care*. London: BMJ
- McCullagh, S., Moore, M., Gawel, M., & Feinstein, A. (1999). Pathological laughing and crying in amyotrophic lateral sclerosis: An association with prefrontal cognitive dysfunction. *Journal of the Neurology Sciences*, *169*, 43-48.
- McGill, F. (1980). *Go Not Gently: Letters from a patient with amyotrophic lateral sclerosis*. California: Arno Press.
- Mehta, S., Pereira, S., Janzen, S., McIntyre, McClure, A., Teasall, R. W. (2012). Effectiveness of psychological interventions in chronic stage of stroke: A systematic review. *Topics in Stroke Rehabilitation*, *19*(6), 536-544.
- Miller, A., Pratt, H., & Schiffer, R. B. (2011). Pseudobulbar affect: the spectrum of clinical presentations, etiologies and treatments. *Expert Reviews in Neurotherapy*, *11*(7), 1077-1088.
- Mitchell, P. H., Veith, R. C., Becker, K. J., Buzaitis, Cain, K.C, Tirschwell, D., & Teri, L. (2009). Brief psychosocial-behavioural intervention with antidepressant reduces poststroke depression significantly more than usual care with antidepressant. *Stroke*, *40*, 3073-3078.
- Moher, D., Liberati, A., Tetzlaff, J., & Altman, D. G. (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. *PLoS Medicine*, *6*(6). Retrieved 19 January 2013 from <http://www.prisma-statement.org/usage.htm>

- Mohr, D. C., & Goodkin, D. E. (1999). Treatment of depression in multiple sclerosis: review and meta-analysis. *Clinical Psychology: Science and Practice*, 6, 1-9.
- Morris, P. L. P., Robinson, R. G., & Raphael, B. (1993) Emotional lability after stroke. *Australian and New Zealand Journal of Psychiatry*, 27(4), 601-605.
- Mouton, P., Remy, A., & Cambon, H. (1994). Spasmodic laughter caused by unilateral involvement of the brain stem. *Reviews in Neurology (Paris)*, 150, 302-303.
- Murphy, K., O'Shea, E., Cooney, A., & Casey, D. (2007). *The Quality of Life of Older People with a Disability in Ireland*. National Council of Aging and Older People: Dublin.
- Park, C. L. & Ai, A. L. (2006). Meaning making and growth: New directions in research on survivors of trauma. *Journal of Loss and Trauma: International Perspectives on Stress & Coping*, 11(5), 389-407.
- Parvizi, J., Anderson, S. W., Martin, C. O., Damasio, H., & Damasio, A. R. (2001). Pathological laughter and crying: a link to the cerebellum. *Brain*, 124, 1708-1719.
- Patel, M. X., Doku, V. & Tennakoon, L. (2003). Challenges in recruitment of research participants. *Advances in Psychiatric Treatment*, 9, 229-238.
- Perrins, J., King, N., & Collins, J. (1998). Assessment of long-term psychological well-being following intensive care. *Intensive and Critical Care Nursing*, 14(3), 108-116.
[plosmedicine.org/article/info%3Adoi%2F10.1371%2Fjournal.pmed.1000097](https://pubmed.ncbi.nlm.nih.gov/1000097/)
- Poeck, K. (1969). Pathophysiology of emotional disorders associated with brain damage. In: Vinken, P. J. & Bruyn, G. W. (Eds.). *Handbook of Clinical Neurology*. (pp. 343-367). Holland: Holland Publishing Co.
- Prigatano, G. P. (2010). *The Study of Anosagnosia*. Oxford University Press: New York. Publishing.
- Racker, H. (2001). *Transference and Counter-Transference*. London: International Universities Press.

- Raffii, M. S. & Hillis, A. E. (2006). Compendium of cerebrovascular diseases. *International Review of Psychiatry*, 5, 395-407.
- *Rasquin, S. M. C., van de Sande, P., Praamstra, A. J., & van Heughten, C. M. (2009). Cognitive-behavioural evaluation for depression after stroke: Five single case studies on effects and feasibility. *Neuropsychological Rehabilitation*, 19(2), 208-222.
- Robinson, R. G., Parikh, R. M., Lipsey, J. R., Starkstein, S. E., & Price, T. R. (1993). Pathological laughter and crying following stroke; Validation of a measurement scale and a double-blind treatment study. *American Journal of Psychotherapy*, 150, 286-293.
- Robinson, R. G. (1997). Neuropsychiatric consequences of stroke. *Annual Review of Medicine*, 48, 217-299.
- Sarantakos, S. (2005). *Social Research*. Hampshire: Palgrave Macmillan.
- Shamir, M. & Travis, J. (2002). *Boys Don't Cry: Rethinking Narratives of Masculinity and Emotion in the U.S.* New York: Columbia University Press.
- SIGN (2008). *SIGN 50: A guideline developer's handbook. Revised Edition, November 2011*. Retrieved 19 February 2013 from <http://www.sign.ac.uk/guidelines/fulltext/50/>
- SIGN (2010). *Management of Patients with Stroke: Rehabilitation, Prevention and Management of Complications and Discharge Planning (SIGN publication 118)*. Retrieved 10 January 2013 from <http://www.sign.ac.uk/pdf/sign118.pdf>
- *Sims, J., Galea, M., Taylor, N., Dodd, K., Jespersen, S., Joubert, L., & Joubert, J. (2009). Regenerate: assessing the feasibility of a strength-training program to enhance the physical and mental health of chronic post stroke patients with depression. *International Journal of Geriatric Psychiatry*, 24, 76-83.
- Starkstein, S. E., Migliorelli, R., Teson, A., Petracca, G., Chemerinsky, E., Manes, F., et al. (1995). Prevalence and clinical correlates of pathological affective display in Alzheimer's disease. *Journal of Neurology, Neurosurgery & Psychiatry*, 59, 55-60.

- Stone, P., Ream, E., Richardson, A., Thomas, H., Andrews, P., Campbell, P., *et al.* (2003). Cancer-related fatigue - a difference of opinion? Results of a survey of healthcare professionals, patients, and caregivers. *European Journal of Cancer Care*, 12(1), 20-27.
- Strauss, A. & Corbin, J. (1990). *Basics of Qualitative Research: Grounded Theory Procedures and Techniques*. London: Sage Publications.
- Strauss, A. L., & Corbin, J. M. (1994). Grounded theory methodology: An overview. In N.K. Denzin & Y.S. Lincoln (Eds.), *Handbook of Qualitative Research* (pp. 273-85). Thousand Oaks, CA: Sage.
- Tang, W. K., Chen, Y. K., Lu, J. Y., Mok, V. C., Xiang, Y. T., Ungvari, G. S., *et al.* (2009). Microbleeds and post-stroke emotional lability. *Journal of Neurology, Neurosurgery and Psychiatry*, 80(10), 1082-1086.
- Tateno, A., Jorge, R. E., & Robinson, R. G. (2004). Pathological laughing and crying following traumatic brain injury. *Journal of Neuropsychiatry and Clinical Neurosciences*, 16, 426-434.
- Tei, H. & Sakamoto, Y. (1997). Pontine infarction due to basilar artery stenosis presenting as pathological laughter. *Neuroradiology*, 39, 190-191.
- *Thomas, S. A., Walker, M. F., Macniven, J. A., Haworth, H., & Lincoln, N. B. (2012). Communication and Low Mood (CALM): a randomized controlled trial of behavioural therapy for stroke patients with aphasia. *Clinical Rehabilitation*, 0(0), 1-11.
- *Towle, D., Lincoln, N. B., & Mayfield, L. M. (1989). Evaluation of social work on depression after stroke. *Clinical Rehabilitation*, 3, 89-96.
- van het Hoofd, M. G. V., Roelse, H., Kwakkel, G., Schepers, V. P. M., & van de Port, I. G. L. (2011). Systematic Review: Does exercise therapy affect depression or depressive symptoms after stroke? *Physical Therapy Review*, 16(3) 191-200.
- Williams, M., & Penman, D. (2011). *Mindfulness: A practical guide to finding peace in a frantic world*. Paitkus: London.

- Willig, C. (2008). *Introducing Qualitative Research in Psychology*. New York: Open University Press.
- Wilson, S. A. K. (1924). Some problems in neurology - Pathological laughing and crying, *Journal of Neurology and Psychopathology*, 16, 299-333.
- Wing, J., Cooper, J., & Sartorius, N. (1974). *Measurement and Classification of Psychiatric Symptoms: Instruction Manual for the PSE*. Cambridge University Press: London.
- Wortzel, H. S., Oster, T. J., Anderson, C. A., & Archiegas, D. B. (2008). Pathological laughing and crying: epidemiology, pathophysiology and treatment. *CNS Drugs*, 22(7): 531-545.

*references marked with asterisks refer to the articles selected for methodological appraisal in the systematic literature review.

Appendix 1: Author guidelines for the Journal

Rehabilitation Psychology

Rehabilitation Psychology[®] is now using a software system to screen submitted content for similarity with other published content. The system compares each submitted manuscript against a database of 25+ million scholarly publications, as well as content appearing on the open web.

This allows APA to check submissions for potential overlap with material previously published in scholarly journals (e.g., lifted or republished material). A similarity report will be generated by the system and provided to the *Rehabilitation Psychology* Editorial office for review immediately upon submission.

All new and revised manuscripts are to be submitted electronically (Word Documents are preferred) through the Manuscript Submission Portal.

Suitable Submissions

Rehabilitation psychology deals with the interplay of biological, psychological, social, environmental, and political factors that affect the functioning of persons with chronic health conditions or disability. Given the breadth of rehabilitation psychology, the journal's scope is broadly defined. Suitable submissions include:

Empirical Articles This format reports original empirical research which can include experimental investigations, survey research, evaluations of interventions, and outcome studies research.

Brief Reports This format may be appropriate for empirically sound studies that are limited in scope, contain novel or provocative findings that need further replication, or represent replications and extensions of prior published work. Brief Reports must use a 12-point Times New Roman type and 1-in. (2.54-cm) margins, and not exceed 265 lines of text plus references. These limits do not include the title page, abstract, author note, footnotes, tables, or figures.

Review Articles This format includes reviews of various types and formats. Reviews can include state-of-the-art review of empirical research (meta-analysis), reviews of professional, theoretical or public policy issues, or reviews designed to help practitioners solve common clinical problems (clinical management reviews).

Commentaries This format supports a submitted or previously published manuscript including explanation, critique or illustration of rehabilitation related issues or topics.

Case studies This format includes written analyses of one or more particular cases or case histories with a view to making generalizations in rehabilitation and that are of sufficient import to warrant attention.

Submissions are welcomed from authors in psychology and other health related disciplines.

Cover Letter

The cover letter accompanying the manuscript submission must include all authors' names and affiliations, addresses and phone numbers, as well as electronic mail addresses and fax numbers for possible use by the editorial office and later by the production office.

The cover letter should identify the type of submission category and include

- a statement of compliance with APA ethical standards in the conduct of the work reported in the manuscript
- a statement that the manuscript or data have not been previously published and that they are not presently under consideration for publication elsewhere
- a statement that all listed authors have contributed significantly to the work submitted for consideration
- a statement that the paper has been seen and approved by all authors

When the manuscript contains data or observations from a larger study, the cover letter should clarify the relationship between this submission and other papers from the study, specifically addressing potential overlap. Authors must be prepared to provide copies of related manuscripts or papers as part of the editorial review process.

Authors may suggest qualified reviewers of the manuscript, but these are considered advisory only.

Title

Should be accurate, descriptive, and no longer than 12 words. If the report is a clinical trial or a brief report this should be included in the title.

Abstract and Keywords

All manuscripts must include a structured abstract containing a maximum of 250 words typed on a separate page (page 2 of the manuscript). Abstracts must contain a brief statement about each of the following:

- Purpose/Objective
- Research Method/Design - including the number and type of participants
- Results
- Conclusions/Implications

After the abstract, please supply up to five keywords.

Impact and Implications Statement

At the start of each paper the authors should provide 2-3 bullet points, with the header "Impact", that states what the current paper adds to the literature and one to two practice or policy implications the findings. This is not a statement of the conclusions, rather a thoughtful series of statements highlighting the novel contribution of the work and translation of the findings for practice or policy. This section should be no more than 200 words.

Style of Manuscripts

The journal considers theoretical, empirical, and commentary papers relevant to rehabilitation psychology. Brief reports are considered.

Additional Information for Specific Publication Categories

Randomized Clinical Trials

Rehabilitation Psychology requires the use of the CONSORT (Consolidated Standards of Reporting Trials) reporting standards (i.e., a checklist and flow diagram) for randomized clinical trials. The checklist may be placed in an Appendix of the manuscript for review purposes.

Visit the CONSORT Statement Web site for more details and resources.

Nonrandomized Trials

Rehabilitation Psychology encourages the use of the most recent version of the TREND criteria (Transparent Reporting of Evaluations with Non-randomized Designs for nonrandomized designs, available on the TREND Web site).

Review Process

Papers will be evaluated for their importance to the field, scientific rigor, novelty, suitability for the journal, and clarity of writing. Manuscripts that do not conform to the submission guidelines may be returned without review.

A masked review process is used. To facilitate masked review, it is incumbent upon authors to see that the manuscript itself contains no clues to their identities. Authors' names, affiliations, and contact information should be included only in the cover letter.

Rehabilitation Psychology encourages translation of information and strives to review submitted articles in a timely manner.

Preparing Files for Production

If your manuscript is accepted for publication, please follow the guidelines for file formats and naming.

Please ensure that the final version for production includes a byline and full author note for typesetting.

Manuscript Preparation

Prepare manuscripts according to the *Publication Manual of the American Psychological Association* (6th edition). Manuscripts may be copyedited for bias-free language (see Chapter 3 of the *Publication Manual*).

Review APA's Checklist for Manuscript Submission before submitting your article.

Double-space all copy. Other formatting instructions, as well as instructions on preparing tables, figures, references, metrics, and abstracts, appear in the *Manual*.

Below are additional instructions regarding the preparation of display equations and tables.

Display Equations

We strongly encourage you to use MathType (third-party software) or Equation Editor 3.0 (built into pre-2007 versions of Word) to construct your equations, rather than the equation support that is built into Word 2007 and Word 2010. Equations composed with the built-in Word 2007/Word 2010 equation support are converted to low resolution graphics when they enter the production process and must be rekeyed by the typesetter, which may introduce errors.

To construct your equations with MathType or Equation Editor 3.0:

- o Go to the Text section of the Insert tab and select Object.
- o Select MathType or Equation Editor 3.0 in the drop-down menu.

If you have an equation that has already been produced using Microsoft Word 2007 or 2010 and you have access to the full version of MathType 6.5 or later, you can convert this equation to MathType by clicking on MathType Insert Equation. Copy the equation from Microsoft Word and paste it into the MathType box. Verify that your equation is correct, click File, and then click Update. Your equation has now been inserted into your Word file as a MathType Equation.

Use Equation Editor 3.0 or MathType only for equations or for formulas that cannot be produced as Word text using the Times or Symbol font.

Tables

Use Word's Insert Table function when you create tables. Using spaces or tabs in your table will create problems when the table is typeset and may result in errors.

Submitting Supplemental Materials

APA can now place supplementary materials online, available via the published article in the PsycARTICLES® database. Please see Supplementing Your Article With Online Material for more details.

References

List references in alphabetical order. Each listed reference should be cited in text, and each text citation should be listed in the References section.

Examples of basic reference formats:

- **Journal Article:** Herbst-Damm, K. L., & Kulik, J. A. (2005). Volunteer support, marital status, and the survival times of terminally ill patients. *Health Psychology, 24*, 225-229.
- **Authored Book:** Mitchell, T. R., & Larson, J. R., Jr. (1987). *People in organizations: An introduction to organizational behavior* (3rd ed.). New York, NY: McGraw-Hill.
- **Chapter in an Edited Book:** Bjork, R. A. (1989). Retrieval inhibition as an adaptive mechanism in human memory. In H. L. Roediger III & F. I. M. Craik (Eds.), *Varieties of memory & consciousness* (pp. 309-330). Hillsdale, NJ: Erlbaum.

Figures

Graphics files are welcome if supplied as Tiff, EPS, or PowerPoint files. The minimum line weight for line art is 0.5 point for optimal printing.

When possible, please place symbol legends below the figure instead of to the side.

Original color figures can be printed in color at the editor's and publisher's discretion provided the author agrees to pay

- \$255 for one figure
- \$425 for two figures
- \$575 for three figures
- \$675 for four figures
- \$55 for each additional figure

Permissions

Authors of accepted papers must obtain and provide to the editor on final acceptance all

necessary permissions to reproduce in print and electronic form any copyrighted work, including, for example, test materials (or portions thereof) and photographs of people.
Download Permissions Alert Form (PDF, 47KB)

Publication Policies

APA policy prohibits an author from submitting the same manuscript for concurrent consideration by two or more publications.

See also APA Journals® Internet Posting Guidelines.

APA requires authors to reveal any possible conflict of interest in the conduct and reporting of research (e.g., financial interests in a test or procedure, funding by pharmaceutical companies for drug research).

Download Disclosure of Interests Form (PDF, 38KB)

Authors of accepted manuscripts are required to transfer the copyright to APA.

Download Publication Rights (Copyright Transfer) Form (PDF, 83KB)

Ethical Principles

It is a violation of APA Ethical Principles to publish "as original data, data that have been previously published" (Standard 8.13).

In addition, APA Ethical Principles specify that "after research results are published, psychologists do not withhold the data on which their conclusions are based from other competent professionals who seek to verify the substantive claims through reanalysis and who intend to use such data only for that purpose, provided that the confidentiality of the participants can be protected and unless legal rights concerning proprietary data preclude their release" (Standard 8.14).

APA expects authors to adhere to these standards. Specifically, APA expects authors to have their data available throughout the editorial review process and for at least 5 years after the date of publication.

Authors are required to state in writing that they have complied with APA ethical standards in the treatment of their sample, human or animal, or to describe the details of treatment.

Download Certification of Compliance With APA Ethical Principles Form (PDF, 26KB)

The APA Ethics Office provides the full Ethical Principles of Psychologists and Code of Conduct electronically on their website in HTML, PDF, and Word format. You may also request a copy by emailing or calling the APA Ethics Office (202-336-5930). You may also read "Ethical Principles," December 1992, *American Psychologist*, Vol. 47, pp. 1597-1611.

APPENDIX 2: Systematic Review Protocol

based on York University's Centre for Reviews and Dissemination
Guidance for undertaking reviews in healthcare (CRD, 2009)

Background

- Pharmacological treatment of PSD is effective, but has been found to put individuals at risk of adverse events (Hackett *et al.*, 2008b), therefore other approaches to treatment are required. This of course has implications for the use of medication in clinical practice, and alternative approaches such as psychotherapy, should ideally be considered
- Evidence to suggest that psychological therapy (particularly cognitive behavioural approaches) lends itself to the treatment of PSD (Bradbury *et al.*, 2008, Broomfield *et al.*, 2010, Fraser *et al.*, 2005, Lustman *et al.*, 1998, Mohr & Goodkin, 1999)
- Lack of research in the area due to the inherent problems of conducting psychological treatment intervention trials with brain-injury populations (Broomfield, *et al.*, 2010)
- Cochrane review focuses solely on high quality randomized-controlled trials
- Valuable information can be gleaned from uncontrolled trials and case series

Previous similar reviews:

- Cochrane review of interventions for treating depression after stroke (Hackett *et al.*, 2008b)
- Cochrane review of the prevention of depression after stroke (Hackett *et al.*, 2008a)
- Evaluation of psychological management of post-stroke depression (Kneebone & Dunmore, 2000)
- Effectiveness of psychological interventions in chronic stage of stroke: A systematic review of randomised controlled trials (Mehta *et al.*, 2012)
- A systematic review of therapeutic interventions for poststroke depression and the role of nurses (de Man-van Ginkel *et al.*, 2010)
- No systematic reviews conducted considering non-RCTs

Review questions

What are the critical issues to consider when designing research trials into the use of non-pharmacological treatments of PSD?

Eligibility criteria

- Published case study, case series, small n studies, quasi-experimental design studies, controlled study or non-controlled study
- English Language publication
- Full-text available
- All publication date ranges
- Intervention studies

Population

- Diagnosis of either ischaemic or hemorrhagic stroke, and post-stroke depression
- Adult populations

Outcomes

- Use of one or more of the depression measures validated for use with stroke populations (Lincoln *et al.*, 2012).

Planned search strategy

- Keyword searches of online databases (Medline, PsycINFO, CINAHL Plus, Psychology and Behavioural Sciences Collection and Embase), using MeSH search terms for stroke ('stroke', 'apoplexy', 'CVA', 'cerebrovascular accident', 'haemorrhage', 'hemorrhage', 'intracranial bleed', 'vascular accident', 'SAH', and 'subarachnoid') in combination with 'depressi* AND treatment', 'depressi* AND therapy', 'low mood AND treatment', and 'low mood AND therapy'
- Trials registers websites (www.ClinicalTrials.gov and www.who.int/trialsearch/) were searched to identify any further relevant studies which may have been in progress or in press
- Manual search of reference lists of papers selected for review
- Manual search of reference lists of the 5 similar previous reviews

Study selection

1. Titles screened for relevance
2. If titles relevant, abstracts reviewed to see if study meets eligibility criteria
3. Full-text of retained studies reviewed to see if meet eligibility criteria
4. Final selection of studies included in methodological appraisal and assessment

Data extraction

The following data was extracted regarding each paper:

- Research/study question
- Study design
- Population(s)/sample(s)
- Measures
- Intervention
- Main findings
- Limitations

Quality Assessment

- Specific criteria for each criteria domain
- Scoring categories of well covered; adequately addressed; poorly addressed; not addressed/not reported; not applicable
- Overall assessment of the study (Good $\geq 70\%$; Fair $\geq 50\%$; Weak $< 50\%$)

Data synthesis

- Summary of individual study findings and characteristics (data from standardised data extraction form)
- Overall rating and quality ratings for each of the dimensions identified
- Overall summary of state of the literature in this area
- Limitations of available literature
- Areas identified for future research

Dissemination

- Chapter in doctoral portfolio thesis
- Submit for publication and presentation

Appendix 3: Systematic Review Data Extraction Form

General information

Date of data extraction:

Author:

Article title:

Citation:

Country of origin:

Study characteristics

Aim/objectives of the study:

Study design:

Study inclusion criteria:

Study exclusion criteria:

Recruitment procedures used:

Participant characteristics

Age:

Gender:

Ethnicity:

Socio-economic status:

Stroke diagnosis:

Co-morbidities (e.g. other long-term physical health conditions):

Number of participants in sample:

Intervention and setting

Setting in which the intervention is delivered/research takes place:

Description of the intervention(s) and control(s) (if applicable):

Outcome data/results

Unit(s) of assessment or measure(s) used:

Statistical techniques used:

Measurement tool or method used (if applicable):

Length of follow-up, number and/or times of follow-up measurements:

For all intervention/experimental group(s) and control group(s):

Number of participants approached/asked to take part:

Number of participants enrolled/took part/returned responses:

Number of participants included in analysis:

Number of withdrawals, deaths, exclusions, lost to follow-up:

Number of participants in each group/condition:

Group/condition mean:

Type of analysis used in study (e.g. group mean comparisons; correlation, etc.):

Results of study analysis:

Details of any additional relevant outcomes reported:

Adverse events/effects:

Other notes:

Appendix 4: Quality Rating Tool for Systematic Review

based on the Scottish Intercollegiate Guidelines Network guidance on systematic literature reviews (SIGN, 2008) and York University's Centre for Reviews and Dissemination guidance for undertaking reviews in healthcare (CRD, 2009).

	Overview of Quality Criteria Domains	Domain numerical score
1	Research question and objectives	/
2	Sampling	/
3	Design and method	/
4	Statistical analysis	/
5	Quality of reporting	/
6	Generalisability	/
7	Overall assessment of study	/
	Total score	/
	Percentage (based on number of items rated as applicable)	
	Descriptive category (Good $\geq 70\%$; Fair $\geq 50\%$; Weak $< 50\%$)	

1. Research question and objectives

1.1 The study addresses an appropriate and clearly focused question, drawn from a theoretical model or previous research.

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

If the study is a randomised controlled trial:

1.2 The assignment of participants to treatment groups is randomised

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

1.3 An adequate concealment method is used

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

1.4 subjects and investigators are kept 'blind' about treatment

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

1.5 The treatment and control groups are similar at the start of treatment

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

1.6 The only difference between the groups is the treatment under investigation

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

1.7 All relevant outcomes are measured in a standard, valid, and reliable way

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

1.8 All the subjects are analysed in the groups to which they were randomly allocated (intention to treat analysis)

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

1.9 Where the study is carried out at multi-centre sites, results are comparable for all sites

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

2. Sampling

2.1 The characteristics of the participants are representative of the group being studied.

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

2.2 The study indicates how many of the people asked to take part did so, in each of the groups studied. Also, if applicable, how many dropped out.

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

3. Design and method

3.1 The constructs/variables under investigation are clearly defined and operationalised.

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

3.2 Variable measurement method is appropriate and demonstrates validity and reliability.

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

3.3 Confounding variables that may have influenced the results are taken into account.

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

4. Statistical analysis

4.1 Statistical analyses are fully reported and appropriate (including confidence intervals where appropriate).

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

5. Quality of reporting

5.1 Reporting of method, analyses and results are sufficiently detailed to allow their replication or justification.

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

6. Generalisability

6.1 The findings could be generalised to similar populations.

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

7. Overall assessment of study

7.1 A judgement of the overall quality of the study.

++ Good to excellent = 3	
+ Adequate to good = 2	
- Poor to adequate = 1	

APPENDIX 5: Participant Information Sheet



Primary and Community Division Lothian MCN Stroke Clinical Neuropsychology

Astley Ainslie Hospital
Department of Clinical Psychology
133 Grange Loan
Edinburgh EH9 2HL
Telephone 0131 537 9000
Direct Dial **0131 537 9140**
Fax **0131 537 9120**

Participant Information Sheet

Post-stroke Emotional Lability: Interviews

Information for Participants

We would like to invite you to take part in some interviews as part of a research study. The topic of the current research is the conceptualisations of a condition called post-stroke emotional lability (PSEL). This is part of a larger research project by the Lothian Stroke Managed Clinical Network on the treatment of PSEL.

Before you make a decision about taking part we would like you to understand why the research is being undertaken and what it would involve for you. Please ask us if anything is unclear.

Part 1

What is the purpose of the study?

The Lothian Stroke MCN is currently conducting some research on the treatment of PSEL. There has been little research done on staff's conceptualisations and experiences of

PSEL, and this research seeks to find out what people think about it and what their clinical experience is of the condition within specialist stroke services.

What would be your role?

You are being invited to take part because you are a professional working in a stroke unit with individuals who have PSEL.

Why have I been invited?

We would like to conduct individual interviews (some in Edinburgh and some in Glasgow) with stroke unit professionals from various disciplines.

What will the study involve?

Each participant will meet with the researcher on an individual basis on a stroke inpatient unit. You will only need to meet with the researcher once, and the interview will last between 45-60mins. The main question that will be asked is: “what does PSEL mean to you?” Some other questions may also be asked about PSEL and your experience of it in your clinical work.

The interview will be recorded digitally and then transcribed by the researcher. Your name will not be identified on the transcript (i.e. the transcript will refer to “Occupational Therapist”, or “Nurse”, not the person’s name). The information that is obtained from the interviews will be used to help us better understand specialist staff’s conceptualisations and experiences of PSEL. While extracts from the interviews will be used in the analysis these will not be attributed to any professional in the final report.

Do I have to take part?

No, you do not have to take part. Participation in the study is entirely voluntary. Unfortunately we are not able to give any form of payment for participating.

If you decide to take part, you will be asked to sign a consent form and will have an opportunity to ask questions. You can change your mind at any time and ask for the information you provided to be destroyed and not to be used in the study.

What are the benefits of taking part?

In order for us to further investigate the approaches to treating PSEL, it is important that we also know more about staff’s experiences and conceptualisations of it. This will help us gauge the need for possible training and support for staff, as well as being able to

further inform treatment guidelines and effectively manage the condition within specialist services. This research hopes to investigate specialist stroke staff's experiences of PSEL, and how they conceptualise and understand it, with the overall aim of developing services and clinical practice guidelines.

What are the disadvantages of taking part?

We do not envisage any significant risks or disadvantages to you from taking part in the research.

As with all research related to experiences of working with vulnerable people who are unwell, there is a slight possibility that interviews may result in some emotional upset for participants. If at any point you do feel upset during the interview, the researcher will support you appropriately at this point and the interview can be paused or stopped. Should you continue to feel upset after the interview, you can discuss how you feel with a trusted colleague or supervisor. You can also contact the researcher to discuss any concerns after the interview if you wish.

If the information in Part 1 has interested you and you are thinking about taking part, please read the rest of this sheet before making a decision.

Part 2: Additional information

What happens to the information?

All information will be kept confidential and stored securely under the protection of the Principal Investigator. All data will be coded so your name will not appear on any written record or on any computer database. Data may be looked at by authorised people to check that the study is being carried out correctly, and all will have a duty of confidentiality to uphold.

The results will be written up as a DCLinPsychol Thesis, and this thesis may be published in a peer reviewed journal. All the information will be kept confidential and no names will be included in the final report of the study. We will share the results of the research with you should you wish.

What if there is a problem?

If you have a concern about any aspect of the research, you can ask to speak to the researchers, who will do their best to answer your questions - the Principal Investigator's details can be found below. If you remain unhappy and wish to complain formally, you can do this using the normal National Health Service complaints mechanisms. The Principal Investigator can give you details of these. You can also make a complaint to the academic project supervisors, Dr Ethel Quayle and Dr Kenneth Laidlaw, University of Edinburgh.

In the unlikely event that you are harmed by taking part in this research study, and this is due to someone's negligence, then you may have grounds for a legal action against NHS Lothian, but you may have to pay your legal costs.

Who has reviewed the study?

All research in the NHS is looked at by an independent group called a Research Ethics Committee, to protect your interests. This study has been given a favourable opinion by the University of Edinburgh Research Ethics Committee, NHS Lothian Quality Improvement Team, and Greater Glasgow and Clyde Research and Development Department.

If you would like further information, please contact the Principal Investigator: Hannah Picton, Trainee Clinical Psychologist, Department of Neuropsychology, Astley Ainslie Hospital, Edinburgh, EH9 2HL (tel: 0131 537 9149, email: Hannah.Picton@nhslothian.scot.nhs.uk). You can also contact Dr David Gillespie, Consultant Clinical Neuropsychologist and Clinical Supervisor for the project on tel: 0131 537 9140 or email: David.Gillespie@nhslothian.scot.nhs.uk.

APPENDIX 6: Interview Questions



Interview Schedule

Post-stroke Emotional Lability: Interviews

1. What does PSEL mean to you: What is it? What causes it? How important is it for us to know about the cause/aetiology of PSEL?
2. How do we identify PSEL?
3. How do we distinguish PSEL from post-stroke depression?
4. Can you give me an example of when you have encountered someone with PSEL? What did you do/not do?
5. Do you notice any difference in the presentation of PSEL depending on the location of the lesion (particularly left and right lesions)? If so, what do you see? Can you give an example?
6. What does PSEL look like in individuals with aphasia?

APPENDIX 7: Consent Form



Consent Form

Post Stroke Emotional Lability: Interviews

	Yes	No
I have been given information about the study.		
I have had a chance to ask questions about the study.		
My questions have been answered.		
I understand what is involved.		
I understand my name will not be used in written reports.		
I agree to take part in the interview.		
I understand I can leave the interview at any time.		
I understand I can ask, at any time, for my interview not to be used.		
I agree for my interview to be recorded.		

Name:.....

Profession:.....

Signature:

Date:

Signed (researcher):

Date:

APPENDIX 8: Supplementary Participant Information



Supplementary participant information

Post Stroke Emotional Lability: Interviews

Profession	
Qualifications	
Number of years working in stroke	
Age	
Approximate number of patients seen per month with emotional lability	
Experience in other Neuro services. What service(s)? How long?	

APPENDIX 9: Ethical Approval



SCHOOL of HEALTH & SOCIAL SCIENCE
CLINICAL PSYCHOLOGY

The University of Edinburgh
Medical School
Dootway 6, Teviot Place
Edinburgh EH8 9AG

Telephone 0131 651 3969
Fax 0131 650 3891
Email submitting.ethics@ed.ac.uk

Hannah Picton
59 (2F1) Jeffrey Street
Edinburgh
EH1 1DH

02 April 2013

Dear Hannah,

Re: The conceptualisation of post-stroke emotionalism: A qualitative investigation with stroke specialist staff

Application for Level 1 Approval

Thank you for submitting the above research project for review by the Section of Clinical Psychology Ethics Research Panel. I can confirm that the submission has been independently reviewed and was approved on the 13th December 2012.

Should there be any change to the research protocol it is important that you alert us to this as this may necessitate further review.

Yours sincerely,

Kirsty Gardner
Secretary
Clinical Psychology

APPENDIX 10: R&D Amendment Approval

University Hospitals Division

Queen's Medical Research Institute
47 Little France Crescent, Edinburgh, EH16 4TJ

KM/SS

21 January 2013

Dr David Gillespie
NHS Lothian
Department of Clinical Psychology
Astley Ainslie Hospital
Edinburgh
EH9 2HL



RESEARCH &
DEVELOPMENT
Room E1.12
Tel: 0131 242 3330
Fax: 0131 242 3343
Email:
R&DOffice@luht.scot.nhs.uk

Director:
Professor David E Newby

Dear Dr Gillespie

REC No: N/A
R&D Project ID No: 2012/P/REH/03
Amendment: Substantial amendment No.1 dated 09 January 2013
Title of Research An investigation of non-pharmacological treatment approaches used by stroke professionals for post-stroke emotional lability

I am writing in reply to recent correspondence in relation to an amendment(s) to the above project and the subsequent updated documents as follows.

- o Information Sheet PSEL Version 3 dated 10 December 2012
- o Consent Form PSEL Interviews
- o CV – Miss Hannah Picton
- o Research Protocol Version 2 dated 07 January 2013

We have now assessed any consequential changes and can confirm that NHS Lothian management approval is extended to cover the specific changes intimated.

Yours sincerely


Mrs Karen Maitland
Research Governance Manager

cc: Hannah Picton, Trainee Clinical Psychologist

APPENDIX 11: NHS-to-NHS Access for Research Approval



Coordinator/Administrator: Michael Barber/Elaine O'Neill
Telephone Number: 0141 211 6208
E-Mail: michael.barber@ggc.scot.nhs.uk
Website: www.nhsggc.org.uk/r&d

R&D Management Office
Western Infirmary
Tennent Building
1st Floor, 38 Church Street
Glasgow, G11 6NT.

19 February 2013

Miss Hannah Picton
Trainee Clinical Psychologist
Dept of Neuropsychology
Astley Ainslie Hospital
113 Grange Loan
Edinburgh EH9 2HL

Dear Miss Picton,

NHS to NHS - Letter of Access for Research

As an existing NHS employee you do not require an additional honorary research contract with this NHS organisation. We are satisfied that the research activities that you will undertake in this NHS organisation are commensurate with the activities you undertake for your employer. Your employer is fully responsible for ensuring such checks as are necessary have been carried out. Your employer has confirmed in writing to this NHS organisation that the necessary pre-engagement checks are in place in accordance with the role you plan to carry out in this organisation. This letter confirms your right of access to conduct research through **NHS Greater Glasgow and Clyde** for the purpose and on the terms and conditions set out below. This right of access commences on **19th February 2013** and ends on **14th July 2013** unless terminated earlier in accordance with the clauses below.

You have a right of access to conduct such research as confirmed in writing in the letter of permission for research from this NHS organisation. Please note that you cannot start the research until the Principal Investigator for the research project has received a letter from us giving permission to conduct the project.

You are considered to be a legal visitor to **NHS Greater Glasgow and Clyde** premises. You are not entitled to any form of payment or access to other benefits provided by this organisation to employees and this letter does not give rise to any other relationship between you and this NHS organisation, in particular that of an employee.

While undertaking research through **NHS Greater Glasgow and Clyde** you will remain accountable to your employer **NHS Lothian** but you are required to follow the reasonable instructions of your nominated manager **Dr Niall Broomfield** in this NHS organisation or those given on her/his behalf in relation to the terms of this right of access.

Where any third party claim is made, whether or not legal proceedings are issued, arising out of or in connection with your right of access, you are required to co-operate fully with any investigation by this NHS organisation in connection with any such claim and to give all such assistance as may reasonably be required regarding the conduct of any legal proceedings.

You must act in accordance with **NHS Greater Glasgow and Clyde** policies and procedures, which are available to you upon request, and the Research Governance Framework.

You are required to co-operate with **NHS Greater Glasgow and Clyde** in discharging its duties under the Health and Safety at Work etc Act 1974 and other health and safety legislation and to take reasonable care for

the health and safety of yourself and others while on **NHS Greater Glasgow and Clyde** premises. Although you are not a contract holder, you must observe the same standards of care and propriety in dealing with patients, staff, visitors, equipment and premises as is expected of a contract holder and you must act appropriately, responsibly and professionally at all times.

If you have a physical or mental health condition or disability which may affect your research role and which might require special adjustments to your role, if you have not already done so, you must notify your employer and the Board via the **HR Department** prior to commencing your research role at the Board.

You are required to ensure that all information regarding patients or staff remains secure and *strictly confidential* at all times. You must ensure that you understand and comply with the requirements of the NHS Confidentiality Code of Practice (<http://www.dh.gov.uk/assetRoot/04/06/92/54/04069254.pdf>) and the Data Protection Act 1998. Furthermore you should be aware that under the Act, unauthorised disclosure of information is an offence and such disclosures may lead to prosecution.

NHS Greater Glasgow and Clyde will not indemnify you against any liability incurred as a result of any breach of confidentiality or breach of the Data Protection Act 1998. Any breach of the Data Protection Act 1998 may result in legal action against you and/or your substantive employer.

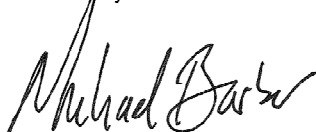
You should ensure that, where you are issued with an identity or security card, a bleep number, email or library account, keys or protective clothing, these are returned upon termination of this arrangement. Please also ensure that while on the premises you wear your ID badge at all times, or are able to prove your identity if challenged. Please note that this NHS organisation accepts no responsibility for damage to or loss of personal property.

We may terminate your right to attend at any time either by giving seven days' written notice to you or immediately without any notice if you are in breach of any of the terms or conditions described in this letter or if you commit any act that we reasonably consider to amount to serious misconduct or to be disruptive and/or prejudicial to the interests and/or business of this NHS organisation or if you are convicted of any criminal offence. You must not undertake regulated activity if you are barred from such work. If you are barred from working with adults or children this letter of access is immediately terminated. Your employer will immediately withdraw you from undertaking this or any other regulated activity and you **MUST** stop undertaking any regulated activity immediately.

Your substantive employer is responsible for your conduct during this research project and may in the circumstances described above instigate disciplinary action against you.

If your circumstances change in relation to your health, criminal record, professional registration or suitability to work with adults or children, or any other aspect that may impact on your suitability to conduct research, or your role in research changes, you must inform the NHS organisation that employs you through its normal procedures. You must also inform your nominated manager in this NHS organisation.

Yours sincerely



Dr Michael Barber
Research Co-ordinator

cc: NHS Lothian HR Dept