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**An investigation of the lived experiences and illness
perceptions of adults with sudden onset neurological
conditions.**

Niamh McAleese

Doctorate in Clinical Psychology

University of Edinburgh

May 2017

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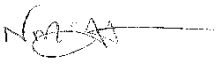
D.Clin.Psychol. Declaration of Own Work

Name: Niamh McAleese

Title of Work: An investigation of patient beliefs and illness perceptions in adults with sudden onset neurological conditions.

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

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Lastly, I would like to extend my gratitude and appreciation to the participants who gave their time to share their experiences. I dedicate this thesis to them.

Portfolio thesis abstract

Purpose: The systematic review summarised the literature on the impact of patient illness perceptions on health outcomes and coping after an acute neurological event, guided by Leventhal's Self-Regulatory Model (SRM). The empirical study investigated individuals' lived experiences of emotionalism, a sudden onset neurological disorder characterised by involuntary laughter and crying. A further aim was to develop a questionnaire measuring beliefs about emotionalism based on patients' perspectives.

Method: The review identified seventeen articles through database searches using predefined inclusion criteria. In the empirical paper, eighteen individuals took part in a qualitative study to explore their experiences of emotionalism.

Results: Findings provided support for the SRM in acute neurological populations. Negative illness perceptions were associated with a range of poor health outcomes and unhelpful coping behaviours. The empirical paper provided rich individual accounts of the social and personal impact of emotionalism. Four themes were identified and used to develop a questionnaire measuring beliefs about emotionalism.

Conclusions: Both chapters emphasise the value of eliciting patient beliefs about their neurological condition and of providing support at the early stages of recovery. The clinical implications and directions for future research were discussed as was the need for further validation of the questionnaire.

Chapter 1: Systematic review

A systematic review of illness perceptions, coping and health outcomes in adults with sudden onset neurological conditions.

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Word count: 8,024

Abstract

Purpose: Sudden onset neurological conditions (e.g. stroke, traumatic brain injury, neuro-infection) are associated with significant and potentially life-limiting health consequences. There is a need to better understand the psychological determinants of illness outcomes and coping in these populations. According to Leventhal's Self-Regulatory model, patients' illness perceptions regulate their emotional responses and health-behaviours. The aim of this study was to review the literature related to the relationships between individuals' illness perceptions, coping behaviours and health outcomes after a sudden neurological event.

Method: A systematic review of the published literature was undertaken. Key electronic databases were searched from January 1995 to January 2017. The references of included articles were manually searched and key authors were contacted for additional publications.

Results: Sixteen studies were of sufficient quality to be included in the review. A synthesis of the results found that beliefs about multiple symptoms with lasting serious consequences, low levels of control and a lack of understanding about the causes of injury were associated with poor health outcomes and coping behaviours.

Conclusions: The results of this review provide support for applying the Self-Regulatory Model to sudden onset neurological populations. Implications for rehabilitation, research directions and clinical practice are discussed.

Keywords: Illness perceptions, brain injury, stroke, subarachnoid haemorrhage, systematic review.

Introduction

Sudden onset neurological conditions account for 15-20% of acute medical admissions [1]. The term refers to damage caused to the brain as a consequence of traumatic brain injury, neuro-infection or disruptions to the cerebrovascular system. Sudden neurological injuries pose serious personal and public health concerns. Stroke is the leading cause of long-term disability [2]. Every year, an estimated 1 million people sustain a traumatic head injury in the UK [3]. Neuro-infections are prevalent and associated with high levels of morbidity and mortality [4].

For some individuals, the impact of a neurological injury is pervasive and long lasting. Up to 30% of those with a mild traumatic brain injury develop post-concussion syndrome (PCS), defined by persistent physical symptoms like dizziness and headaches [5]. PCS causes heightened social and functional disability, as well as increased pressure on public health services [6,7]. Failure to return to work is a frequent occurrence after stroke and associated with reduced life satisfaction, social isolation, maladaptive coping and clinical mood disorders [8,9,10]. The latter is a common after neurological injury. One in three people who sustain a stroke develop depression within 12 months of the injury [11] and 37% of individuals with a subarachnoid haemorrhage go on to develop post-traumatic stress disorder [12,13]. Kreutzer *et al.* [14] found that 42% of patients with a traumatic brain injury met DSM-IV criteria for depression.

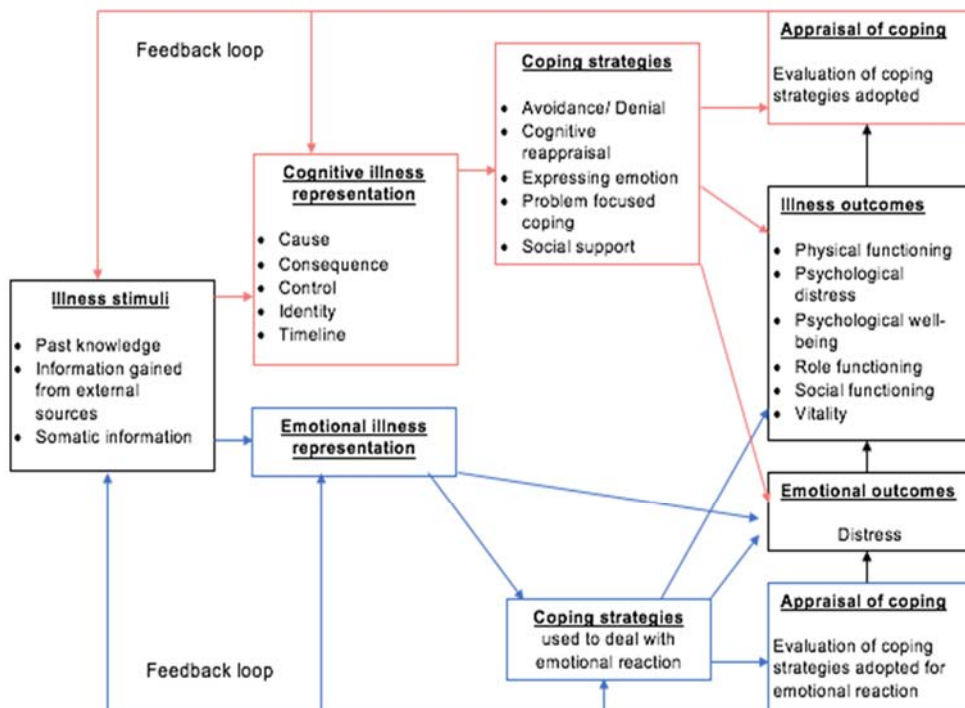
Clearly, sudden onset neurological conditions place significant burden on the individual. Yet injury severity does not fully account for the variance in psychosocial outcomes or quality of life within these populations [15]. Indeed, many individuals fail to resume pre-injury social and functional roles despite being physically well and cognitively able [16]. Recently, research has begun to establish links between psychological constructs such as patients' beliefs and recovery trajectories after sudden neurological injury [17].

An influential theory of patient beliefs and health behaviour is that of Leventhal and colleagues' Self-Regulatory Model (SRM) [18]. The model proposes that once diagnosed with a health threat, the person develops an organised set of cognitive beliefs and emotional responses about the condition. These patterns of beliefs (known as 'illness perceptions') are used to guide the person's emotional responses and coping behaviours to manage the health condition [19]. Leventhal *et al.* [20] described five inter-related components of the model: (1) beliefs about how long the illness will last (*timeline*); (2) understanding the cause of the condition (*cause*); (3) expectations about the impact of symptoms on their life (*consequences*); (4) identifying symptoms that belong to the condition (*identity*) and (5) beliefs about controlling the illness through personal or treatment means (*control*). Morris-Morris [21] added two further dimensions to the original model; understanding of the condition and associated symptoms (*coherence*) and emotional reactions (*emotional representation*).

Illness perceptions are thought to arise from three sources of information; the individual's socio-cultural knowledge of illness, somatic information and their

past illness experience [22]. According to Leventhal *et al.* [18] the person engages in a dynamic process of making sense of their illness by continuously evaluating the effectiveness of their coping strategies with respect to their health outcomes (see *Figure 1.1.*).

Figure 1. Schematic representation of Leventhal's (1980) Self-Regulatory Model of illness perceptions adapted from Hagger and Orbell (2003).



In 1996, Weinman *et al.* developed the Illness Perception Questionnaire (IPQ) based on the SRM to formally assess illness perceptions in clinical settings [23]. A further revised version of the scale (IPQ-R) included two additional dimensions, *coherence* and *emotional representation* [21]. The length of the original scale motivated Broadbent and colleagues to create an abbreviated version to accommodate shorter assessments [24]. The psychometric validity and reliability of the three measures has been tested across a wide range of health conditions [25, 26]. Authors of the scales recommend that researchers

modify items on the questionnaire to suit the characteristics of the health condition of interest [23].

Since the inception of the IPQ, a large body of research has shown relationships between patient illness perceptions and health outcomes [27,28]. Hagger and Orbell summarised these associations in a comprehensive meta-analysis [26]. The authors used content analysis to examine the existing health literature and group health outcomes and coping strategies into categories (see *Figure 1.1.*). Health outcomes were classified as; physical functioning, psychological distress and well-being, role functioning, social functioning and vitality. Coping strategies were classified as; avoidance/denial, cognitive re-appraisal, expressing emotions, problem focused coping and using social support. The results of Hagger and Orbell's review suggested that people who endorsed negative perceptions about their illness engaged in unhelpful ways of coping (i.e. rumination or avoidance) and reported poorer health outcomes. Conversely, positive illness perceptions were predictive of improved health outcomes such as vitality, well-being and role functioning across a range of health populations.

Since Hagger & Orbell's review, many studies have shown that illness perceptions are important factors in predicting medical (e.g. pain severity), psychological (e.g. quality of life, depression or anxiety) and functional (e.g. return to work) outcomes across a range of health conditions [29,30,31]. To date, research into illness perceptions has focused on chronic illness [32,33].

It is not known whether the same psychological processes apply to those who suffer an acute neurological condition.

Rationale for Review

The onset of a neurological injury is immediate and traumatic. The fatalistic nature of the event forces the person to consider their mortality in a sudden way [34]. Yet, the psychological mechanisms that the person employs to make sense of their condition are largely unknown. The early stages of neurological injury involve high levels of distress, uncertainty and an influx of information about disease course and prognosis, at a time of maximum physical or cognitive impairment. All of these factors will likely affect the formation of patient beliefs. Given that illness perceptions are important determinants of outcomes and coping in people with chronic illness, investigating illness perceptions and health outcomes in acute neurological populations is indicated. Greater clarity about these relationships could inform health care interventions and build on emerging research.

Aims and Objectives

The study aimed to provide a systematic overview of the literature exploring relationships between illness perceptions, health outcomes and coping in those with a sudden onset neurological condition. The review used Hagger and Orbell's categories to classify health outcomes and coping [26]. The review hypothesised that individuals who endorsed stronger beliefs regarding

the severity of consequences, longevity of their neurological condition and lower levels of perceived control would demonstrate poorer coping abilities and more negative health outcomes.

The study addressed the research objective with reference to participants, interventions, comparators and outcomes (PICO) as recommended by the Centre for Reviews and Dissemination [35].

Method

Inclusion criteria

Studies were included in the review if they met the following inclusion criteria:

1. Observational studies with quantitative design.
2. Studies conducted in an adult population with a diagnosed sudden onset neurological condition (i.e. stroke, brain injury or infection).
3. Studies including any version of the IPQ.
4. At least one measure of coping or health outcome as defined by Orbell and Hagger. These were physical functioning, psychological distress or well-being, role functioning, social functioning, vitality, avoidance/denial, cognitive re-appraisal, expressing emotions, problem-focused coping or social support [26].
5. Full text papers published in the English language.
6. Studies conducted after 1995 (the date of development of the IPQ).

Studies that recruited sudden onset neurological populations with other acute medical populations were included if the majority of the study sample comprised of those with an acute injury (as per Cochrane guidance) [36].

Literature search strategies

A systematic literature review was conducted by searching the following electronic databases for articles published between January 1995 to 2017:

PsychINFO, Web of Science, EMBASE, MEDLINE, Cochrane Library and CINAHL. Grey literature was searched using the following engines; Google Scholar, OpenGrey and the British Library Electronic Theses Online System (EThOS). The search terms were developed based on terms relevant to previously published studies in the field and expert opinion (see *Figure 2*). Citation searches of the included articles and a manual search of their reference lists was conducted to locate additional papers. The lead authors of the included studies were contacted to request any studies in preparation for publication to minimise bias. See the study selection guidance procedure (*Appendix A*) and the study protocol (*Appendix B*) for further details. A review protocol was published online to provide transparency in the systematic review process [37].

Figure 2. Boolean search terms

| |
|--|
| <p>("illness perception*" OR "self- regulation" OR "patient attitude*" OR "illness cognition" OR "health belief*" OR "common sense" OR "illness representation" OR IPQ)</p> <p style="text-align: center;">AND</p> <p>("head injur*" OR "brain injur*" OR "brain infection" OR stroke OR CVA OR "cerebrovascular accident" OR "subarachnoid hemorrhag*" OR "subarachnoid haemorrhag*" OR ischemi* OR TBI OR ABI OR concussion)</p> <p style="text-align: center;">NOT</p> <p>("myocardial infarct*")</p> |
|--|

Data extraction

One researcher read titles and abstracts of studies identified by database searches. A data extraction form (see *Appendix C*) was used by two reviewers to independently assess the abstracts of full text articles for their relevance to

the review. Through consensus, 16 articles were included for the final narrative synthesis. Four studies were merged into two as the same dataset was used by the authors. The reviewers were not blinded to authors or institution. The first authors of the included studies were contacted if information was unavailable from the published article, or if something was unclear from the published account. Three authors replied to provide supplemental information (*see Results*).

Quality assessment

Evaluating the methodological quality of studies and their susceptibility to bias determines the strength and the generalisability of review findings. The *Cochrane Collaboration* recommends the use of formal quality assessment tools within non-randomised control studies [36,38]. The Newcastle-Ottawa Scale (NOS) is one of the most frequently used tools for non-randomised or observational studies [39]. It has acceptable face validity and criterion validity and the original authors of are currently investigating its construct validity [41].

Studies included in the review were rated based on their methods used to recruit a representative sample (selection), to control for confounding variables (comparability) and to assess the outcomes of interest (assessment of outcome) [40]. One point (in the form of a star) was awarded for each criterion achieved (*see Appendix D*). Higher ratings were associated with better quality studies.

The Centre for Reviews and Dissemination suggest that the tool of choice be guided by the review question, study design and the ability to detect bias [35]. The NOS was adapted for the current review by adding two questions to examine each study's fidelity to the use of and reporting of the IPQ. The quality criteria were operationalised as follows:

Selection / Representativeness

1. Was the recruited sample representative of adults with a sudden onset neurological condition?
2. Was the sample size adequate and justified?
3. Was comparability between participants and non-respondents established and were the response rates satisfactory?
4. Was the health outcome or coping style measured with a validated tool?
5. Was the IPQ administered as recommended by the developers (i.e. adapted for the target population and the use of a test reliability analysis on each subscale)?

Comparability

6. Did the study control for injury variables (e.g. injury severity)?
7. Did the study control for demographic factors (e.g. age, gender)?

Assessment of outcome

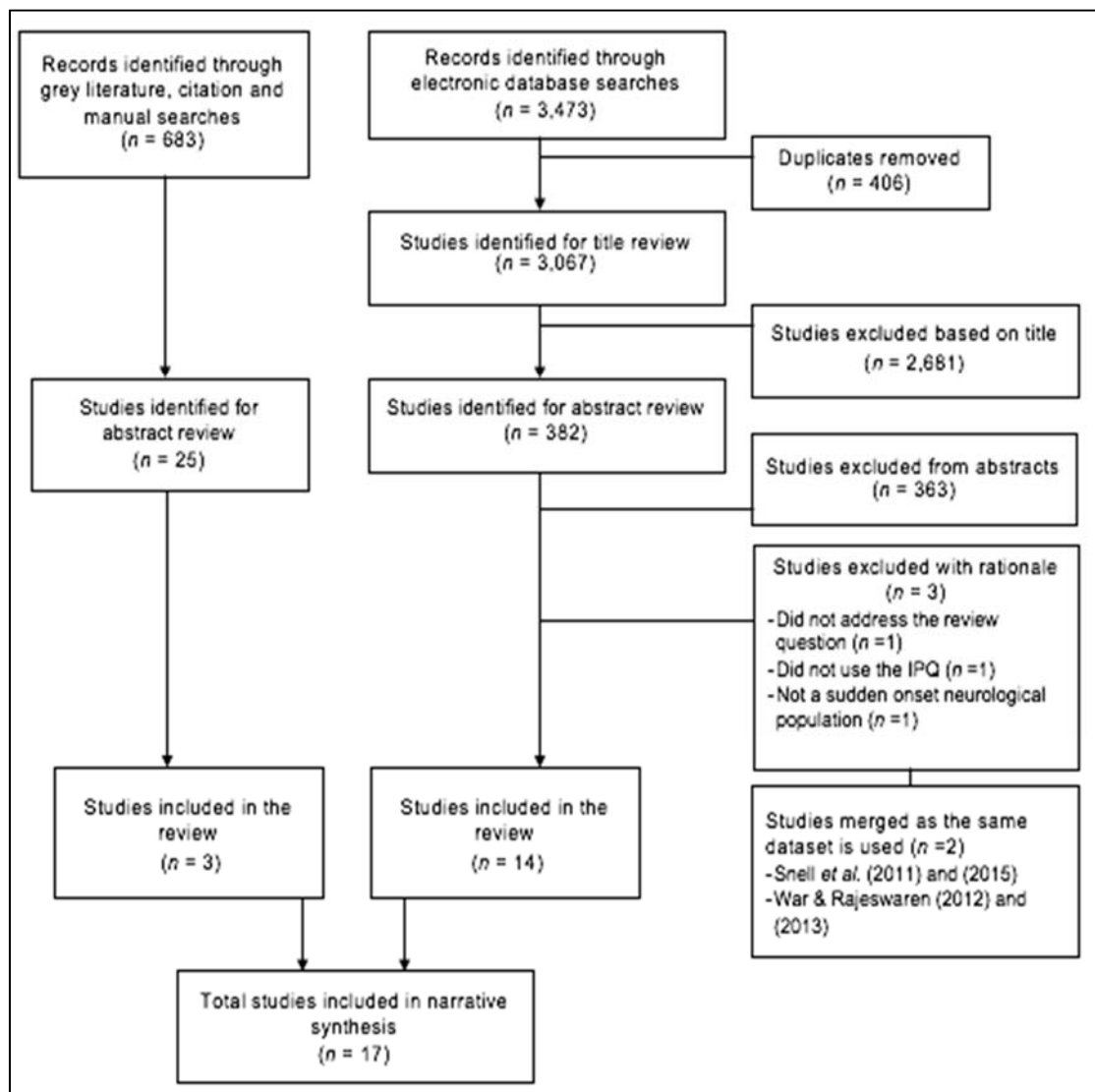
8. Were the statistical tests clearly described and appropriate?
9. Was the assessment of outcome confirmed by secure clinical records?
10. Were all of the dimensions of the IPQ reported?

SIGN 50 guidelines recommend the inclusion of a second reviewer when assessing the quality of studies to minimise bias [42]. Inter-rater reliability was found to be 93% on the total quality score of a random selection of half of the included papers. Two independent raters resolved discrepancies through discussion.

Results

The search procedure identified 4,156 articles from manual and electronic database searches. Duplicates were removed and the titles of 3,750 studies were screened. This process left 407 studies for abstract review where 388 studies were omitted for failing to meet the inclusion criteria. One additional study was included from searching the reference lists of the included articles. Two studies were merged for using the same dataset [15, 50].

Figure 3. PRISMA study selection procedure



Characteristics of the included studies

Traumatic brain injury was the most common condition identified ($n=8$), followed by stroke ($n=6$) then subarachnoid haemorrhage ($n=2$). One study included a heterogeneous population of neurological conditions including traumatic brain injury, stroke, hypoxia, tumour and encephalitis. Six of the eight TBI studies classified the sample as having a mild traumatic brain injury [15,43,44,46,50,53].

The majority of studies ($n=14$) reported the time since injury to be between 1 to 12 weeks. However, three studies described post-injury mean times of 70.43, 71.1 and 21.8 months [47,57, 54]. Two studies did not report on the time since injury [51,55]. Sample sizes ranged from 27 to 578 ($M=118$ $SD=131.7$). The follow-up period for participants in the longitudinal studies was at either 3 or at 6 months' post-injury [43,44,46,48,52,53,57]. Participants were predominantly male, Caucasian and English speaking.

No studies had a control group but three used a comparison population [44,45,48]. Sheldrick TARRIER, Berry and Kincey compared illness perceptions between those with myocardial infarction and subarachnoid haemorrhage [48]. Saltapidas and Ponsford examined two cultural populations [45] and Jones *et al.* compared the illness perceptions of those who completed brain drawings to those who did not [44]. The latter was a novel method of recording illness perceptions by asking the patient to draw a picture of their brain before and after their traumatic brain injury. See *Table 1* for a summary of the key findings.

Table 1. Data extraction and study characteristics table.

| Author | Population | Sample | Gender | Age | Design | IPQ Type | Comparison | Outcome of interest | Outcome |
|---|------------|----------------|--------------------|-------------------------------------|----------------------------|-------------------------|-------------------------------------|--|---|
| Snell <i>et al.</i> ¹⁵ (2011) (2015) | Mild TBI | <i>n</i> = 147 | M:44% F:56% | <i>M</i> =41.8 <i>SD</i> =15.7 | Cross sectional | IPQ-R | Brief COPE RPQ RHIFUQ HADS | Coping behaviour Physical functioning Social functioning Distress | Stronger beliefs about the nature and consequences of TBI were linked to greater distress, poor social and functional outcomes at 3 months. Participants were clustered into 3 groups based on perceptions. |
| Snell <i>et al.</i> ⁴³ (2013) | Mild TBI | <i>n</i> = 125 | M:42.4% F:57.6% | <i>M</i> =43.6 <i>SD</i> =15.8 | Longitudinal (6 months) | IPQ-R | RPC Brief COPE HADS | Physical functioning Coping behaviour Distress | Stronger injury identity beliefs, expectations of severe consequences and distress at time one predicted poor physical outcomes at time two. |
| Jones <i>et al.</i> ⁴⁴ (2016) | Mild TBI | <i>n</i> = 245 | M: 60% F:40% | <i>M</i> =37.38 <i>SD</i> =17.32 | Longitudinal (6 months) | B-IPQ Brain drawings | SF-36 RPQ | Quality of life Physical functioning | Negative Illness perceptions at 1 month predicted PCS and poorer quality of life at 6 months. |
| Saltapidas & Ponsford ⁴⁵ (2008) | TBI | <i>n</i> = 70 | M:58% F: 42% | <i>M</i> =39.06 <i>SD</i> =14.85 | Cross sectional | IPQ-R | CHART | Physical & social functioning | Poor understanding of TBI and greater emotional representations were associated with unemployment, poor social & functional outcomes. |

| Author | Population | Sample | Gender | Age | Design | IPQ Type | Comparison | Outcome of interest | Outcome |
|---|--|----------------|--------------------|-------------------------------------|-------------------------|--------------|---|--|---|
| Harris, ⁹ (2014) | aSAH | <i>n</i> = 134 | M:28.3% F:71.7% | <i>M</i> =52.2 <i>SD</i> =8.8 | Cross sectional | B-IPQ | FSQ | Role functioning (return to work) | Negative Illness perceptions were associated with greater levels of unemployment 1-2 years post injury. |
| Hou <i>et al.</i> ⁴⁶ (2012) | Mild TBI | <i>n</i> = 107 | M:62% F:37% | <i>M</i> =40.43 <i>SD</i> =15.44 | Longitudinal (6 months) | B-IPQ | BRIQ HADS SSQ RPQ | Distress Social functioning Physical functioning | Negative illness perceptions were the most important predictor of post-concussive syndrome at 6 months. |
| Rogan, Fortune & Prentice ⁴⁷ (2013) | TBI, CVA, Tumour, abscess, hypoxia, encephalitis | <i>n</i> = 70 | M: 70% F:30% | 19-65 yrs. <i>SD</i> =12 | Cross sectional | IPQ-R | PTGI FIM + FAM Brief COPE HADS | Physical functioning Coping behaviour Distress | Attributing the cause to psychological factors was correlated with distress. Stronger beliefs about treatment control linked post-traumatic growth. |
| Sheldrick, Tarrier, Berry & Kinsey ⁴⁸ (2006) | SAH | <i>n</i> = 27 | Not stated | Not stated | Longitudinal (3 months) | IPQ-R | DTS | Distress | High emotional representation, poor understanding of SAH treatment and consequences were predictive of developing PTSD. |
| Sjölander, Eriksson & Glader ⁴⁹ (2013) | CVA | <i>n</i> = 578 | M: 60% F: 40% | <i>M</i> =70.1 | Cross sectional | BIPQ Adapted | BMQ MARS | Physical functioning | Treatment control was associated with beliefs about medicines but not directly associated with medication adherence. |

| Author | Population | Sample | Gender | Age | Design | IPQ Type | Comparison | Outcome of interest | Outcome |
|--|-------------------|------------------------------|---------------------------------|--|-------------------------|----------|--|--|---|
| War & Rajeswaran (2012) ⁵⁰ (2013) | Mild-moderate TBI | <i>n</i> =31 <i>n</i> =30 | M:100% F: 0% | <i>M</i> =38.13 <i>SD</i> =8.82 | Cross sectional | B-IPQ | DAS-21 RPQ WHOQOL-BREF DAS-21 | Distress Physical functioning Psychological well-being | Negative beliefs about consequences, low personal or treatment control and high emotional representation were correlated with PCS and lower quality of life. |
| Nogueira & Teixeira ⁵¹ (2011) | CVA | <i>n</i> = 50 | M: 50% F: 50% | <i>M</i> = 59.6 <i>M</i> = 53.4 | Cross sectional | IPQ-R | WCPS VAS BDI | Coping behaviour Physical functioning Distress | Illness perceptions influenced coping styles in response to central pain in stroke. |
| Twiddy, House & Jones ⁵² (2012) | CVA & carers | <i>n</i> = 42 dyads | M: 57% F: 43% (CVA group) | <i>M</i> = 65.12 <i>SD</i> =10.2 (CVA group) | Longitudinal (3 months) | IPQ-R | GHQ-28 SOS | Distress Role functioning | Stronger illness identity, beliefs about more serious consequences & the role of psychological factors in causing stroke were linked to greater distress at 3mths. |
| Whittaker, Kemp & House ⁵³ (2007) | Mild TBI | <i>n</i> = 73 | M: 43% F: 57% | <i>M</i> =41.8 | Longitudinal (3 months) | B-IPQ | HADS IES RPQ | Distress Psychological well-being Physical functioning | Greater negative perceptions about consequences were linked with heightened risk of post-concussive syndrome. |
| Medley <i>et al.</i> ⁵⁴ (2010) | Severe TBI | <i>n</i> = 37 | M: 84% F: 16% | <i>M</i> = 39.5 <i>SD</i> =12.2 | Cross sectional | IPQ-R | WCCL-R EBIQ | Coping behaviour Psychological well-being | Low control and poor understanding were linked to avoidance. Greater timeline, consequences and illness identity beliefs were linked to positive coping strategies. |

| Author | Population | Sample | Gender | Age | Design | IPQ Type | Comparison | Outcome of interest | Outcome |
|--|--------------|---------------------|-----------------------|--------------------------------------|-----------------|----------|---|---|---|
| Evans, ⁵⁵ (2008) <i>D.ClinThesis</i> | CVA | <i>n</i> = 61 | M: 54.1% F: 45.9%% | <i>M</i> = 68.92 <i>SD</i> = 9.47 | Cross sectional | IPQ-R | COPE HADS PANS SSQ | Coping Distress Social support | Stronger illness identity, a cyclical timeline and believing that CVA was caused by psychosocial factors predicted anxiety. |
| Avison, ⁵⁶ (2009) <i>D.ClinThesis</i> | CVA & carers | <i>n</i> = 51 dyads | M: 64.7% F: 35.3%% | <i>M</i> = 64.9 <i>SD</i> = 9.25 | Cross sectional | IPQ-R | HADS | Distress | Patients and carers held discrepant perceptions but these did not significantly influence levels of anxiety or depression. |
| Dinsmore, ⁵⁷ (2010) <i>PhD Thesis</i> | CVA | <i>n</i> = 155 | M: 48.8% F: 51.6% | <i>M</i> = 71.1 <i>SD</i> = 13.3 | Longitudinal | IPQ-R | HADS SSQOL RLOC MSPSS SEIQOL-DW | Distress Social support Psychological well-being Quality of life | High levels of control, greater understanding and low illness identity or consequences were associated with improved quality of life and functional recovery. |

Key terms: CVA: cerebrovascular accident/ stroke, TBI: Traumatic brain injury, aSAH: Aneurysmal subarachnoid haemorrhage, PTG: Post traumatic growth

Key outcome measures:

IPQ: Illness Perception Questionnaire, **B-IPQ:** Brief Illness Perception Questionnaire, **IPQ-R:** Illness Perception Questionnaire Revised, **RPQ:** Rivermead Postconcussion Symptoms Questionnaire, **RHIFUQ:** Rivermead Head Injury Follow-up Questionnaire, **HADS:** Hospital Anxiety and Depression Scale, **SF-36:** Self-reported health-related QoL, **CHART:** Craig Handicap Assessment Reporting Technique, **FSQ:** Functional Status Questionnaire, **BRIQ:** Behavioural Response to Illness Questionnaire, **SSQ:** Brief Social Support Questionnaire, **PTGI:** Post Traumatic Growth Inventory, **FIM+FAM:** Functional Independence Measure & Functional Assessment Measure, **DTS:** Davidson Trauma Scale, **BMQ:** Beliefs about Medicines Questionnaire, **MARS:** Medication Adherence Report Scale, **DAS-21:** Depression Anxiety Stress Scale **WHOQOLBREF:** WHO Quality of Life BREF Version, **IES:** The Impact of Events Scale, **VAS:** Visual Analog Scale, **WCPS:** Ways of Coping with Problems Scale, **BDI:** Beck Depression Inventory, **EBIQ:** European Brain Injury Questionnaire, **WCCL-R:** The Ways of Coping Checklist- Revised, **SOS:** Significant Others Scale, **GHQ-28:** General Health Questionnaire, **PANS:** Positive and Negative Affect Schedule, **SEIQOL:** Schedule for the Evaluation of Individual Quality Of Life, **RLOC:** Recovery Locus of Control Scale, **MSPSS:** Multi-dimensional Perceived Social Support Scale. **SSQOL:** Stroke Specific Quality of Life scale.

Table 2. Adapted Newcastle-Ottawa Scale (NOS) quality assessment table.

| Authors | Selection/ Sample Representativeness | | | | | Comparability | | Assessment of outcome | | |
|---|--------------------------------------|----------------|--------------------|---------------------|------------------------|------------------------------------|---------------------------------------|-------------------------|--------------------------|---------------------------------|
| | 1. Representative population | 2. Sample size | 3. Non-respondents | 4. Exposure defined | 5. Fidelity to the IPQ | 6. Controlled for Injury variables | 7. Controlled for demographic factors | 8. Statistical analysis | 9. Assessment of outcome | 10. All IPQ dimensions reported |
| Snell <i>et al.</i> (2011) (2015) | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ |
| Saltapidas & Ponsford, (2008) | - | ☆ | - | ☆ | ☆ | ☆ | ☆ | ☆ | - | ☆ |
| Snell <i>et al.</i> (2013) | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ |
| Harris (2014) | ☆ | ☆ | ☆ | ☆ | - | ☆ | ☆ | ☆ | ☆ | ☆ |
| Rogan, Fortune & Prentice, (2013) | - | ☆ | - | ☆ | ☆ | ☆ | ☆ | ☆ | - | ☆ |
| Sheldrick, Tarrier, Berry & Kincey (2006) | ☆ | - | ☆ | ☆ | ☆ | - | ☆ | ☆ | - | ☆ |
| Jones <i>et al.</i> (2016) | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ |
| Sjölander, Eriksson & Glader (2013) | ☆ | ☆ | ☆ | ☆ | - | ☆ | ☆ | ☆ | ☆ | ☆ |

| Authors | Selection | | | | | Comparability | | Assessment of outcome | | |
|-----------------------------------|------------------------------|----------------|--------------------|---------------------|------------------------|------------------------------------|---------------------------------------|-------------------------|--------------------------|---------------------------------|
| | 1. Representative population | 2. Sample size | 3. Non-respondents | 4. Exposure defined | 5. Fidelity to the IPQ | 6. Controlled for injury variables | 7. Controlled for demographic factors | 8. Statistical analysis | 9. Assessment of outcome | 10. All IPQ dimensions reported |
| War & Rajeswaran (2012) (2013) | - | - | - | ☆ | - | ☆ | ☆ | ☆ | - | ☆ |
| Hou <i>et al.</i> , (2011) | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ | - | ☆ |
| Medley <i>et al.</i> , (2010) | - | - | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ | - | ☆ |
| Twiddy, House & Jones, (2012) | ☆ | - | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ | - | ☆ |
| Whittaker, Kemp & House, (2007) | ☆ | ☆ | - | ☆ | ☆ | - | ☆ | ☆ | - | ☆ |
| Nogueira & Teixeira, (2011) | ☆ | - | - | ☆ | - | - | ☆ | ☆ | - | ☆ |
| Evans (2008) | - | ☆ | - | ☆ | ☆ | - | ☆ | ☆ | ☆ | ☆ |
| Avison (2009) | - | ☆ | - | ☆ | - | - | ☆ | ☆ | - | - |
| Dinsmore (2010) | - | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ | ☆ |

Operationalising the quality criteria

Table 2 provides an overview of the quality ratings for each study in relation to the review question. Snell *et al.*, Jones *et al.*, Harris, Sjölander, Eriksson and Glader, Twiddy House and Jones, Dinsmore, and Hou *et al.* conducted the methodologically strongest studies, achieving 8 or more of the quality criteria [9,15,43,44,46,49,52,57]. Avison was the weakest study and was excluded from the narrative synthesis [56]. The remaining studies provided a good level of quality to warrant inclusion in the review [45,47,48,50,51,53,54,55].

Selection

The majority of studies (n=10) ensured participant representativeness by using multiple recruitment methods [9,15,43,44,46,48,49,51,52,53]. However, seven studies used inclusion or exclusion criteria that limited the generalisability of their findings. Dinsmore, Rogan, Fortune and Prentice and Medley recruited a sample that were at a later stage of recovery from their sudden neurological condition [47,54,57]. The latter two studies also used a rehabilitation service as a single recruitment site, which is likely to have led to a sample with a higher level of need and support [47,54]. Five studies recruited participants using just one site hospital site at a single time point [9,46,50,51,53]. Four studies omitted participants with significant cognitive or language impairments [47,48,52,55].

The longitudinal studies reported moderate attrition rates (15% to 35%). Five studies reported the use of a power calculation to determine their sample sizes [15,43,49,55,57] and the author of the review calculated post-hoc effect sizes for the remaining studies. Overall, eleven studies met the quality criteria for an adequate sample size [9,15,43,44,45,46,47,49,53,55,57].

Comparability

The methodologically stronger studies used multiple clinical tools to control for injury and demographic variables i.e. brain scans, assessment measures (such as the Glasgow Coma Scale), medical records and databases [9,15,43,44,46,47,49, 52,57]. Four studies used a single classification criteria (i.e. Glasgow Coma Scale) [45,49,50,52] and four studies did not control for the severity of participant's neurological condition [47,48,51,55].

Assessment of outcome

All of the included studies used appropriate statistical analysis. Snell *et al.* and Medley *et al.* computed additional analysis to cluster individuals according to illness perceptions, coping and health outcomes [15,45]. Eight studies failed to meet the assessment of outcome criteria as these studies relied on participant's self-report only which increased the risk of bias [46,47,48,50,51,52,53,54].

Fidelity to the use of the IPQ and the use of standardised health and coping outcomes measures were of particular relevance to the review. Thirteen studies met this quality criteria [15,43,44,45,46,47,48,52,53,54,55,57]. The above studies adapted the IPQ in accordance with the developer's recommendations by omitting or revising the wording of items and calculating Cronbach's alpha coefficients for each subscale for the target population (see *Table 3*). Three studies did not give a rationale for the partial administration and reporting of individual IPQ dimensions, suggestive of reporting bias [45,51,56].

Table 3. Adaptations or omissions to the Illness Perception Questionnaire

| Authors | Item omissions | Item adaptations | Item additions | Analysis/ Cronbach's alpha |
|--|--|---|--|---|
| Saltapidas & Ponsford, (2008) | | "Illness" replaced with "injury" | "Internal locus of control " "External locus of control" | CA: 0.84-0.96 Int. LOC (CA:0.82) Ext. LOC (CA:0.81) Principal Component Analysis |
| Sheldrick, Tarrier, Berry & Kinsey, (2006) | Causal attribution | | | Factor Analysis All 8 IPQ items 62.27% of variance |
| Rogan, Fortune & Prentice, (2013) | Coherence Consequences Personal control (CA:<0.7) | | | Principal Component Analysis CA:0.88 – 0.17 |
| Sjölander, Eriksson & Glader, (2013) | Causal attribution | "Illness" replaced with "stroke" | | Principal Component Analysis All CA: >0.7 |
| Twiddy, House & Jones, (2012) | Personal control (CA:0.59) | "Illness" replaced with "stroke" | "Physical risk factors" "Psychological risk factors" | Phys. RF (CA:0.75) Psych. RF (CA:0.78) Principal Component Analysis |
| Jones <i>et al.</i> , (2016) | Causal attribution | "Illness" replaced with "injury" | | Principal Component Analysis All CA:>0.7 |
| Snell <i>et al.</i> , (2011/15) (2013) | | "Illness" replaced with "injury" | "Concentration", "balance", "irritability", "memory" | Factor analysis All CA: >0.7 |
| Medley <i>et al.</i> , (2010) | | "Illness" replaced with "brain injury/problems" | "Lack of sleep", "drugs", "other people", "assault" | All CA: >0.7 Principal Component Analysis |
| Hou <i>et al.</i> , (2012) | Treatment control | "Illness" replaced with "injury" | | All CA: 0.85 |
| Evans (2008) | All causal items except "personality" | | "Behaviour risk factors" "Psychosocial attributions" "Unpredictable causes" "Health risk factors" | Principal Component Analysis |
| Dinsmore (2010) | | "illness" replaced with "stroke" | "Stress/ worry" "Personal health/ immunity" "Risk factors" "Behaviour/ ageing" "Genetics" "Immunity/ medical care" | Principal Component Analysis 6 additional items had scores >0.7 |
| Whittaker (2007) | | "Illness" replaced with "injury" | | All CA: >0.7 |

Narrative synthesis

A meta-synthesis was not performed as the population and the outcomes of interest (health questionnaires) were highly heterogeneous. Five of the six health outcomes from Hagger and Orbell's (2003) model (physical, social and role functioning, psychological distress and psychological well-being) and three coping strategies (emotion-focused, cognitive re-appraisal and problem-focused) were addressed in the included studies.

(1) Physical functioning

Five studies investigated the relationships between illness perceptions and PCS in those with a mild TBI [15,43,44,46,53]. Of these, three studies were rated to be of high methodologically quality. Snell *et al.* [15,43] and Jones *et al.* [44] found that individuals who attributed multiple symptoms to their brain injury (identity), believed symptoms to be severe (consequences) and long-lasting (timeline) and had low levels of control were at greater risk of developing PCS at three months post-injury. Snell *et al.* and Jones *et al.* also identified that early negative illness beliefs and not illness severity was predictive of PCS at six months [43,44]. Regarding their limitations, Jones *et al.* relied on self-report only to measure outcomes which increased the risk of response bias. Snell and colleagues reported sampling bias in that participants differed from non-respondents who were young and male. This is a TBI population known to be less likely to engage with healthcare services [58] and Snell *et al.* attempted to control for this in their statistical analysis.

Whilst Hou *et al.* [46], Whittaker, Kemp and House [53] also found similar relationships between illness perceptions and PCS, both studies relied on self-report only to assess outcomes. Whittaker and colleagues did not control for injury variables which further restricted their findings.

Sjölander, Eriksson and Glader found that illness perceptions did not influence patient adherence to stroke medication [49]. Despite significant results, this study did not report all dimensions of the IPQ, which may be suggestive of bias.

(2) Social, role functioning and quality of life

Six studies investigated the influence of illness perceptions on social or role functioning and quality of life [9,44,45,50,52,57]. Of these, Dinsmore [57] and Jones [44] were rated to be of the highest quality. Both studies showed that greater levels of perceived negative consequences or emotional reactions (i.e. anxiety or depression), lower coherence and a lack of personal control were predictive of poorer quality of life and unemployment. Although Dinsmore was rated as excellent overall, the study recruited participants later into their injury. It is not known whether illness perceptions between those with a chronic and acute injury are similar thus, these findings must be interpreted cautiously.

War & Rajeswaran also found that negative illness perceptions were correlated with lower quality of life, however, the study had methodological limitations that significantly affected its validity. These included the use of an

unequal and small sample (all male participants), recruiting from a single site and failing to use the IPQ as recommended by the developers [50].

Harris was rated to be of very good quality and found that 44% of patients did not return to work after their injury and those with lower levels of social support were at higher risk of a poorer employment outcome [9].

(3) Psychological distress and well-being

Five studies investigated the impact of illness perceptions on psychological distress and well-being [15,43,47,52,55]. The two strongest studies, conducted by Snell *et al.*, concluded that patients who did not fully understand their condition, who indicated greater emotional reactions to symptoms, higher personal control reported greater emotional distress [15,43].

Whilst relationships between illness perceptions, distress and well-being were shown in other studies included the review, the quality of those studies was variable. For instance, Rogan, Fortune and Prentice [47] found that those who attributed psychological factors to be the cause of injury reported greater distress. Similarly, Evans found that those who believed that their stroke was caused by psychosocial factors reported greater anxiety [55]. Both of these studies were constrained by their selection procedures. Rogan and colleagues recruited participants who were later into their injury from a single rehabilitation service and Evan's sample consisted of individuals who had been living with a spouse only for at least a year.

Sheldrick and colleagues identified that negative perceptions at the acute stage of injury were predictive of the development of post-traumatic stress disorder at 3 months [48]. However, the study had a relatively high attrition rate (35%) and found that non-respondents were more distressed than respondents at baseline, reporting more PTSD symptoms. It is likely that this study's findings are more applicable to those with less severe PTSD symptoms, somewhat limiting its generalisability.

Twiddy, House and Jones found that discrepancies in illness perceptions amongst stroke survivors and their carers were predictive of distress over time [52]. Although it was rated highly overall, the study had a small sample size and was reliant on self-report as the only method to assess outcomes. Further replication studies with a larger sample size of dyads are needed to add weight to these findings.

(4) Coping responses

Five studies investigated coping behaviours and illness perceptions [15,43,46,51,54]. The three methodologically strongest studies found that individuals who reported higher levels of personal or treatment control, lasting symptoms and a greater understanding of injury consequences used positive coping strategies like cognitive-reappraisal or problem-focused coping [15,43,46]. Additionally, Hou *et al.* found that adopting an all-or-nothing coping

style was found to be the most important predictor of PCS at three months [46].

The remaining two studies found significant relationships between negative illness perceptions and unhelpful ways of coping but had notable methodological flaws. Nogueira and Teixeira found that individuals who believed the cause of their pain to be due to their emotional reactions to stroke used more emotion-focused coping strategies [51]. However, this study was based on an underpowered sample size where the authors did not control for injury variables or use the IPQ in line with recommendations. Medley *et al.* found that individuals with TBI could be clustered based on their differing illness perceptions and coping styles [54]. This study used a singular method of recruitment with a population who were later into their neurological condition, thus limiting its validity to the population under study in the review.

Discussion

This systematic review aimed to determine whether and how illness perceptions influence health outcomes and coping in individuals with sudden onset neurological conditions. The review found that the individual's beliefs and interpretation of the impact of the event, not illness severity, consistently influenced their social, functional or psychological outcomes. The results support the use of the Self-Regulatory Model to elicit illness perceptions at the acute stages of neurological injury.

Patients with TBI who believed that their neurological condition had long lasting symptoms with life-limiting consequences and with low levels of control were more likely to develop post-concussion syndrome. The review findings extend the work of Meares *et al.* [59] and Silverberg & Iverson who found that early psychological factors play a crucial role in the development and maintenance of PCS [60]. Negative illness perceptions were also found to be strong predictors of unemployment, social isolation and poorer quality of life [44,45,50]. The listed factors have been linked to negative outcomes after sudden onset neurological conditions [61,11]. Consistent with existing research, the review found that a patient's perception of how the injury would affect their lives was more predictive of their social functioning than injury severity [62].

Interestingly, Sjölander, Eriksson and Glader found that illness perceptions were not significantly associated with medication adherence [49]. Instead, the

authors proposed that illness perceptions acted as a mediator between the individual's beliefs about medicines and their medication adherence. The indirect relationship between illness perceptions and medication has been supported by studies of other health populations, such as asthma [63] and HIV [64].

Patients that endorsed stronger emotional representations (i.e. low mood or anxiety), an internal locus of control and who attributed the cause of their neurological injury to psychological factors (i.e. endorsing beliefs like "*stress caused my stroke*") reported greater psychological distress [15,43,47,48,55]. These results support the work of Gómez-de-Regil who found that individuals who held low self-efficacy beliefs and increased personal responsibility for the onset of their illness reported greater psychological distress in psychosis [65]. Illness perceptions were also found to predict PTSD, suggesting that the disorder may be mediated by a patient's emotional responses to subarachnoid haemorrhage, their understanding of the impact on their lives and their confidence in medical treatment [48].

Another important finding was that discrepant patient-carer illness perceptions predicted greater distress for both parties over time [52]. Similar to Grice *et al.*'s study, patients reported greater distress when their carers endorsed more negative illness perceptions [66]. Research examining discrepant illness perceptions has yielded mixed findings. Some studies have found carer beliefs to predict patient outcomes [67] whereas others have failed to find consistent relationships [68].

Studies found that greater levels of personal control and understanding as well as attributing lifestyle factors to the cause of the neurological condition led to the use of adaptive coping styles [15,43,47]. This finding not only aligns to the Self-Regulatory Model but also draws parallels with Lazarus' general coping theory [69]. The theory proposes that greater emotional representations will foster emotion-focused coping and that higher levels of control will facilitate more emotion-focused approaches.

This review highlighted that the stage of injury and environment determined which coping styles were helpful or unhelpful. One study found that the use of problem-focused strategies was detrimental at the initial stages of injury [46]. Here, individuals who engaged in all-or-nothing coping behaviours (i.e. bursts of activity followed by exhausted rest) were more likely to develop post-concussion syndrome at 3 months post injury.

Additionally, studies found that some negative perceptions can be beneficial for recovery. Medley *et al.* clustered patients based on their coping styles, illness perceptions and health outcomes [54]. The "high salience" group demonstrated greater self-awareness, reported greater negative consequences and used a range of adaptive coping strategies. The "high optimism" and "ambivalent" groups tended to use avoidance coping which led to poorer outcomes. Conflicting accounts suggest that the relationship between coping and outcomes after neurological injury is complex and may be mediated by other factors like recovery trajectory or self-awareness.

Beliefs are likely to change over time depending on the nature of a person's health condition [63]. Two studies found that patients altered their perceptions of the impact of mild TBI and considered their condition to have more serious consequences at six-months compared to three-months post injury [15,43]. This result is consistent with Petrie's study that found that patients with myocardial infarction endorsed lower timeline and higher control beliefs at the acute stages of injury [63]. Petrie argued that patients at acute stages often believe that their treatment will be effective and that symptoms will alleviate over time. It is likely that individuals will develop more realistic beliefs about the chronicity of symptoms when discharged home and as the initial shock of the event passes [70].

Quality of the evidence

The majority of the studies ($n= 16$) were rated to be of good methodological quality. The strongest studies employed systematic methods of recruitment, appropriately controlled for demographic and injury variables, demonstrated fidelity to the IPQ and used standardised health outcome measures [9,15,43,44,46,49,52,57].

However, the review also highlighted methodological weaknesses. Four studies excluded those with significant cognitive or language impairment which is a common consequence of a sudden onset neurological condition [47,48,52,55]. Additionally, individuals were recruited through hospitals or

rehabilitation settings. Often, individuals with mild TBI do not present to services due to a delay in the emergence of symptoms [71]. Patients recruited in hospitals may demonstrate systematic differences in their needs and illness perceptions compared with the same population not in receipt of treatment, or those who receive treatment outside of hospital settings [15]. Selective recruitment may indicate self-selection bias or over-inflation of outcomes, both of which limit the external validity of findings [72].

Small sample sizes were conspicuous areas of weakness [48,50,51,54]. With weak statistical power, it is difficult to draw firm conclusions regarding the generalisability of results. One studies used correlational analysis only [50] which precluded the ability to establish causal links between illness perceptions and outcomes. Over half of the studies were cross-sectional and assessed all outcomes of interest concurrently [15,45,47,49,50,51,54,55,56]. Thus, inferences about the predictive relationships between illness perceptions and outcomes in the cross-sectional studies were limited.

Finally, there was heterogeneity in the study design, types of neurological injuries and the health outcomes investigated. This was unavoidable, given the limited literature available on any one condition, but did reduce the specificity of results.

Strengths and limitations of the review

A strength of the study lay in its use of a systematic search strategy to review the literature in a novel area of research. Citation searches were conducted on the included studies and authors of the included studies were contacted to request unpublished findings to limit publication bias. A second rater was used to assess the methodological quality of studies to further mitigate bias.

The main limitation of this review was that only English language studies were included. This may have led to possible language bias in the review process. In addition, three studies reported a longer time since injury [47,54,57] and two studies recruited participants from rehabilitation services [47,54]. These studies were included based on the wide inclusion criteria of the review. However, the findings cannot be directly comparable to other studies that reported on populations at the acute stages of injury (1-6 months). Nonetheless, all studies found significant associations between illness perceptions, coping and health outcomes which contributes to the research question.

Future research directions

The mechanisms of injury in all of the included papers were either cerebrovascular or traumatic in nature. No studies were identified in the literature that explored illness perceptions in people with neuro-infections or inflammation. Illness perceptions may invariably differ in individuals with an acute infection compared with other neurological conditions. For instance, a person may expect longer lasting and more severe effects from a traumatic brain injury compared with an acute infection. There is a lack of knowledge about neuro-infections in the general population [73]. Less understanding about symptoms, timeline or causes may increase the likelihood of a person developing idiosyncratic beliefs or misconceptions which could delay treatment-seeking or impede recovery [74]. Further investigation is warranted as the literature develops in these neurological conditions.

Cognitive status, motivation and levels of awareness are likely to fluctuate hugely within the first few months of a sudden neurological event [75]. Reduced insight and lasting cognitive deficit will interfere with an individual's ability to synthesise and understand information to make sense of their condition [76]. The mediating role of neurocognitive factors on illness perceptions has yet to be explored [54]. Future studies investigating the relationship between cognition and illness perceptions should incorporate a brief, easy to administer screening tool like the Mini-Mental State Examination [77], Addenbrooke's Cognitive Examination-III [78] or the Montreal Cognitive Assessment [79].

The review found no evidence of gender differences but studies did find that socio-cultural beliefs [45] and education status [43,45] influenced the individual's internal working model of their condition. This fits with Leventhal and colleagues' theory that illness perceptions are moderated by past knowledge of illness, personality characteristics and information gained from others [18]. Further assessment of demographic and socioeconomic factors in acute settings with respect to illness perceptions may unveil important considerations.

Weinman *et al.* recommended that the IPQ be condition-specific [23]. The development of condition-specific adaptations (i.e. the IPQ adapted for brain injury) would allow for within-group comparisons which would strengthen the validity of findings. An avenue for further research would be to develop adaptations based directly on the patient's perspectives (patient-reported outcome measures, PROM). An advantage of PROM is that it captures the patient's own opinions and beliefs about the impact of their health condition on their lives [80]. Merging condition-specific PROMs with the IPQ in future studies may ensure more robust within-group comparisons in the neurological population of interest.

All longitudinal studies included in this review assessed whether illness perceptions at a discrete time point could predict health outcomes at 3 or 6 months [43,46,48,52,53]. A more comprehensive study would measure

associations across multiple time points to assess the predictive validity of illness perceptions on outcomes along the trajectory of a person's recovery.

Illness perceptions have been used as a means of predicting coping behaviours in physical health populations [81]. Friedman delineates coping into two categories; (1) a person's decision to seek medical care and (2) their adherence to medical recommendations [82]. The predicative relationship has been most evident in the myocardial infarction (MI) population. Cooper, Jackson, Weinman and Horne illustrated that individuals with MI who endorsed stronger beliefs about personal and treatment control at the early stages of illness were more likely to attend cardiac rehabilitation [83]. Assessing the predictive value of illness perceptions on adherence to cognitive rehabilitation or self-management behaviours after neurological injury would be another interesting avenue for research.

Are illness perceptions predictive of outcomes because they contain information that cannot be picked up on clinical tools, or because illness perceptions influence the person's present behaviours which then affects their future outcomes? This review found strong associations between illness perceptions and post-concussive syndrome. Using this relationship as an example, a patient may report fatigue and irritability due to PCS which may be accurate symptoms of their condition. On the other hand, negative perceptions of PCS and brain injury may lead the person to cope passively, withdraw from others and reduce their activities. Avoidance may lead to a further decline in physical and mental health, causing poorer outcomes in the future. In this

case, it is difficult to ascertain whether fatigue and irritability are due to the person's physical condition or due to poor health behaviours as a result of negative illness perceptions. Further research is required to uncover the mechanisms that could account for such effects [28].

Clinical implications

The IPQ has good discriminant validity and can highlight specific profiles within health conditions [24]. For instance, individuals with diabetes have been found to report longer timeline beliefs. Individuals with myocardial infarction describe higher treatment control beliefs as a consequence of hospitalisation and high volumes of health information [29]. This review found that groups of individuals with traumatic brain injury could be clustered based on their health outcomes and coping styles [15,54]. Condition-specific illness perceptions could be used as prognostic indicators to predict recovery trajectories in specific neurological conditions. This could be used in future to inform clinical-decision making and treatment guidelines.

There is growing support for the use of psychological interventions to restructure maladaptive illness perceptions in chronic health conditions like cancer, coronary heart disease, non-cardiac chest pain and chronic lower back pain [25,84,85,86]. For instance, Petrie and colleagues found that targeting negative illness perceptions improved functional outcomes and led to a quicker return to work rate in individuals with myocardial infarction [63]. Evidence for the efficacy of using psychological therapy in those with sudden onset

neurological conditions is modest [87,88]. In a recent meta-analysis, Waldron, Caserly and O'Sullivan found that CBT demonstrated moderate effect sizes when used to treat anxiety and depression in TBI [89]. However, many studies only reported partial reductions in symptoms. In the first randomised control trial of CBT for stroke, Lincoln & Flannaghan found insufficient evidence for the merits of CBT [90]. They attributed this to the limitations of the protocol and the use of a one-size-fits all approach. More recently, Taylor and colleagues suggested the use of an augmented form of CBT, tailored to the individual, accounting for stroke-related disability and cognitive impairment [91].

In light of the review findings, there is considerable potential to develop cognitive-based interventions to re-structure maladaptive illness perceptions at the acute stages of neurological injury. Augmented psychological therapy could be useful in encouraging patients to develop more realistic expectations about recovery (timeline) and adjust to the impact of their injury (consequences). Psychosocial interventions could support the person to make gradual changes to resume their social and functional participation (control and coping).

Current clinical practice guidelines recommend the early provision of information, support and advice [92]. Reassurance and education in the first few weeks or months after neurological injury will normalise symptoms and improve the patient's understanding of their condition (cause and coherence). Person-centred interventions may prevent patients from developing

idiosyncratic or maladaptive illness perceptions that may predispose them to poorer outcomes.

The significance of emotional adjustment and the impact of this on the timing of interventions is an important consideration [93]. Dependent upon the injury, patients will undergo a process of bereavement associated with loss of functioning, which often occurs simultaneous to their physical recovery [94]. Thus, information about the emotional consequences of injuries should be staged accordingly. Rehabilitation should occur in tandem with medical care at the acute stages of neurological injury and patients should be considered to be active recipients in this process [95]. Eliciting patient illness perceptions at the early stages of their condition may not only inform clinicians of the patient's levels of adjustment but also their motivation to engage in rehabilitation and their likelihood to seek support in the months that follow.

Implications for Rehabilitation

- The Self-Regulatory Model has utility in understanding patient beliefs and expectations about recovery after a sudden neurological injury.
- Beliefs about multiple symptoms with lasting serious consequences, low levels of control and a lack of understanding about the causes of injury are linked with poor psychosocial and physical outcomes. Negative illness perceptions can delay help-seeking and impede recovery.
- Illness perceptions are modifiable factors that can be the target of early individualised interventions that can restructure maladaptive beliefs, alleviate future distress and encourage adaptive coping.
- Whilst the literature in this field is expanding, further high-quality research is needed to confirm the predictive validity of illness perceptions on outcomes and coping in neurological populations.

References

- [1] Chowdhury RN, Hasan AH, Rahman YU, Khan SI, Hussain AR, Ahsan S. Pattern of neurological disease seen among patients admitted in tertiary care hospital. BMC. 2014;7(1):202.
- [2] World Health Organization. The world health report 2002: reducing risks, promoting healthy life. World Health Organization. 2002. Accessed_May2017from:http://www.who.int/whr/2002/en/w hr02_en.pdf?ua=1
- [3] National Institute for Clinical Health and Excellence. Clinical guideline CG176: Head injury: assessment and early management. 2014. Accessed_May2017from:<https://www.nice.org.uk/guidance/cg176 /chapter/3-Otherinformation>.
- [4] Granerod J, Ambrose HE, Davies NW, Clewley JP, Walsh AL, Morgan D, Ward KN. Causes of encephalitis and differences in their clinical presentations in England: a multicentre, population-based prospective study. The Lancet. 2010;10(12): 835-844.
- [5] King, NS. (2003). Post-concussion syndrome: clarity amid the controversy? B.J.Psych. 2003; 276-278.
- [6] Ryan LM, Warden DL. Post-concussion syndrome. International review of psychiatry. 2003 Nov 1;15(4):310-6.
- [7] King, N. & Kirwilliam, S. Permanent Post-Concussion Symptoms after Mild Head Injury. Brain Injury. 2011; 25: 462-470.

- [8] Coole C, Radford K, Grant M, Terry J. Returning to work after stroke: perspectives of employer stakeholders, a qualitative study. *Journal of Occupational Rehabilitation*. 2013;23(3):406-18.
- [9] Harris C. Return to Work After Stroke. *Stroke*. 2014;45(9):174-6.
- [10] Foxwell R, Morley C, Frizelle D. Illness perceptions, mood and quality of life: A systematic review of coronary heart disease patients. *Journal of Psychosomatic Research*. 2013;75(3):211-22.
- [11] Hackett, M. L., Anderson, C. S., House, A. O., & Xia, J. Interventions for treating depression after stroke. *Stroke*. 2009; 40(7): 487.
- [12] Berry E. Post-traumatic stress disorder after subarachnoid haemorrhage. *British Journal of Clinical Psychology*. 1998; 37(3): 365-367.
- [13] Powell, J, Kitchen, N, Heslin, J, Greenwood R. Psychosocial outcomes at three and nine months after good neurological recovery from aneurysmal subarachnoid haemorrhage: predictors and prognosis. *Journal of Neurology, Neurosurgery & Psychiatry*. 2002;72(6):772-781.
- [14] Kreutzer JS, Rapport LJ, Marwitz JH, Harrison-Felix C, Hart T, Glenn M, Hammond F. Caregivers' well-being after traumatic brain injury: a multicentre prospective investigation. *Archives of Physical Medicine and Rehabilitation*. 2009;90(6):939-46.
- [15] Snell DL, Siegert RJ, Hay-Smith EJ, Surgenor LJ. Associations between illness perceptions, coping styles and outcome after mild traumatic brain injury: preliminary results from a cohort study. *Brain Injury*. 2011;25(11):1126-38.

- [15] Snell DL, Surgenor LJ, Hay-Smith EJ, Williman J, Siegert RJ. The contribution of psychological factors to recovery after mild traumatic brain injury: Is cluster analysis a useful approach? *Brain injury*. 2015;29(3):291-9.
- [16] Vestling M, Tufvesson B, Iwarsson S. Indicators for return to work after stroke and the importance of work for subjective well-being and life satisfaction. *Journal of Rehabilitation Medicine*. 2003 Jan 1;35(3):127-31.
- [17] Van Mierlo ML, van Heugten CM, Post MW, Hajós TR, Kappelle LJ, Visser-Meily JM. Quality of life during the first two years post stroke: the Restore4Stroke cohort study. *Cerebrovascular Diseases*. 2016;41(1-2):19-26.
- [18] Leventhal H, Meyer D, Nerenz D. The common-sense representation of illness danger. *Contributions to Medical Psychology*. 1980; 2:7-30.
- [19] Petrie KJ, Weinman J. Patients' perceptions of their illness: The dynamo of volition in health care. *Current Directions in Psychological Science*. 2012;21(1):60-5.
- [20] Leventhal H, Benyamini Y, Brownlee S, Diefenbach M, Leventhal EA, Patrick-Miller L, Robitaille C. Illness representations: theoretical foundations. *Perceptions of Health and Illness*. 1997; 2:19-46.
- [21] Moss-Morris R, Weinman J, Petrie K, Horne R, Cameron L, Buick D. The revised illness perception questionnaire (IPQ-R). *Psychology and Health*. 2002;17(1):1-6.

- [22] Leventhal H, Leventhal EA, Contrada RJ. Self-regulation, health, and behavior: A perceptual-cognitive approach. *Psychology and Health*. 1998;13(4):717-33.
- [23] Weinman J, Petrie KJ, Moss-Morris R, Horne R. The illness perception questionnaire: a new method for assessing the cognitive representation of illness. *Psychology and Health*. 1996;11(3):431-45.
- [24] Broadbent E, Petrie KJ, Main J, Weinman J. The brief illness perception questionnaire. *Journal of Psychosomatic Research*. 2006;60(6):631-7.
- [25] Broadbent E, Niederhoffer K, Hague T, Corter A, Reynolds L. Headache sufferers' drawings reflect distress, disability and illness perceptions. *Journal of Psychosomatic Research*. 2009;66(5):465-70.
- [26] Hagger MS, Orbell S. A meta-analytic review of the common-sense model of illness representations. *Psychology and Health*. 2003;18(2):141-84.
- [27] Petrie KJ, Weinman J. Perceptions of health and illness: current research and applications. Taylor & Francis; 1997; pg. 149-155.
- [28] Benyamini Y. Health and illness perceptions. *The Oxford handbook of health psychology*. 2011; pg. 281-314.
- [29] Broadbent E, Wilkes C, Koschwanez H, Weinman J, Norton S, Petrie KJ. A systematic review and meta-analysis of the Brief Illness Perception Questionnaire. *Psychology & Health*. 2015; 30(11): 1361-85.

- [30] O'Donovan CE, Painter L, Lowe B, Robinson H, Broadbent E. The impact of illness perceptions and disease severity on quality of life in congenital heart disease. *Cardiology in the Young*. 2016; 26(01): 100-9.
- [31] Schoormans D, Mulder BJ, van Melle JP, Pieper PG, van Dijk AP, Sieswerda GT, Hulsbergen-Zwarts MS, Plokker TH, Brunninkhuis LG, Vliegen HW, Sprangers MA. Illness perceptions of adults with congenital heart disease and their predictive value for quality of life two years later. *European Journal of Cardiovascular Nursing*. 2014;13(1):86-94.
- [32] Goldstein LH, Holland L, Soteriou H & Mellers JD. Illness representations, coping styles and mood in adults with epilepsy. *Epilepsy Res*. 2005; 67(1):1–11.
- [33] Vaughan R, Morrison L, Miller E. The illness representations of multiple sclerosis and their relations to outcome. *British Journal of Health Psychology*. 2003;8(3):287-301.
- [34] Miller L. The "trauma" of head trauma: Clinical, neuropsychological, and forensic aspects of posttraumatic stress syndromes in brain injury. *Journal of Cognitive Rehabilitation*. 1993;16(1) 100-110.
- [35] Centre for reviews, dissemination (CRD). Systematic reviews: CRD's guidance for undertaking reviews in health care. Centre for Reviews and Dissemination; 2009. Accessed May 2017 from: https://www.york.ac.uk/media/crd/Systematic_Reviews.pdf.
- [36] Higgins JP, Green S, editors. *Cochrane handbook for systematic reviews of interventions*. John Wiley & Sons. 2011.

- [37] McAleese N, Gillespie D, O'Rourke S & Guzman A. A systematic review of illness perceptions, coping and health outcomes in adults with sudden onset neurological conditions. PROSPERO 2017: CRD42017054487 Available from http://www.crd.york.ac.uk/PROSPERO/display_record.asp?ID=CRD42017054487
- [38] di Ruffano LF, Dinnes J, Sitch AJ, Hyde C, Deeks JJ. Test-treatment RCTs are susceptible to bias: a review of the methodological quality of randomized trials that evaluate diagnostic tests. *BMC medical research methodology*. 2017;17(1):35.
- [39] Wells G.A, Shea B, O'Connell D, Peterson J, Welch V, Losos M. & Tugwell, P. The Newcastle-Ottawa Scale (NOS) for assessing the quality of non-randomised studies in meta-analyses. Ottawa Hospital Research Institute. Accessed May 2017: http://www.ohri.ca/programs/clinical_epidemiology/oxford.asp .
- [40] Viswanathan M, Ansari MT, Berkman ND, Chang S, Hartling L, McPheeters M, Santaguida PL, Shamliyan T, Singh K, Tsertsvadze A, Treadwell JR. Assessing the risk of bias of individual studies in systematic reviews of health care interventions. 2012. Accessed May 2017 at: www.effectivehealthcare.ahrq.gov/
- [41] Hartling L, Milne A, Hamm MP, Vandermeer B, Ansari M, Tsertsvadze A, Dryden DM. Testing the Newcastle Ottawa Scale showed low reliability between individual reviewers. *Journal of Clinical Epidemiology*. 2013;66(9):982-93.

- [42] Scottish Intercollegiate Guidelines Network (SIGN). SIGN 50: a guideline developer's handbook. Edinburgh: SIGN; 2015. (SIGN pub.no. 50). Accessed May 2017 from: <http://www.sign.ac.uk/pdf/sign50.pdf>.
- [43] Snell DL, Hay-Smith EJ, Surgenor LJ, Siegert RJ. Examination of outcome after mild traumatic brain injury: The contribution of injury beliefs and Leventhal's Common-Sense Model. *Neuropsychological rehabilitation*. 2013;23(3):333-62.
- [44] Jones KM, Kydd R, Broadbent E, Theadom A, Barker-Collo S, Edwards H, Feigin VL. Brain drawings following traumatic brain injury (TBI) and links to illness perceptions and health outcomes—Findings from a population-based study. *Psychology & Health*. 2016;31(10):1182-202.
- [45] Saltapidas H, Ponsford J. The influence of cultural background on experiences and beliefs about traumatic brain injury and their association with outcome. *Brain Impairment*. 2008;9(01):1-3.
- [46] Hou R, Moss-Morris R, Peveler R, Mogg K, Bradley BP, Belli A. When a minor head injury results in enduring symptoms: a prospective investigation of risk factors for post-concussional syndrome after mild traumatic brain injury. *Journal of Neurology, Neurosurgery & Psychiatry*. 2012;83(2):217-23.
- [47] Rogan C, Fortune DG, Prentice G. Post-traumatic growth, illness perceptions and coping in people with acquired brain injury. *Neuropsychological Rehabilitation*. 2013;23(5):639-57.

- [48] Sheldrick R, Tarrier N, Berry E, Kincey J. Post-traumatic stress disorder and illness perceptions over time following myocardial infarction and subarachnoid haemorrhage. *British Journal of Health Psychology*. 2006;11(3):387-400.
- [49] Sjölander M, Eriksson M, Glader EL. The association between patients' beliefs about medicines and adherence to drug treatment after stroke: a cross-sectional questionnaire survey. *BMJ*. 2013;3(9):3551.
- [50] Var FA, Rajeswaran J. Perception of illness in patients with traumatic brain injury. *Indian Journal of Psychological Medicine*. 2012; 34(3):223.
- [50] Var FA, Rajeswaran J. Quality of life and perception of illness in patients with traumatic brain injury. *The Indian Journal of Neuro-Trauma*. 2013; 10(1): 115-119
- [51] Nogueira M, Teixeira MJ. Central pain due to stroke: cognitive representation and coping according to gender. *Arquivos de neuro-psiquiatria*. 2012;70(2):125-8.
- [52] Twiddy M, House A, Jones F. The association between discrepancy in illness representations on distress in stroke patients and carers. *Journal of Psychosomatic Research*. 2012;72(3):220-5.
- [53] Whittaker R, Kemp S, House A. Illness perceptions and outcome in mild head injury: a longitudinal study. *Journal of Neurology, Neurosurgery & Psychiatry*. 2007;78(6):644-6.
- [54] Medley AR, Powell T, Worthington A, Chohan G, Jones C. Brain injury beliefs, self-awareness, and coping: a preliminary cluster analytic

- study based within the self-regulatory model. *Neuropsychological Rehabilitation*. 2010;20(6):899-921.
- [55] Evans S. Stroke, psychological outcomes and the self-regulatory model. Doctoral dissertation, Lancaster University. 2008; 53-100.
- [56] Avison R. Discrepant illness perceptions in stroke survivor-partner dyads: relationship to psychological adjustment and expressed emotion. Doctoral dissertation, University of Hull. 2009; 54-94.
- [57] Dinsmore JG. Quality of life and illness perception in Stroke Patients: A 12 Month Study. Doctoral dissertation, Queen's University Belfast. 2010.
- [58] Snell D, Surgenor L. An analysis of referees and referrals to a specialist concussion clinic in New Zealand. *The New Zealand Medical Journal*. 2006; 119:1231.
- [59] Meares S, Shores EA, Batchelor J, Baguley IJ, Chapman J, Gurka J, Marosszeky JE. The relationship of psychological and cognitive factors and opioids in the development of the post-concussion syndrome in general trauma patients with mild traumatic brain injury. *Journal of the International Neuropsychological Society*. 2006;12(06):792-801.
- [60] Silverberg, ND, Iverson, GL. Etiology of the post-concussion syndrome: physiogenesis and psychogenesis revisited. *NeuroRehabilitation*. 2011;29(4):317-329
- [61] Cacioppo JT, Hawkley LC. Social isolation and health, with an emphasis on underlying mechanisms. *Perspectives in Biology and Medicine*. 2003;46(3): S39-52.

- [62] Bergman S, Jacobsson LT, Herrström P, Petersson IF. Health status as measured by SF-36 reflects changes and predicts outcome in chronic musculoskeletal pain: a 3-year follow up study in the general population. *Pain*. 2004;108(1):115-23.
- [63] Petrie KJ, Cameron LD, Ellis CJ, Buick D, Weinman J. Changing illness perceptions after myocardial infarction: an early intervention randomized controlled trial. *Psychosomatic Medicine*. 2002; 64(4):580-6.
- [64] Weiss JJ, Konstantinidis I, Boueilh A, Fierer DS, Gardenier D, Barber MG, Kang T, Kress A, Ericson K, Lira MC, Yostos MP. Illness perceptions, medication beliefs, and adherence to antiretrovirals and medications for comorbidities in adults with HIV infection and hypertension or chronic kidney disease. *JAIDS Journal of Acquired Immune Deficiency Syndromes*. 2016;73(4):403-10.
- [65] Gómez-de-Regil L, Kwapil TR, Barrantes-Vidal N. Predictors of expressed emotion, burden and quality of life in relatives of Mexican patients with psychosis. *Journal of Psychiatric and Mental Health Nursing*. 2014;21(2):170-9.
- [66] Grice SJ, Kuipers E, Bebbington P, Dunn G, Fowler D, Freeman D, Garety P. Carers' attributions about positive events in psychosis relate to expressed emotion. *Behaviour Research and Therapy*. 2009 Sep 30;47(9):783-9.
- [67] Dempster M, McCorry NK, Brennan E, Donnelly M, Murray LJ, Johnston BT. Do changes in illness perceptions predict changes in

psychological distress among oesophageal cancer survivors?
Journal of Health Psychology. 2011;16(3):500-9.

- [68] Kaptein AA, Scharloo M, Helder DI, Snoei L, Van Kempen GM, Weinman J, van Houwelingen JC, Roos RA. Quality of life in couples living with Huntington's disease: the role of patients' and partners' illness perceptions. *Quality of Life Research*. 2007;16(5):793-801.
- [69] Lazarus RS. Coping theory and research: past, present, and future. *Psychosomatic Medicine*. 1993;55(3):234-47.
- [70] Alsén P, Brink E, Persson LO, Brändström Y, Karlson BW. Illness perceptions after myocardial infarction: relations to fatigue, emotional distress, and health-related quality of life. *Journal of Cardiovascular Nursing*. 2010;25(2): e1-10.
- [71] Corrigan, J. D., Selassie, A. W., & Orman, J. A. L. The epidemiology of traumatic brain injury. *The Journal of Head Trauma Rehabilitation*. 2010; 25(2): 72-80.
- [72] McCullagh S, Feinstein A. Outcome after mild traumatic brain injury: an examination of recruitment bias. *Journal of Neurology, Neurosurgery & Psychiatry*. 2003;74(1):39-43.
- [73] Modabbernia A, Poustchi H, Malekzadeh R. Neuropsychiatric and psychosocial issues of patients with hepatitis C infection: a selective literature review. *Hepatitis monthly*. 2013;13(1): e8340.
- [74] MacInnes JD. The illness perceptions of women following symptoms of acute myocardial infarction: a self-regulatory approach. *European Journal of Cardiovascular Nursing*. 2006; 5(4):280-8.

- [75] Carron SF, Alwis DS, Rajan R. Traumatic brain injury and neuronal functionality changes in sensory cortex. *Frontiers in Systems Neuroscience*. 2016;10.
- [76] Morris JC, Storandt M, Miller JP, McKeel DW, Price JL, Rubin EH, Berg L. Mild cognitive impairment represents early-stage Alzheimer disease. *Archives of Neurology*. 2001 Mar 1;58(3):397-405.
- [77] Folstein MF, Folstein SE, McHugh PR. "Mini-mental state": a practical method for grading the cognitive state of patients for the clinician. *Journal of Psychiatric Research*. 1975;12(3):189-98.
- [78] Mathuranath PS, Nestor PJ, Berrios GE, Rakowicz W, Hodges JR. A brief cognitive test battery to differentiate Alzheimer's disease and frontotemporal dementia. *Neurology*. 2000;55(11):1613-20.
- [79] Nasreddine ZS, Phillips NA, Bédirian V, Charbonneau S, Whitehead V, Collin I, Cummings JL, Chertkow H. The Montreal Cognitive Assessment, MoCA: a brief screening tool for mild cognitive impairment. *Journal of the American Geriatrics Society*. 2005;53(4):695-9.
- [80] Black N. Patient reported outcome measures could help transform healthcare. *BMJ (Clinical research edition)*. 2013; 346: 367.
- [81] Searle A, Norman P, Thompson R, Vedhara K. A prospective examination of illness beliefs and coping in patients with type 2 diabetes. *British Journal of Health Psychology*. 2007;12(4):621-38.
- [82] Friedman HS. *The Oxford Handbook of Health Psychology*. 2nd edition. Oxford University Press. 2011; pg. 205-210.

- [83] Cooper AF, Jackson G, Weinman J, Horne R. Factors associated with cardiac rehabilitation attendance: a systematic review of the literature. *Clinical rehabilitation*. 2002;16(5):541-52.
- [84] Harman G, Clare L. Illness representations and lived experience in early-stage dementia. *Qualitative Health Research*. 2006; 16(4): 484-502.
- [85] Jonsbu E, Martinsen EW, Morken G, Moum T, Dammen T. Change and impact of illness perceptions among patients with non-cardiac chest pain or benign palpitations following three sessions of CBT. *Behavioural and Cognitive Psychotherapy*. 2013;41(04):398-407.
- [86] Siemonsma PC, Stuive I, Roorda LD, Vollebregt JA, Walker MF, Lankhorst GJ, Lettinga AT. Cognitive treatment of illness perceptions in patients with chronic low back pain: a randomized controlled trial. *Physical Therapy*. 2013;93(4):435.
- [87] Comper P, Bisschop SM, Carnide N, Tricco A. A systematic review of treatments for mild traumatic brain injury. *Brain Injury*. 2005;19(11):863-80.
- [88] Soo C, Tate RL. Psychological treatment for anxiety in people with traumatic brain injury. *The Cochrane Library*. 2007.
- [89] Waldron B, Casserly LM, O'Sullivan C. Cognitive behavioural therapy for depression and anxiety in adults with acquired brain injury. What works for whom? *Neuropsychological Rehabilitation*. 2013;23(1):64-101.
- [90] Lincoln NB, Flannaghan T. Cognitive behavioral psychotherapy for depression following stroke. *Stroke*. 2003;34(1):111-5.

- [91] Taylor GH, Todman J, Broomfield NM. Post-stroke emotional adjustment: A modified social cognitive transition model. *Neuropsychological Rehabilitation*. 2011;21(6):808-24.
- [92] Scottish Intercollegiate Guidelines Network (SIGN). SIGN 118: Management of patients with stroke: Rehabilitation, prevention and management of complications, and discharge planning A national clinical guideline 2010. Retrieved on 9th May from: <http://www.sign.ac.uk/pdf/sign118.pdf>
- [93] Wade DT. Measurement in neurological rehabilitation. *Current Opinion in Neurology*. 1992;5(5):682-6.
- [94] Coetzer R. Traumatic brain injury rehabilitation: A psychotherapeutic approach to loss and grief. Nova Publishers; 2006.
- [95] Wade D. Rehabilitation—a new approach. Overview and Part One: the problems. *Clinical Rehabilitation*. 2015; 1041-1050.

Appendix A: Preferred Reporting Items for Systematic review and Meta-Analysis Protocols (PRISMA) protocol for systematic reviews checklist

| Section/topic | # | Checklist item |
|------------------------------------|----|---|
| TITLE | | |
| Title | 1 | Identify the report as a systematic review, meta-analysis, or both. |
| ABSTRACT | | |
| Structured summary | 2 | Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number. |
| INTRODUCTION | | |
| Rationale | 3 | Describe the rationale for the review in the context of what is already known. |
| Objectives | 4 | Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS). |
| METHODS | | |
| Protocol and registration | 5 | Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number. |
| Eligibility criteria | 6 | Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale. |
| Information sources | 7 | Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched. |
| Search | 8 | Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated. |
| Study selection | 9 | State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis). |
| Data collection process | 10 | Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators. |
| Data items | 11 | List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made. |
| Risk of bias in individual studies | 12 | Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis. |
| Summary measures | 13 | State the principal summary measures (e.g., risk ratio, difference in means). |
| Synthesis of results | 14 | Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I^2) for each meta-analysis. |

Appendix B: Systematic review protocol based on York University's Centre for Reviews and Dissemination Guidance for undertaking reviews in healthcare (CRD, 2009)

Review title: A systematic review of the impact of patient's illness perceptions on coping and outcomes after brain injury.

Review question: Are illness perception dimensions related to coping styles and health outcomes in adults with an acquired or traumatic brain injury?

Searches

1. The following electronic databases will be used: MEDLINE, EMBASE, PsycINFO, Ebsco host CINAHL, Web of Science, Cochrane library, Google Scholar, ETHoS, OpenGrey.
2. Manual searches of the reference lists from relevant papers will locate additional citations of articles.
3. Key authors identified during the review process will be contacted to provide information regarding missing data or to inform the authors of ongoing work.

Types of study to be included

Inclusion criteria:

- Quantitative research design
- Full text articles
- Cross-sectional, cohort and longitudinal studies
- From 1996 – 2017
- Diagnosis of either an acquired or traumatic brain injury
- A validated measure of coping or illness outcome as categorised by Hagger & Orbell [26].
- Use of the Illness Perception Questionnaire or adapted versions
- Studies written in English
- Adults (18+)

Exclusion criteria:

- Studies with a qualitative or mixed-methods research design
- Editorials, conference articles or reviews
- Abstracts not accompanied by full text
- Studies where the primary focus is care givers/ partners or care professionals

Search terms:

("illness perception" OR "self- regulation" OR "patient attitude*" OR "illness cognition" OR "health belief*" OR "common sense" OR "illness representation" OR IPQ)*

AND

("head injur" OR "brain injur*" OR "brain infection" OR stroke OR CVA OR "cerebrovascular accident" OR "subarachnoid hemorrhag*" OR "subarachnoid haemorrhag*" OR ischemi* OR TBI OR ABI OR concussion)*

NOT

("myocardial infarction)

Condition or domain being studied

Illness perceptions, coping and illness outcomes.

Participants/ population

Adults (+18 years) with a diagnosis of an acquired or a traumatic brain injury.

Intervention(s), exposure(s)

Not applicable

Comparator(s)/ control

No comparator or control group

Context

Any primary, secondary or tertiary care/community setting.

Primary outcomes

Illness perceptions, coping and illness outcomes.

Data extraction, (selection and coding)

Selection –

1. Titles and abstracts of studies will be retrieved using the search strategy. Those from additional sources will be screened independently by two review authors to identify studies that meet the inclusion criteria. Full texts of potentially eligible studies will be retrieved and independently assessed for suitability for inclusion by two review authors.

Data extraction –

2. A coding form adapted will be used to extract data for the assessment of study quality and evidence synthesis. Two review authors will extract data independently. Discrepancies will be identified and resolved through discussion.

Risk of bias (quality) assessment

The Newcastle-Ottawa scale (Wells *et al.*, 2012) will be used to assess the methodological quality of the selected studies. This tool is an 8-item checklist and has been listed as one of the most useful quality assessment instruments according to a review by Deeks *et al.* (2003). It is also recommended by the Cochrane Handbook in the assessment of non-randomised studies (Higgins & Green, 2011).

Strategy for data synthesis

Study characteristics, associations and outcomes will be reported following Cochrane recommended guidelines for narrative synthesis (Popay *et al.*, 2006). Should an assumption of homogeneity exist then an additional meta-analysis will be computed to measure the size of the effect of illness perceptions on coping and outcomes.

Dissemination plans

The results will be submitted to a relevant psychology peer-reviewed journal and shared with health professionals at a local and national level via conferences.

Appendix C: Data extraction form

| |
|--|
| Study ID (<i>surname of first author and year first full report of study was published e.g. Smith 2001</i>) |
| |
| Report IDs of other reports of this study (<i>e.g. duplicate publications, follow-up studies</i>) |
| |
| Notes: |
| |

1. General Information

| | |
|--|--|
| 1. Date form completed (<i>dd/mm/yyyy</i>) | |
| 2. Name/ID of person extracting data | |
| 3. Report title (<i>title of paper/ abstract/ report that data are extracted from</i>) | |
| 4. Report ID (<i>if there are multiple reports of this study</i>) | |
| 5. Reference details | |
| 6. Report author contact details | |
| 7. Publication type (<i>e.g. full report, abstract, letter</i>) | |
| 8. Study funding source (<i>including role of funders</i>) | |
| Possible conflicts of interest (<i>for study authors</i>) | |
| 9. Notes: | |

2. Eligibility

| Study Characteristics | Review Inclusion Criteria (<i>Insert inclusion criteria for each characteristic as defined in the Protocol</i>) | Yes/No/Uncl ear | Location in text (<i>pg & ¶/fig/table</i>) |
|--------------------------------------|---|------------------------|--|
| 10. Type of study | | | |
| 11. Participants | | | |
| 12. Types of intervention | | | |
| 13. Types of outcome measures | | | |
| 14. Decision: | INCLUDED | | |
| 15. Reason for exclusion | | | |

| Study Characteristics | Review Inclusion Criteria <i>(Insert inclusion criteria for each characteristic as defined in the Protocol)</i> | Yes/ No / Uncl ear | Location in text <i>(pg & ¶/fig/table)</i> |
|------------------------------|---|---------------------------------------|--|
| 16. Notes: | | | |

DO NOT PROCEED IF STUDY EXCLUDED FROM REVIEW

3. Population and setting

| | Description <i>Include comparative information for each group (i.e. intervention and controls) if available</i> | Location in text <i>(pg & ¶/fig/table)</i> |
|---|---|--|
| 17. Population description <i>(from which study participants are drawn)</i> | | |
| 18. Setting <i>(including location and social context)</i> | | |
| 19. Inclusion criteria | | |
| Exclusion criteria | | |
| 20. Method/s of recruitment of participants | | |

4. Methods

| | Descriptions as stated in report/paper | Location in text <i>(pg & ¶/fig/table)</i> |
|---|---|--|
| 21. Aim of study | | |
| 22. Design <i>(e.g. parallel, crossover, non-RCT)</i> | | |
| 23. Start date | | |
| 24. End date | | |
| 25. Duration of participation <i>(from recruitment to last follow-up)</i> | | |

5. Participants

| | Description as stated in report/paper | Location in text (pg & ¶/fig/table) |
|--|---------------------------------------|--|
| 26. Sample size | | |
| 27. Age | | |
| 28. Sex | | |
| 29. Race/Ethnicity | | |
| 30. Severity of illness | | |
| 31. Co-morbidities | | |
| 32. Other treatment received <i>(additional to study intervention)</i> | | |
| 33. Other relevant socio demographics | | |

6. Results

| | Description as stated in report/paper | Location in text (pg & ¶/fig/table) |
|--|---------------------------------------|--|
| 34. IPQ Type | | |
| 35. Comparison | | |
| 36. Subgroup | | |
| 37. No. missing participants and reasons | | |
| 38. Any other results reported | | |
| 39. Statistical methods used and appropriateness of these methods <i>(e.g. adjustment for correlation)</i> | | |
| 40. Results | | |
| 41. Reanalysis required? <i>(if yes, specify why)</i> | | |
| 42. Reanalysis possible? | | |

| | Description as stated in report/paper | Location in text (pg & ¶/fig/table) |
|------------------------|---------------------------------------|--|
| 43. Reanalysed results | | |

7. Applicability

| | | |
|--|--|--|
| 44. Have important populations been excluded from the study? <i>(consider disadvantaged populations, and possible differences in the intervention effect)</i> | | |
| 45. Does the study directly address the review question? <i>(any issues of partial or indirect applicability)</i> | | |

8. Other information

| | Description as stated in report/paper | Location in text (pg & ¶/fig/table) |
|--|---------------------------------------|--|
| 46. Key conclusions of study authors | | |
| 47. References to other relevant studies | | |
| 48. Correspondence required for further study information <i>(what and from whom)</i> | | |
| 49. Further study information requested <i>(from whom, what and when)</i> | | |
| 50. Correspondence received <i>(from whom, what and when)</i> | | |

Appendix D: Adapted Newcastle-Ottawa Scale

| | Rating |
|--|---|
| <p>Selection (max. 5 stars)</p> <p><u>1. Representativeness:</u></p> <p>a. Truly representative of adults with a sudden onset neurological condition? (all subjects or random sampling) ★</p> <p>b. Somewhat representative (non-random sampling) ★</p> <p>c. Selected group of users</p> <p>d. No description</p> <p><u>2. Sample size:</u></p> <p>a. Is the sample size adequate and justified? ★</p> <p>b. Not justified</p> <p><u>3. Non-respondents:</u></p> <p>a. Is comparability between respondents and non-respondents established and response rates satisfactory? ★</p> <p>b. Comparability between respondents and non-respondents established and response unsatisfactory.</p> <p>c. No description of response rates or characteristics of responders/non-responders</p> <p><u>4. Ascertainment of the exposure:</u></p> <p>a. Is the exposure measured using a validated tool? ★</p> <p>b. Non-validated tool but the tool is described.</p> <p>c. No description of measurement tool.</p> <p><u>5. Was the IPQ administered as recommended by the developers?</u></p> <p>a. Yes ★</p> <p>b. No</p> | <p>1)</p> <p>2)</p> <p>3)</p> <p>4)</p> <p>5)</p> |
| <p>Comparability:</p> <p>a. The study controls for injury variables and demographic factors? ★ ★</p> <p>b. The study controls for demographic factors only. ★</p> <p>c. No evidence of controlling for confounding factors.</p> | |
| <p>Outcome:</p> <p><u>1. Assessment of outcome</u></p> <p>a. Was assessment of outcome confirmed by secure records & self-report? ★</p> <p>b. Self-report only.</p> <p>c. No description.</p> <p><u>2. Statistical tests:</u></p> <p>a. Statistical tests clearly described and appropriate, the measure of association is presented, including confidence intervals and probability value. ★</p> <p>b. The statistical test is not appropriate, described or incomplete.</p> <p><u>3. Were all dimensions of the IPQ reported?</u></p> <p>a. Yes ★</p> <p>b. Partially</p> <p>None</p> | <p>1)</p> <p>2)</p> <p>3)</p> |
| <p>8-10 stars = Very Good, 7-5 stars = Good 4 stars = Satisfactory 0-3 stars = Unsatisfactory</p> | |

Chapter 2: Empirical paper

The post-stroke emotionalism cognition questionnaire: a development study.

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Abstract

Purpose: Outbursts of involuntary laughing or crying is a common consequence of stroke and is called post-stroke emotionalism. Little is known about the psychological consequences of the disorder. This study explored the lived experiences of people with emotionalism and developed a questionnaire based on their narratives to assess beliefs about the condition. This questionnaire could be used in future to understand individuals' perspectives on the impact of emotionalism and identify those who may benefit from further support to manage their symptoms.

Methods: Eighteen semi-structured interviews were conducted using framework analysis.

Results: Four global themes were found; the sudden and uncontrollable nature of emotionalism; incongruence; the stigma of expressed emotion and convalescence. These themes formed the basis of the questionnaire, entitled the Post-Stroke Emotionalism Cognitions Questionnaire (PEC-Q).

Conclusions: The social and emotional impact of emotionalism was crucial in understanding how participants made sense of the condition. People with negative experiences described heightened disability, a loss of valued roles and social withdrawal. Positive experiences were shaped by a greater understanding of emotionalism, an increased sense of control over symptoms, social support and maintaining hope about recovery. Implications for

rehabilitation, and recommendations for the future validation and use of the beliefs questionnaire were discussed.

Keywords: Post-stroke emotionalism, stroke, patient-reported outcome measure, qualitative, cognition questionnaire

Introduction

Stroke is the leading cause of disability worldwide, the fourth leading cause of death and affects over 1.2 million people in the United Kingdom [1]. There are many emotional consequences of stroke but one of the most dramatic is that of emotionalism [2]. Emotionalism is an acute neurological disorder, characterised by sudden, involuntary and uncontrollable episodes of crying or laughing [3]. In some instances, laughter or crying is incongruent to the person's underlying emotional state. On other occasions, the emotional reaction is an exaggerated response to emotional stimuli [4].

The prevalence and aetiology of emotionalism

The aetiology of emotionalism is strongly associated with cerebral pathology [5]. The disorder occurs secondary to a wide range of neurological conditions like stroke, multiple sclerosis, dementia, amyotrophic lateral sclerosis and traumatic brain injury [6]. According to a recent meta-analysis, the prevalence of emotionalism is estimated to be 17% at the acute phase (<1 month after stroke) and 20% at the post-acute phase (1-6 months) of stroke [7]. However, the number of well-conducted prevalence studies is low and further work is needed to obtain better prevalence estimates.

Studies suggest that emotionalism arises from lesions to the frontal lobes and descending corticobulbar-cerebellar circuits that regulate motor control and the co-ordination of emotional expression [8,9]. Impaired communicating

pathways from frontal and cortical motor inputs are thought to disrupt the cerebellum's ability to modulate the motor expression of emotion [10]. Maruzairi and Koh argue that emotionalism arises from dysfunctional serotonergic and glutaminergic neurotransmission in the cerebellum, thought to play an important role in emotion processing [11]. Disrupted neurotransmission is theorised to lead to a loss of, or reduction in, a patient's voluntary control over their emotional expression [10].

The pathophysiology of emotionalism is not entirely understood and there remains inconclusive links between lesion location and neuropathology [2,10]. However, it is recognised that stroke-related disruption to the pathways between frontal, parietal and brain stem regions typically leads to involuntary laughter or crying, characteristic of emotionalism [6].

The crying variant of emotionalism is frequently mistaken for clinical depression [8, 9] Diagnosis is complicated by the fact that emotionalism can co-occur with mood disorders [2] Yet, both are distinct conditions. Crying caused by emotionalism is brief and uncontrollable where there is a lack of depressive beliefs associated with crying [6, 10, 12]. In the case of depression, crying is prolonged and patients report depressive beliefs relating to a sense of hopelessness, worthlessness and despair [13].

Current treatments for emotionalism

National guidelines recommend anti-depressant medications as the first line of treatment for emotionalism [14]. However, the overall effectiveness of medications in symptom reduction is rather weak [15,16]. Behavioural treatments like competing response training, an intervention which involves learning to anticipate the onset of symptoms and employing movements to counteract symptoms, have shown promising results [17]. Distraction and relaxation training have been seen to reduce the emotional distress associated with symptoms [5]. However, non-pharmacological interventions are still in their infancy in this population and further empirical studies are required.

The psychosocial impact of emotionalism

Almost nothing is known about the persistence of emotionalism or about the factors that exacerbate symptoms. The longer-term social and functional impact is under-studied and existing research relies heavily on cross-sectional case studies [18,19,20]. The condition has been associated with distress, limited social participation and a reduced quality of life [11,21, 22, 23, 24]. Yet, the factors that precipitate these outcomes are not yet understood.

Rationale for the current study

Individuals with emotionalism are at heightened risk of developing mood disorders, presumably due to the socially isolating nature of symptoms [3,25].

Post-stroke depression impedes functional recovery, reduces a person's quality of life and increases their risk of mortality [26, 27]. Post-stroke anxiety has similarly poor outcomes [28]. To date, the psychological impact of emotionalism is under-investigated. It is not known why some individuals develop mood disorders whilst others do not.

The way that a person conceptualises their illness can significantly influence their recovery from stroke [29,30]. According to Beck, it is the *meaning* that the person ascribes to an event rather than the *nature* of the event itself that determines their coping responses [31]. Indeed, research consistently indicates that patient beliefs predict coping behaviours [32], treatment adherence [33], social functioning [34], emotional well-being and quality of life [35] in chronic health populations.

It's hypothesised that a person's beliefs about emotionalism may contribute to some of the negative outcomes described in the literature. For instance, a person who endorses negative assumptions about emotionalism (i.e. "*crying is weak or childish*") may think critically of themselves, avoid activities that trigger symptoms and withdraw from others for fear of negative evaluation. Avoidance and social isolation are key determinants of poor psychosocial outcomes after stroke [27,36]. Avoiding activity can lead to a loss of achievement or a sense of mastery, consistent with behavioural models of depression. Similarly, avoiding social contact is likely to maintain a person's anxiety by preventing habituation to the social and cognitive triggers of

emotionalism. Clearly, a greater understanding of the role of patient beliefs is warranted as these relationships are speculative.

Eliciting patients' assumptions about emotionalism may lend insight into how they are managing their symptoms. The development of a questionnaire that assesses patient beliefs about emotionalism could be used to identify those who hold maladaptive beliefs who may be vulnerable to avoidance, distress or poor outcomes. Questionnaires that measure patient beliefs are often created solely on clinical expertise or on the existing literature. This method of assessing illness risks missing features of the experience that the patient deems to be important [37]. Patient-reported outcome measures (PROMs) are gaining increasing recognition as a way of measuring the impact of illness based on patients' perspectives [38,39]. To develop a patient-reported outcome measure, one must know of the important aspects of the experience in a target population. There is a paucity of research investigating patients' perspectives on emotionalism and to our knowledge, no qualitative studies exist.

Primary research objective

This qualitative project aimed to investigate the lived experiences of people with emotionalism after stroke. It further aimed to identify important aspects of the experience of emotionalism to inform the development of a measure of beliefs associated with the condition. This study was the first stage of the questionnaire development.

Method

Design

The study used a purposive sampling design and aimed to recruit individuals with emotionalism across a wide range of ages, stroke classifications and at different stages of recovery. Data were analysed using framework analysis, a form of thematic analysis that focuses on relationships within the data to draw conclusions based on themes [40]. This method was chosen based on its flexibility and systematic methods of analysing and interpreting data [41].

Ethical considerations

Ethical approval was granted by the West of Scotland Research Ethical Committee, NHS Lothian Research and Development and the University of Edinburgh Clinical Psychology Research Ethics Committee (see *Appendix E*).

Inclusion and exclusion criteria

Participants were eligible for inclusion if they had a primary diagnosis of stroke and probable emotionalism as defined by the ICD-10 diagnostic criteria (see *Appendix F*) Participants were required to have sufficient cognitive and language abilities as well as no known significant psychiatric condition or neurodegenerative disorder to take part in the study.

Screening measures

Patients are routinely screened for functional and cognitive impairments as well as for the presence of mood disorders as part of service delivery in NHS Lothian acute stroke settings. This is as recommended by SIGN clinical guidelines [14]. The Barthel Index (BI) is a 20-item, valid and reliable measure of activities of daily living [42]. It is commonly used to assess for functional independence after stroke [43]. The Addenbrooke's Cognitive Examination (ACE-III) is a well-established measure of cognitive functioning [44]. The Hospital Anxiety and Depression Scale (HADS) is a 14-item self-report questionnaire measuring symptoms of anxiety and depression [45]. This questionnaire has acceptable psychometric properties in stroke [46]. All participants completed these measures in advance of the study as part of their routine care. This information was used for descriptive purposes and to confirm participant eligibility for inclusion in the study.

Participants

Of the 29 participants who were invited to take part in the study, four participants did not meet the ICD-10 diagnostic criteria for emotionalism and instead were diagnosed with depression. Two participants declined to take part, two had severe communication difficulties and three participants had cognitive impairments that interfered with their ability take part in an interview. A total of eighteen participants took part in the study.

Procedure

Participants were recruited via stroke specialist physicians, consultant neurologists, clinical psychologists and nurses from three NHS acute stroke units and one NHS post-acute stroke service. Members of the stroke teams identified individuals who met the ICD-10 criteria for emotionalism and approached them to introduce the study. Once potential individuals expressed an interest in taking part, they met with the lead researcher (NM) who explained the details of the study and provided them with an information sheet and consent form (*Appendix G*). Participants were given a week to decide if they wished to take part. If they consented, a suitable time for the interview was arranged.

When consenting to take part, the participants gave permission for the researcher to access their medical notes to ascertain their functional status (Barthel Index score), cognitive functioning (ACE-III score) and mood questionnaire score (HADS). See the research protocol in *Appendix I* for details.

Development of the interview guide

The semi-structured interview guide was based on guidelines outlined by Ritchie and colleagues [41]. Given the exploratory nature of the study, the interview guide was used flexibly to prevent constricting participant narratives [47]. Questions were omitted, altered or explored in more detail dependent on

the individual [41]. The guide was developed based on the literature investigating patient beliefs in clinical health populations [48] and in stroke [49, 30]. The guide was reviewed by five stroke specialists (3 consultant stroke physicians, 1 consultant geriatrician and 1 clinical neuropsychologist) and amended based on their recommendations.

Figure 4. Semi-structured interview guide

Questions based on the literature:

- In what way has your ability to control your emotions changed since your stroke?
- How able do you feel that you can control your emotions?
- What do you think are the causes of your emotionalism?
- What impact does emotionalism have on your life?
- How long do you believe that your emotionalism will last for?

Questions based on expert opinion:

- How do you feel when you become emotional?
- What thoughts are passing through your mind when you become emotional?
- How do you cope when you become emotional?
- What do you think that other people are thinking when you're emotional in public?
- Is there anything that you would do but can't because of emotionalism?
- What advice would you give to someone who has emotionalism?

Data Analysis

Framework analysis uses a matrix-based approach to categorise and organise data according to emergent themes on an individual and group basis [41]. The seven stages of framework analysis were followed as recommended by Ritchie, Spencer and O'Connor [50]. Analysis was an iterative process, where the researcher moved back and forth between stages to consolidate the final themes.

Stage 1: Transcription

All interviews were transcribed by the researcher verbatim. Two transcripts were reviewed by an independent researcher to confirm the researcher's fidelity to the recordings. Field notes on non-verbal forms of communication and the researcher's reflections on the interview process were also documented at this stage.

Stage 2: Familiarisation

Familiarisation is a universal concept across all qualitative methods, often referred to as "immersion" [51]. Listening to the interviews again reminded the researcher of the emotional tone of the interviews as well as of the severity and duration of participant's emotional reactions. Re-reading transcripts and noting preliminary codes or reflections primed the researcher for the next stage of analysis.

Stage 3: Coding

The first six transcripts were analysed by assigning statements or “codes” to each line (see *Appendix I*). Using Gibb’s guidelines, codes were initially developed by summarising what each participant was describing [52]. Participants’ own words were used in order to stay true to the data at this initial stage [41]. Notes as to the reasons why codes were created and reflections on how codes were linked, were described in the form of memos and attached to the transcripts. Over time, the process of coding became more categorical where multiple statements were classified under one code. For instance, a line of text describing a person’s inability to work after stroke and another line referring to a person’s inability to walk after stroke would be defined under the same code as “loss”. These analytical codes were clustered into preliminary themes and used to create a coding matrix as per Gale *et al.* [40].

Stage 4: Developing a working analytical framework

The coding matrix and interview guide were then used to develop an analytical framework that aimed to categorise the data according to emerging themes [53]. This was an iterative process of constantly refining the framework by clustering passages of text and re-reading transcripts (see *Appendix J*).

Stage 5: Indexing

The analytical framework was piloted on two transcripts where passages of text were labelled with one or more of the framework themes and refined as new insights emerged. The final framework was then applied to all subsequent transcripts using a computerised qualitative software package known as MAXQDA (<http://www.maxqda.com/products/maxqda-standard>).

Stage 6: Charting

Data was charted onto a matrix Excel spreadsheet and categorised based on the framework themes. The matrix comprised of one column per participant and one row per framework theme. Conceptual ideas were then abstracted from within each framework theme and between each participant [41]. The final analytical accounts were summarised using verbatim text.

Stage 7: Developing explanatory accounts and the questionnaire

At this stage of analysis, the emphasis moved beyond descriptive accounts of individual narratives towards a wider understanding of the emotionalism experience. A questionnaire was developed based on the final analytical framework, entitled the Post-Stroke Emotionalism Cognition Questionnaire (PEC-Q) (see *Appendix K*).

Ensuring methodological rigour

A reflective diary was kept throughout the interview process to log theoretical ideas, decisions about codes and the dynamics between interviewees and interviewer. This was to provide transparency in the researcher's perspective on the sensitive content of interviews. The diary was also used to document the researcher's experiences and assumptions about stroke and emotionalism as well as the influence that this had upon their interviewing style and approach to the analytical process (see *Appendix L*).

Multiple coding with two separate researchers was used on four transcripts. This was in order to be transparent with the progressive development of themes and to strengthen the reliability and validity of the analysis process.

Consensus Expert Panel

A summary of the final analytical framework and a draft of the PEC-Q was sent to participants who requested to be contacted (see *Appendix M*). A draft of the questionnaire was also sent to a panel of stroke specialists for review. This was to ensure respondent validation of the themes as well as the content and face validity of the emotionalism questionnaire.

Results

Eighteen single session interviews were conducted and audio recorded on an encrypted device. Interviews were transcribed verbatim. The length of interviews ranged from 12-60 minutes ($M=35.3$ mins $SD=10.9$). The duration of interviews depended on environmental variables (i.e. busy inpatient wards), the participant's physical ability and the depth to which they wished to explore their experiences.

Demographic information

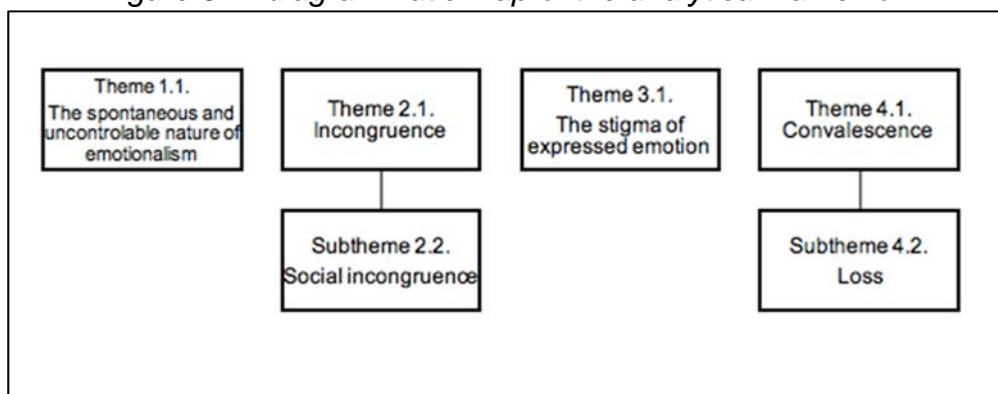
Six males and twelve females were recruited to the study (age range= 39-81 years, $M=58.9$ yrs, $SD=10.4$). Six participants were inpatients in a NHS stroke ward and twelve were interviewed in their own homes. The length of time since stroke ranged from 2 weeks to 17 months ($M=4.3$ months $SD=3.7$). The sample consisted of four haemorrhagic and fourteen ischemic strokes. Of these, ten were right-sided, three were left-sided and five were bilateral strokes. The functional ability of individuals varied from needing total support for all aspects of daily living (Barthel Index=1) to being fully independent (Barthel Index =20). Three individuals scored in the clinical ranges for mild depression and anxiety and were receiving psychological therapy. One participant was from Italy, another from South America and the remaining participants were from the United Kingdom.

Analysis was completed in two phases. Phase 1 reported on the themes reflecting participants' experiences of emotionalism. Phase 2 outlined the development of the questionnaire based on these themes.

Analysis phase 1: Emergent themes

Four global themes and two subthemes concerning the experience of emotionalism emerged. Although these themes overlap to some degree, they represent a mapping of the most salient aspects of participants' experience.

Figure 5. A diagrammatic map of the analytical framework



Theme 1.1: The spontaneous and uncontrollable nature of emotionalism

This theme reflected participants' attempts to make sense of emotionalism. Participants frequently referred to their outbursts of laughter or crying as occurring for no "*rhyme or reason*" (Participant 12:10).

(Participant 17: L2-4)

"I get no warning, it just... one minute I'm ok and the next minute I just burst into tears ... it

could be a happy thing, it can be a sad thing or it's just for no reason."

(Participant 5: L165)

"I laugh or cry for no reason."

(Participant 16: L2)

"My emotions would normally work at the right time but now they're just sporadic it just happens, it could be triggered by a word, triggered by looking at somebody or hearing somebody's name."

Some participants described their laughter or crying as being exaggerated responses to mildly emotive triggers and that their emotions always feel *"close to the surface"* (Participant 6: L30). Emotive triggers included talking about family members, seeing grandchildren or watching a television programme.

(Participant 4: L8)

"If I get a nice text or I get a well wish or something. Somebody's thinking about me, I just burst into tears."

Many participants seemed to be unaware that their stroke caused emotionalism. In the absence of a medical reason, participants attributed their emotional reactions to different causes. Some assumed that their uncontrollable laughter or crying was a sign that their mental state was deteriorating. Understandably this was incredibly distressing for them.

(Participant 3: L167)

"I thought I had em ... lost my marbles."

(Participant 17: L290)

"I thought I was having a nervous breakdown, I thought I was really cracking up."

Others believed that their uncontrollable reactions were an unconscious expression of coping and adjusting to stroke. Understanding emotionalism in this way seemed to be less distressing for participants.

(Participant 14: L150)

"I assumed it was just coming to terms with what I couldn't do and realising it."

(Participant 7: L27)

"I suppose either laughing or crying, it must be me just trying to cope with things. I think that's a way it comes."

(Participant 6: L159)

"I was just told that that's just what happens when you've had a stroke."

All participants were unable to control their laughter or crying to some degree. The emotional impact of this lack of control varied according to their beliefs about the severity of symptoms and their pre-stroke assumptions or values.

(Participant 8: L24)

"It's irritating more than anything else...you're not in control when you're overly emotional."

A number of participants believed that the condition had damaged their self-confidence. There were many references to feelings of embarrassment and frustration at being unable to control laughter or crying.

(Participant 12: L216)

"I get embarrassed when I cry cause like I say there's nothing I can do about it."

Theme 2.1: Incongruence

The juxtaposition between participants' internal state and their external emotional responses was described as “*confusing*”, “*worrying*” and “*strange*”. In their attempts to identify the reasons for their reactions, participants often experienced confusion and uncertainty.

(Participant 14: L62)

“There’s nothing emotional... there’s nothing inside me that feels bad, I don’t feel... I don’t feel stressed, I don’t feel any of that kind of thing. I just feel I’m about to cry and no sort of work up toward it.”

(Participant 4: L301-306)

“You know yourself and you know when you come across situations, you know how you’re going to react. You know how your mind works ... but it’s almost like when you’ve had a stroke, you have no control of your mind anymore ... with the giggles or the tears it comes from nowhere, out of character, not the norm and you think “oh where has that come from, what’s that all about?”

Emotionalism seemed to be more distressing for participants who rarely cried before their stroke. They found the discontinuity between their emotional selves before and after stroke to be frightening.

(Participant 2: L7)

“Tears and everything running down my face... and that’s not me ... I don’t get emotional.”

(Participant 6: L26-28)

“It’s scary cause you’re not used to that ... It just isnae me.”

Participants who held strong beliefs about their sense of independence and strength severely criticised their inability to control their emotions. They felt that the expression of emotion was a “*sign of weakness*” (Participant 10: L84) and that individuals should be “*mentally in control and... disciplined*” (Participant 8: L227). Individuals with high achieving jobs who lived alone and described themselves as strong before their stroke particularly identified with these beliefs.

Subtheme 2.2: Social incongruence

The symptoms of emotionalism were rarely triggered by internal emotions. Nonetheless, the public responded to participants’ emotional responses as though they were genuine. This incongruence was “*intensely embarrassing*” for some people.

(Participant 14: L34-35)

“Sometimes you feel really stupid you know, sitting there and tears are falling down and you’re trying to ... and people are all fussing about you because they think there’s something wrong and you just keep saying no I’m absolutely fine I just can’t stop crying for some reason or another.”

Often participants felt “*ashamed*” of their inability to control their emotional reactions in public. They voiced many critical beliefs about other people’s perceptions of them.

(Participant 9: L176-177)

“They give you this expression, it’s like “oh look at her she’s crazy” ... or they’re thinking maybe

you're high on drugs or something like that because it's not normal (for) a person who has nothing to laugh (at to be) laughing right?"

(Participant 13: L248)

"I think people get a different opinion of you because they see it as something wrong with you... that you're not coping with life ... and I don't want that to be my life."

(Participant 10: L210)

"Other people probably see them as being weak and feeling sorry for themselves. You know eh... not strong."

The hidden nature of emotionalism was often attributed to be the reason for why people misinterpret their reactions.

(Participant 17: L87-88)

"Because I look ok, I don't physically have anything to see from the stroke, folk say 'well you look ok'. God help me if I look ok em... (I) don't look like (I've) had a stroke but em ... I feel that I have had a stroke."

Compassion or empathy from others seems to exacerbate emotional reactions. Many participants mentioned that they become emotional when people show concern for their well-being.

(Participant 13: L42-43)

"I cry more when somebody says something nice to me... tries to help me...or be kind."

Some participants felt guilty about the impact that emotionalism has on other people. This was salient for individuals who held beliefs about distressing others, particularly their friends and family members.

(Participant 13: L290).

“I don’t want to upset other people and make them feel that they’ve said something wrong.”

Many participants engaged in subtle avoidances to cope with incongruence. These included only going out with family members, avoiding certain topics in conversation or busy environments. For some, the incongruence associated with emotionalism felt so disabling that they rarely left their home.

(Participant 7: L21)

“If I’m on my own then nobody thinks I’m crazy.”

Theme 3.1: The stigma of expressed emotion

Multiple references to social stereotypes were made when discussing the public perceptions of those who are emotional. Some participants believed that the older generations are more “*sympathetic*” or “*compassionate*” towards tearfulness. These participants believed that older individuals assume that they are grieving and can relate to this better than younger people. Male participants also often referred to societal assumptions about gender and beliefs about how men should not cry.

(Participant 18: L350-351)

“It’s something that men don’t do... show their emotions. Men were always felt to be the stronger ones”.

The laughter variant of emotionalism is less prevalent [7]. Participants reflected on how people easily misinterpret their laughing as a sign of humour and will laugh alongside them or encourage further laughter. This was often very stressful or anxiety provoking for participants. One individual conveyed the disabling nature of her inappropriate laughter during a time where a passer-by misperceived her laughter and threatened her. She found this event incredibly frightening and now rarely leaves her home.

(Participant 9: L58-159)

“I was walking and laughing my head off why I don’t know, [laughs] but the guy behind me, he thought that I was laughing with him or about him and then (he) stopped me and gave me abuse.”

Theme 4.1: Convalescence

This theme reflected the process of recovering from stroke and emotionalism. Here, participants convey the grief associated with their loss after stroke and their struggle in reconciling their new emotional selves with their past identities. This theme further illustrated participants' coping responses and their determination to regain a sense of emotional equilibrium.

The majority of participants reported noticing gradual improvements in their ability to manage their emotions as they recuperated from stroke. A desire to regain continuity or "*getting back to normal*" was common in those who were at the earlier stages of stroke. Participants reflected on the challenge of adjusting their expectations when they did not fit with the reality of their recovery. One participant believed that she is "*getting better, it's just no... as good as (she) would've thought it would've been at this stage*" (Participant 6: L429). Those who were in hospital with lower functional abilities regarded their physical recovery to be more of a priority than emotionalism.

(Participant 2: L102).

"I'm thinking how I can get myself better. All the things I can do, physio-wise... this is more physical for me."

The impact of emotionalism became more distressing as individuals progressed in their stroke trajectory and experienced a plateau in their functional abilities.

(Participant 11: L173-174)

“You’re concentrating I think after the stroke on the physical side you know and getting yourself back and trying to do things and what have you that you... its only after that that... the emotional side kicks in and you start to realise the different problems it’s causing.”

Participants developed various methods of coping with emotionalism. Some strategies were helpful like humour, distraction or to just simply *“let it out.”*

(Participant 4: L276). Others means of coping were unhelpful, like self-criticism or avoidance.

(Participant 8: L5).

“I’m a tearful fool and a blithering idiot who’s just angry at the world.”

The importance of remaining hopeful was strongly emphasised. Those who held a sense of hope and optimism about their continued recovery felt better equipped to manage the difficulties associated with emotionalism.

(Participant 14: L306-307)

“Just say to yourself, well look this is just something that’s happened and you know why it’s happening, because of your stroke so you just have to try and ride this bit of the storm and see what you can put in play.”

(Participant 9: L246)

“Keep trying but be patient because if you lose patience you lose heart... keep cheerful.”

(Participant 9: L246)

“Try to find happiness even... if the day is dark, tomorrow might be a better day. “

Subtheme 4.2 - Loss

Loss was a dominant theme in participants' narratives. For some individuals, this reflected their experiences of a loss of their former emotional strength. For others, it signified a loss of confidence and uncertainty about their future. The saliency of this theme related to the participant's stage of recovery. Those interviewed in the inpatient wards spoke of the loss of their physical functioning and independence. Participants reflected on the challenges of now having to depend on others where they previously were independent. Many individuals at this stage also voiced uncertainty about their futures and fears around sustaining another stroke.

(Participant 4: L184-185)

"You know if somebody could say to you like if you come in with a broken arm you know in three months' time it'll heal, it'll be fine. But this is the unknown."

(Participant 18: L56)

"I'm terrified about having another stroke, that would be awful."

Participants interviewed later into their stroke recovery spoke of losing their sense of place in society. One participant admitted that she believed that she "would never be needed" (L488) by her family after her stroke and that she feels "a bit useless." (L103). Loss of occupation was difficult for participants. Many considered their professions to be a defining feature of their personality.

(Participant 9: L198)

“I’m not in work for a year now, I’m no longer able... no job, I feel I’m not good for nothing.”

(Participant 12: L670)

“I can’t do my job now, it totally destroys me.”

For other participants, loss was in the context of physical recovery. For one participant, her loss of mobility was devastating. Being in a wheelchair makes her feel “*dependent*” and “*stuck*” as she can no longer walk away from situations like before. This loss threatened her assumptions about her autonomy and control before her stroke.

(Participant 10: L35-39)

“I feel as though I’ve lost all control of my life. Before, I always felt I knew where I was going and I was eh, in my own home. I had my own social life and I was in control of everything and I felt I had something to offer.... people would listen to me. But now, I feel as if I’m ... just overlooked, not here.”

Table 4. Summary of key findings from the analytical framework

| Theme | Key findings |
|--|---|
| <p>1.1 The spontaneous and uncontrollable nature of emotional reactions</p> | <p>Emotionalism is sudden and uncontrollable. Participants struggled to understand their emotional reactions. Some individuals attributed their emotional reactions to adjusting to stroke. Others believed that their uncontrollable laughter or crying reflected a deterioration in their mental health and found this to be very distressing.</p> |
| <p>2.1 Incongruence</p> | <p>Most individuals spoke of how being emotional is not in keeping with who they were before their stroke. This incongruence led many individuals to question their own self-concept which was detrimental to their psychological well-being.</p> <p>Subtheme: Social incongruence - Participants described how other people react to their emotional reactions as though they are genuine. Many participants spoke of the embarrassment and shame of being emotional in public and the guilt of upsetting others around them.</p> |
| <p>3.1 The stigma of expressed emotion</p> | <p>Participants described the influence of culture, age and gender stereotypes on how they cope with emotionalism.</p> |
| <p>4.1 Convalescence</p> | <p>Participants spoke of rebuilding their lives and accommodating emotionalism. Many used strategies like distraction, humour and maintaining optimism or social support. Some participants have become socially withdrawn since their stroke and avoid others due to embarrassment.</p> <p>Subtheme: Loss- Many forms of loss were discussed. Grief and despair were often reflected in the narratives.</p> |

Analysis phase 2: Development of the questionnaire

The second phase of analysis concerned the development of the PEC-Q. Items for the questionnaire were developed based on the four themes from the analytical framework. The wording of statements was derived from participants' descriptions of their experiences or adapted slightly to better reflect the themes (see *Table 5*). This is in keeping with similar studies that have developed measures based on patients' perspectives [39,54]

Questionnaire statements were organised into four sections based on the global themes. A 5-point Likert scale was used to score responses along a continuum of beliefs about emotionalism from “*strongly disagree*” (0) to “*strongly agree*” (5). The middle position was labelled “*neither agree or disagree*” to reflect a neutral stance rather than an inability to answer the question [55]. Higher scores on the questionnaire indicated more pessimistic beliefs. Reverse scoring was applied to both statements of Theme 4.1 “*convalescence*” as higher scores reflected more positive beliefs (see *Appendix K* for scoring details).

Expert consultation on the face validity of the PEC-Q

The final analytical framework and a draft of the PEC-Q were sent to all participants who agreed to be contacted (see *Appendix K*). The questionnaire was also sent to three stroke specialists for review (one consultant geriatrician, one consultant stroke physician and one consultant neurologist). Feedback

from participants consisted of re-wording four of the statements. One consultant suggested revising the statement *“I should be in control of my emotions at all times”* to a statement that reflected what it may mean for the person if they were unable to be in control. This item was changed to *“my inability to control my emotions is a sign that I am losing my mind”* as this belief was reported by participants when describing the personal meaning of their loss of control.

Table 5. Phase 2 questionnaire item development

| Theme | Quotes illustrative of themes | PEC-Q statement |
|---|--|--|
| 1.1 The spontaneous and uncontrollable nature of emotional reactions | <i>“I cannot control this crying and laughing.”</i> (P9) | (1) “I have no control over my laughter or crying.” |
| | <i>“I’ve no ability at all to control emotions.”</i> (P18) | |
| | <i>“You can’t stop it.”</i> (P3) | |
| | <i>“It’s something you can’t control.”</i> (P7) | |
| | <i>“I feel I’m just going crazy”</i> (P12) | (2) “My inability to control my emotions is a sign that I am losing my mind.” |
| | <i>“Thought I was cracking up”</i> (P17) | |
| | <i>“There’s something psychologically wrong with me”</i> (P3) | |
| | <i>“It’s like I’ve lost my mind”</i> (P15) | |
| | <i>“I can’t handle my emotions anymore.”</i> (P6) | (3) “When I become emotional, I feel unable to cope.” |
| | <i>“I’m not able to cope with all of these emotions”</i> (P17) | |
| | <i>“I feel like my emotions are taking over.”</i> (P9) | |
| | <i>“Feeling of shame, because this is not me.”</i> (P9) | |
| 2.1 Incongruence | <i>“I’ve never been one to cry. No, things never easily upset me normally. I would say it just isn’t me.”</i> (P6) | (4) “Being emotional just isn’t me.” |

| | | |
|--|---|---|
| | <p><i>"Cause it's not me... it's totally out of character for me." (P2)</i></p> <p><i>"It's so alien to who I am." (P14)</i></p> | |
| | <p><i>"I don't want people to look at me and think that I'm a weak person who cries about everything." (P9)</i></p> <p><i>"I see it as a sign of weakness." (P11)</i></p> <p><i>"You feel weak and debilitated. You don't feel you're in control. You're just weak." (P8)</i></p> <p><i>"I'm being weak...I'm not a weak person." (P2)</i></p> | <p>(5)</p> <p>"I feel weak when I am emotional."</p> |
| | <p><i>"I try to avoid people or strange situations so that way I won't feel embarrassed." (P9)</i></p> <p><i>"I just need to make sure that I don't get dragged into anything that's going to be emotional, be careful of situations that I get myself into." (P2)</i></p> <p><i>"I avoid talking about subjects that I know will trigger it." (P17)</i></p> <p><i>"There are certain subjects that I tend to avoid, like when people ask you how you are, for example." (P14)</i></p> | <p>(6)</p> <p>"I try to avoid people or situations that may trigger my laughter or crying."</p> |
| <p>3.1</p> <p>The stigma of expressed emotions</p> | <p><i>"I'm worried that they will look at me and say look at that bloody idiot, what's he crying for, silly boy." (P12)</i></p> <p><i>"It did kinda worry me a bit, that maybe I would get extreme at completely the wrong time with strangers who would just think "oh somethings odd about that person". They would probably just think it was some mental deficiency, they might have thought, you know "what's wrong with her?" (P7)</i></p> <p><i>"They're all sorta looking at you thinking, "what's wrong with you." (P17)</i></p> | <p>(7)</p> <p>"I worry that other people will think that there is something wrong with me if I become emotional."</p> |

| | | |
|---|--|---|
| | <p><i>"I was scared of upsetting people. I was scared that people would take it personally."</i> (P3)</p> <p><i>"I would feel bad that I was doing that to them. I don't want people to feeling upset because I'm getting upset."</i> (P13)</p> <p><i>"I don't like crying in front of other people cause they feel bad."</i> (P4)</p> <p><i>"I try not to cry when my husband and folk were around and my daughters and that type of thing because I knew it would upset them."</i> (P14)</p> | <p>(8)</p> <p>"I worry that my laughter or crying will upset other people."</p> |
| | <p><i>"I felt an idiot [laughs] just trying to stop myself, I was so embarrassed."</i> (P17)</p> <p><i>"You feel really stupid."</i> (P15)</p> <p><i>"A mixture of embarrassment and poor me if it was to go through my mind, a total embarrassment."</i> (P10)</p> <p><i>"I get embarrassed when I cry cause like I say there's nothing I can do about it."</i> (P12)</p> | <p>(9)</p> <p>"I feel embarrassed when I become emotional in public."</p> |
| <p>4.1</p> <p>Convalescence</p> | <p><i>"I believe it will get better, I hope so."</i> (P8)</p> <p><i>"Hopefully it'll settle down, I think it will. I'm hoping to find something to make it work."</i> (P14)</p> <p><i>"It has to run its course."</i> (P3)</p> <p><i>"It will get better over time."</i> (P6)</p> | <p>(10)</p> <p>"I believe that my emotionalism will improve over time."</p> |
| | <p><i>"I now know when it's going to happen. I just walk away and pull myself together and come back again."</i> (P8)</p> <p><i>"I just kinda get on with things now regardless of the crying."</i> (P13)</p> <p><i>"I just walk away and shrug it off."</i> (P16)</p> <p><i>"I like using a bit of humour."</i> (P2)</p> | <p>(11)</p> <p>"I have ways to manage my laughter and crying."</p> |

Discussion

This study investigated participants' beliefs about emotionalism to inform the conceptual underpinning and content of a patient-reported questionnaire (the PEC-Q). Framework analysis revealed four main themes: The spontaneous and uncontrollable nature of emotional reactions; incongruence; the stigma of expressed emotions and convalescence. The following is a theoretical discussion of the analytical framework in relation to the evidence-base.

For most participants, the uncontrollable nature of emotionalism was detrimental to their psychological well-being. Individuals that described inflexible assumptions about their loss of control (i.e. "*always being in control*" before their stroke) seemed particularly distressed. Studies have shown that patients who hold a low sense of perceived control over their stroke recovery report greater levels of disability, anxiety and depression [36,56,57]. Findings suggest that supporting patients to adopt ways to manage their emotional reactions may challenge some of their negative beliefs around control and increase their positive perceptions towards recovery.

Few participants understood the causes of their emotionalism. This finding was not surprising given that Picton found even stroke professionals struggle to identify emotionalism consistently [58]. In acute and non-specialist services, outbursts of emotions are often regarded to be part of a "normal" adjustment process thus, emotionalism is easily missed [21]. Poor understanding of a health condition has been associated with greater emotional distress and

social disability [29, 59]. Stack *et al.* found that misconceptions about symptoms delay help-seeking behaviours [60]. This study found that participants often misperceived their uncontrollable emotional reactions and few sought medical advice.

Emotionalism affected participants' self-confidence, their social relationships and interfered with their previously valued roles. The results substantiate and extend previous studies that have found that emotionalism is associated with higher levels of social disability [22] and a reduced quality of life [21, 61, 9].

Individuals in the study referred to being in a constant state of flux between their former and present emotional selves (i.e. "*being emotional just isn't me*"). Ellis-Hill, Payne and Ward argue that a person's self-narrative of their past, present and future selves is ruptured after stroke [62]. In their meta-synthesis, Salter and colleagues identified the person's sense of identity as a crucial part of adjustment [63]. Here the individual engages in a continuous cycle of re-definition as they negotiate their new post-stroke self. The process of reconciling identities was especially challenging for participants who valued their emotional resilience as part of their pre-stroke self. For these individuals, emotionalism violated their assumptions about their identity as being stable and this led to feelings of despair and hopelessness.

Interestingly, participants were unable to explain why other people showing them compassion exacerbated their emotional reactions. To understand why

this may be, it is useful to draw on theories that conceptualise the expression of emotion as a social behaviour.

From an evolutionary perspective, displays of emotion increase the person's chance of survival by communicating their internal feelings to others [64]. People instinctively smile when they see others laugh and respond with empathy when they see someone crying [65]. Attachment theory posits that crying is a care-seeking behaviour and a non-verbal signal of distress [66]. Crying elicits reciprocal care-taking behaviours in other people that are intrinsic to the attachment process [67]. The incongruence between participants' internal emotions (for example, "*crying and I don't feel sad*") and the responses from others may rupture this normally reciprocal exchange. Small displays of emotion shown by others are likely to elicit exaggerated emotional responses in participants (given that emotionalism is a disorder of emotional control). The narratives suggest that participants find this incongruence embarrassing and withdraw or avoid as a means of coping.

Participants made multiple references to stigma and stereotypes that fit with socio-cultural views of the expression of emotion. For example, in Western cultures, autonomy, self-control and the suppression of emotions are seen to be valued attributes [68].

Individuals voiced concerns about societal reactions to their uncontrolled laughter or crying and described the shame attached to these assumptions (for example "*other people think that I'm a weak person who can't cope with*

life”). Shamir and Travis proposed that societal views of masculinity reinforce shame as crying is seen as a sign of weakness or of inferiority [69]. Indeed, many participants referred to the stigma associated with men crying. Gillbert and Proctor described the experience of shame as to feel “*rejectable*” in the eyes of others, creating a desire to hide or avoid [70]. The study found that participants often avoid people, employ safety behaviours (like always being with a family member) or use humour to minimise shame when they become emotional.

Loss was a prominent theme and a key concept in adjustment after stroke [63, 71]. This study reaffirms Dowswell and colleagues’ findings that a sense of grief and loss is felt when the individual compares their past and present stroke selves [72]. The loss of emotional strength, valued roles, autonomy and a sense of purpose described by participants align to other qualitative studies [73, 74]

These findings highlight the importance of considering the stage of a person’s stroke recovery when investigating emotionalism. In acute settings, participants prioritised their functional rehabilitation. Emotionalism was more distressing to participants in the months following discharge, as they began adjusting to their life after stroke. Kirkevold [75] argued that the transition from acute services to home is an important milestone, where the person begins to negotiate the discrepancies between their expectations and the reality of their recovery. This was a critical period for participants and a crucial time for support [76].

Salisbury and colleagues consider successful adjustment to be the process of finding ways to accept the loss of identity and previously defining roles and to create new ones that are valuable and fulfilling [77]. In the past, researchers have argued for the use of bereavement models to understand the traumatic loss in stroke [78, 79]. However, staged models have been criticised for their lack of depth and linear approach to adjustment [80]. Taylor, Todman and Broomfield's social-cognitive transition model emphasises the role of the person's assumptions when adjusting to stroke [26]. These assumptions are mediated by the person's past experiences and social-cultural factors. Indeed, participants in this study who held rigid or inflexible beliefs and who placed high value on autonomy and emotional control seemed to struggle to adjust to emotionalism.

Strengths and Limitations

This study used a convenience sample based on inclusion criteria that excluded individuals with significant cognitive or language impairment. The experiences of those with emotionalism and aphasia or cognitive impairment may be different in the way that they perceive the condition. Four participants in the study did have mild communication difficulties. There was also an unequal gender ratio, although no significant gender differences were reported. Both of these factors may have caused a sampling bias.

Participants were middle-aged from higher socio-economic backgrounds and in employment before their stroke, which is significant given that stroke has a higher incidence and poorer prognosis in those with a lower socioeconomic status [81]. It is possible that younger individuals or those from lower socioeconomic backgrounds hold different assumptions about emotionalism due to a lack of knowledge about stroke or by having less access to support services [82].

Strengths of the project were that it was one of the first studies to investigate the lived experiences of those with post-stroke emotionalism and develop a questionnaire based on patients' perspectives. Further strength lay in the range of participants, reflecting a variety of ages and stages of stroke recovery. The use of framework analysis provided a systematic and clear audit trail which ensured transparency in the generation of themes and the questionnaire items.

Directions for further research

This is the first stage of the development of the PEC-Q to elicit emotionalism-specific beliefs. The next phase of the project would be to pilot test the questionnaire to assess its psychometric properties.

The PEC-Q has the potential to identify patients that hold negative beliefs about their emotionalism who may be more susceptible to distress or to adopting unhelpful ways of coping, like avoidance, which may lead to poor outcomes. These relationships remain theoretical. Further analysis is needed to test out the predictive validity of the questionnaire. This would involve assessing whether negative beliefs (as measured by the PEC-Q) at one time point would predict poorer social or emotional outcomes at another time later into the person's recovery.

It is worth considering the cultural context from which the study sample was drawn. Western cultures hold stricter rules about the individual controllability of emotional expressions [83]. It is possible that emotionalism may be perceived differently across cultures depending on whether the society deems their emotional behaviours to be functional. Though the disorder has been shown to be common in different cultural contexts [22]. This would be an interesting avenue for future research.

Clinical implications

Like any patient-reported outcome measure, the PEC-Q is a means of opening up patient-clinician discussions about the person's subjective experiences. This information could be used to tailor individualised interventions that challenge unhealthy beliefs as well as inform clinicians of areas that the patient is likely to need further support with.

Whilst the evidence-base for the efficacy of psychosocial interventions in emotionalism is lacking, it is a burgeoning area for research [55]. Individual case studies have shown promising preliminary results for reducing the emotional impact of symptoms [5,17]. Should psychological therapy be deemed efficacious in future randomised trials, then the themes from this study may provide possible areas to target. For instance, a cognitive-behavioural approach may be used to challenge the person's self-critical beliefs about their inability to control their emotions or about becoming emotional in public [84]. Behavioural interventions could address a person's social avoidance through the use of graded-exposure techniques [85]. The application of these models requires further investigation.

Lastly, few participants knew that emotionalism was caused by stroke. Individuals do not often seek medical advice for emotionalism as they find their symptoms embarrassing or shameful [86]. Current clinical practice guidelines recommend providing information and advice early to patients with emotionalism and their carers [14]. Lending these explanations to patients will

normalise their experiences, prevent them from developing self-critical beliefs (i.e. that they are weak or foolish) and encourage them to seek the support that they need to improve their quality of life.

Implications for Rehabilitation

- This study found that the unpredictable and uncontrollable nature of emotionalism can be confusing, frustrating and frightening. Many participants spoke of the embarrassment of being emotional in public as well as the guilt of upsetting others around them. Avoidance and social isolation were frequently referred to in the narratives. The use of distraction techniques, maintaining humour, optimism and social support were seen to be helpful ways of managing emotionalism.
- The post-stroke emotionalism cognition questionnaire (PEC-Q) was developed based on patient perspectives. It will be a useful measure of beliefs about emotionalism that could inform clinicians of how patients make sense of and in turn, cope with symptoms. The next step is to test the psychometric properties of the questionnaire and examine whether early negative beliefs can predict patients' recovery outcomes.
- Findings highlight a lack of understanding of the nature and causes of emotionalism. There is a need for better recognition of symptoms in clinical settings and the earlier provision of information and advice to prevent patients from developing maladaptive or distressing beliefs.
- The psychosocial consequences of the disorder are under-investigated. Further research is required to provide insights into the impact of emotionalism on an individual's well-being and quality of life.

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References

- [1] Stroke Association. State of the Nation: stroke statistics. 2017. Retrieved 05 May 2017 https://www.stroke.org.uk/sites/default/files/state_of_the_nation_2017_final_1.pdf
- [2] Carota, A, Calabrese P. "Poststroke Emotionalism." *Journal of Neurological Disorders*. 2013.
- [3] House, Knapp & Calvert, "Emotionalism after stroke." *BMJ*. 1989; 298.6679: 991-994.
- [4] House A, Hosker C. Emotional disorders in neurological rehabilitation. *Handbook of clinical neurology*. 2013; 110:01(1): 389.
- [5] Kasprisin A. "Alternative cognitive therapy for emotional instability (pathologic laughing and crying)." *Physical medicine and rehabilitation clinics of North America* 2004; 15(4): 883-917.
- [6] Colamonic J, Formella A, and Bradley W. "Pseudobulbar affect: burden of illness in the USA." *Advances in therapy*. 2012: 1-24.
- [7] Gillespie DC, Cadden AP, Lees R, West RM, Broomfield NM. Prevalence of Pseudobulbar Affect following Stroke: A Systematic Review and Meta-Analysis. *Journal of Stroke and Cerebrovascular Diseases*. 2016 03: 31;25(3):688-94.
- [8] Engelman W, Hammond FM, Malec JF. Diagnosing pseudobulbar affect in traumatic brain injury. *Neuropsychiatric disease and treatment*. 2014; 10:1903.

- [9] Strowd RE, Cartwright MS, Okun MS, Haq I, Siddiqui MS. Pseudobulbar affect: prevalence and quality of life impact in movement disorders. *Journal of neurology*. 2010 08: 1;257(8):1382-7.
- [10] Cummings JL, Arciniegas DB, Brooks BR, Herndon RM, Lauterbach EC, Piro EP, Robinson RG, Scharre DW, Schiffer RB, Weintraub D. Defining and diagnosing involuntary emotional expression disorder. *CNS spectrums*. 2006: 06:1;11(S6):1-1.
- [11] Maruzairi H, Koh CH. Pseudobulbar affect post stroke. *Brunei Int.Med.J.* 201511(1): 59-61
- [12] McCullagh S, Moore M, Gawel M, Feinstein A. Pathological laughing and crying in amyotrophic lateral sclerosis: an association with prefrontal cognitive dysfunction. *Journal of the neurological sciences*. 1999 10: 31;169(1):43-8.
- [13] Leahy RL, Holland SJ, McGinn LK. Treatment plans and interventions for depression and anxiety disorders. Guilford press; 2011 10: 26.
- [14] Scottish Intercollegiate Guidelines Network. Management of patients with stroke: Rehabilitation, prevention and management of complications and discharge planning publication. 2010. Retrieved 04 April 2017 from <http://www.sign.ac.uk/pdf/sign118.pdf>
- [15] Hackett ML, Yang M, Anderson CS, Horrocks JA, House A. Pharmaceutical interventions for emotionalism after stroke. *Stroke*. 2010 Jul 1;41(7): 460-1.
- [16] Schneider MA, Schneider MD. Pseudobulbar Affect: What Nurses, Stroke Survivors, and Caregivers Need to Know. *Journal of Neuroscience Nursing*. 2017 04: 1;49(2):114-7.

- [17] Perotti LP, Cummings LD, Mercado J. Behavioral Treatment of Pseudobulbar Affect: A Case Report. *Perspectives in psychiatric care*. 2016 04; 1;52(2):82-7.
- [18] Bingeliene A, Shapiro CM, Sockalingam S, Hawa RJ. Pathological Laughing and Crying Post-stroke: Liaison Psychiatrist Beware. *Journal of Neuropsychiatry*. 2016.
- [19] Sacco S, Sarà M, Pistoia F, Conson M, Albertini G, Carolei A. Management of pathologic laughter and crying in patients with locked-in syndrome: a report of 4 cases. *Archives of physical medicine and rehabilitation*. 2006 11; 1;13(6):353-79.
- [20] Tateno A, Jorge RE, Robinson RG. Pathological laughing and crying following traumatic brain injury. *The Journal of neuropsychiatry and clinical neurosciences*. 2004 11;16(4):426-34.
- [21] Calvert T, Knapp P, House A. Psychological associations with emotionalism after stroke. *Journal of Neurology, Neurosurgery & Psychiatry*. 1998; 12: 1;65 (6):928-9.
- [22] Wei N, Yong W, Li X, Zhou Y, Deng M, Zhu H, Jin H. Post-stroke depression and lesion location: a systematic review. *Journal of neurology*. 2015 01; 1;262(1):81-90.
- [23] Ahmed A, Simmons Z. Pseudobulbar affect: prevalence and management. *Ther Clin Risk Manag*. 2013;01:1; (9): 483-9.
- [24] Horrocks JA, Hackett ML, Anderson CS, House AO. Pharmaceutical interventions for emotionalism after stroke. *Stroke*. 2004 Nov 1;35(11):2610-1.

- [25] Carota A, Berney A, Aybek S, Iaria G, Staub F, Ghika-Schmid F, Annable L, Guex P, Bogousslavsky J. A prospective study of predictors of poststroke depression. *Neurology*. 2005 Feb 8;64(3):428-33.
- [26] Taylor GH, Todman J, Broomfield NM. Post-stroke emotional adjustment: A modified social cognitive transition model. *Neuropsychological rehabilitation*. 2011 12: 1;21(6):808-24.
- [27] Kneebone II, Lincoln NB. Psychological problems after stroke and their management: state of knowledge. *Neuroscience and Medicine*. 2012 Mar 1;3(1):83.
- [28] Campbell Burton C, Murray J, Holmes J, Astin F, Greenwood D, Knapp P. Frequency of anxiety after stroke: a systematic review and meta-analysis of observational studies. *International Journal of Stroke*. 2013 Oct 1;8(7):545-59.
- [29] Hagger MS, Orbell S. A meta-analytic review of the common-sense model of illness representations. *Psychology and health*. 2003 01: 1;18(2):141-84.
- [30] Twiddy, M. *Beliefs about stroke: negotiating shared understandings*. Diss. University of Leeds, 2008.
- [31] Beck JS. *Cognitive therapy*. John Wiley & Sons, Inc.; 1979. In Leahy, R. L, Holland SJ, & McGinn LK. *Treatment plans and interventions for depression and anxiety disorders*. Guilford press. 2011.
- [32] Whitmarsh A, Koutantji M, Sidell K. Illness perceptions, mood and coping in predicting attendance at cardiac rehabilitation. *British journal of health psychology*. 2003 May 1;8(2):209-21.

- [33] Petrie KJ, Jago LA, Devcich DA. The role of illness perceptions in patients with medical conditions. *Current opinion in psychiatry*. 2007 03: 1;20(2):163-7.
- [34] Harris C. Return to Work After Stroke. *Stroke*. 2014;45(9):174-6.
- [35] Foxwell R, Morley C, Frizelle D. Illness perceptions, mood and quality of life: A systematic review of coronary heart disease patients. *Journal of Psychosomatic Research*. 2013 Sep 30;75(3):211-22.
- [36] Van Mierlo ML, Van Heugten CM, Post MW, De Kort PL, Visser-Meily JM. Psychological factors determine depressive symptomatology after stroke. *Archives of physical medicine and rehabilitation*. 2015 06: 30;96(6):1064-70.
- [37] Engström SM, Leksell J, Johansson UB, Gudbjörnsdóttir S. What is important for you? A qualitative interview study of living with diabetes and experiences of diabetes care to establish a basis for a tailored Patient-Reported Outcome Measure for the Swedish National Diabetes Register. *BMJ open*. 2016;6(3).
- [38] Black N. Patient reported outcome measures could help transform healthcare. *BMJ (Clinical research ed)*. 2013; 346: 367.
- [39] Patchick EL, Horne M, Woodward-Nutt K, Vail A, Bowen A. Development of a patient-centred, patient-reported outcome measure (PROM) for post-stroke cognitive rehabilitation: qualitative interviews with stroke survivors to inform design and content. *Health Expectations*. 2015 Dec 1;18(6):3213-24.
- [40] Gale NK, Heath G, Cameron E, Rashid S, Redwood S. Using the framework method for the analysis of qualitative data in multi-

disciplinary health research. BMC medical research methodology. 2013
09: 18;13(1):117.

- [41] Ritchie J, Spencer L, O'Connor W. Carrying out qualitative analysis. Qualitative research practice: A guide for social science students and researchers. 2003 Feb 18:219-62.
- [42] Wade DT, Collin C. The Barthel ADL Index: a standard measure of physical disability? International disability studies. 1988;10(2):64-7.
- [43] Sangha H, Lipson D, Foley N, Salter K, Bhogal S, Pohani G, Teasell RW. A comparison of the Barthel Index and the Functional Independence Measure as outcome measures in stroke rehabilitation: patterns of disability scale usage in clinical trials. International Journal of Rehabilitation Research. 2005 06: 1;28(2):135-9.
- [44] Mioshi E, Dawson K, Mitchell J, Arnold R, Hodges JR. The Addenbrooke's Cognitive Examination Revised (ACE-R): a brief cognitive test battery for dementia screening. International journal of geriatric psychiatry. 2006 11: 1;21(11):1078-85.
- [45] Zigmond AS, Snaith RP. The hospital anxiety and depression scale. Acta psychiatrica scandinavica. 1983 06: 1;67(6):361-70.
- [46] Aben I, Verhey F, Lousberg R, Lodder J, Honig A. Validity of the Beck Depression Inventory, Hospital Anxiety and Depression Scale, SCL-90, and Hamilton Depression Rating Scale as screening instruments for depression in stroke patients. Psychosomatics. 2002; 10:43:5:386-93.
- [47] Smith, JA, and Eatough V. "Analysing qualitative data in psychology." (2007): 35-50.

- [48] Leventhal H, Meyer D, Nerenz D. The common-sense representation of illness danger. *Contributions to medical psychology*. 1980; 2:7-30.
- [49] Nicholl CR, Lincoln NB, Muncaster K, Thomas S. Cognitions and post-stroke depression. *British Journal of Clinical Psychology*. 2002 09: 1;41(3):221-31.
- [50] Ritchie J, Lewis J, Nicholls CM, Ormston R, editors. *Qualitative research practice: A guide for social science students and researchers*. Sage; 2013 11: 1.
- [51] Parkinson S, Eatough V, Holmes J, Stapley E, Midgley N. Framework analysis: A worked example of a study exploring young people's experiences of depression. *Qualitative Research in Psychology*. 2016 Apr 2;13(2):109-29.
- [52] Gibbs GR. *Analysing qualitative data*. Sage; 2008 Mar 5.
- [53] Smith J, Firth J. *Qualitative data analysis: the framework approach*. *Nurse researcher*. 2011 01: 21;18(2):52-62.
- [54] Brédart A, Marrel A, Abetz-Webb L, Lasch K, Acquadro C. Interviewing to develop Patient-Reported Outcome (PRO) measures for clinical research: eliciting patients' experience. *Health and quality of life outcomes*. 2014; 5:12:1:15.
- [55] Streiner, DL, Norman GR, and Cairney J. "Health management scales: a practical guide to their measurement and use." (2015).
- [56] Thomas SA, Lincoln NB. Factors relating to depression after stroke. *British Journal of Clinical Psychology*. 2006 Mar 1;45(1):49-61.

- [57] Teoh V, Sims J, Milgrom J. Psychosocial predictors of quality of life in a sample of community-dwelling stroke survivors: a longitudinal study. *Topics in stroke rehabilitation*. 2009 Mar 1;16(2):157-66.
- [58] Picton H. 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' conceptualisation of PSEL. 2014. *Edinburgh Research Archive*. Accessed May 2017 from: <https://www.era.lib.ed.ac.uk/handle/1842/9760>
- [59] Hale, ED, Treharne GJ, and Kitas GD. "The Common-Sense Model of self-regulation of health and illness: how can we use it to understand and respond to our patients' needs?" (2007): 904-906.
- [60] Stack RJ, Simons G, Kumar K, Mallen CD, Raza K. Patient delays in seeking help at the onset of rheumatoid arthritis: the problem, its causes and potential solutions. *Aging Health*. 2013 Aug;9(4):425-35.
- [61] Jain S. Pseudobulbar affect: No laughing matter. *Current Psychiatry*. 2014 Apr 1;13(4):66.
- [62] Ellis-Hill CS, Payne S, Ward C. Self-body split: issues of identity in physical recovery following a stroke. *Disability and rehabilitation*. 2000 01: 1;22(16):725-33.
- [63] Salter K, Hellings C, Foley N, Teasell R. The experience of living with stroke: a qualitative meta-synthesis. *Journal of rehabilitation medicine*. 2008 09: 5;40(8):595-602.
- [64] Keltner D, Gross JJ. Functional accounts of emotions. *Cognition & Emotion*. 1999 09: 1;13(5):467-80.

- [65] Gilbert P. Introducing compassion-focused therapy. *Advances in psychiatric treatment*. 2009 05: 1;15(3):199-208.
- [66] Bowlby J. Processes of mourning. *The International journal of psychoanalysis*. 1961; 01;1: 42:317.
- [67] Nelson JK. The meaning of crying based on attachment theory. *Clinical Social Work Journal*. 1998 03: 1;26(1):9-22.
- [68] Butler EA, Lee TL, Gross JJ. Emotion regulation and culture: are the social consequences of emotion suppression culture-specific? *Emotion*. 2007: 02;7(1):30.
- [69] Shamir M, Travis J. *Boys don't cry? rethinking narratives of masculinity and emotion in the US*. Columbia University Press; 2002.
- [70] Gilbert P, Procter S. Compassionate mind training for people with high shame and self-criticism: Overview and pilot study of a group therapy approach. *Clinical Psychology & Psychotherapy*
- [71] Sarre S, Redlich C, Tinker A, Sadler E, Bhalla A, McKeivitt C. A systematic review of qualitative studies on adjusting after stroke: lessons for the study of resilience. *Disability and rehabilitation*. 2014 04: 1;36(9):716-26.
- [72] Dowswell G, Lawler J, Dowswell T, Young J, Forster A, Hearn J. Investigating recovery from stroke: a qualitative study. *Journal of clinical nursing*. 2000: 07: 6;9(4):507-15.
- [73] Crowe C, Coen RF, Kidd N, Hevey D, Cooney J, Harbison J. A qualitative study of the experience of psychological distress post-stroke. *Journal of health psychology*. 2016 Nov;21(11):2572-9.

- [74] Satink T, Cup EH, Ilott I, Prins J, de Swart BJ, Nijhuis-van der Sanden MW. Patients' views on the impact of stroke on their roles and self: a thematic synthesis of qualitative studies. *Archives of physical medicine and rehabilitation*. 2013 Jun 30;94(6):1171-83.
- [75] Kirkevold M. The unfolding illness trajectory of stroke. *Disability and rehabilitation*. 2002 01: 1;24(17):887-98.
- [76] Burton CR. Re-thinking stroke rehabilitation: The Corbin and Strauss chronic illness trajectory framework. *Journal of Advanced Nursing*. 2000:09:1;32(3):595-602.
- [77] Salisbury L, Wilkie K, Bulley C, Shiels J. 'After the stroke': patients' and carers' experiences of healthcare after stroke in Scotland. *Health & social care in the community*. 2010 07: 1;18(4):424-32.
- [78] Laidlaw, K. "Post-stroke depression and CBT with older people." *Handbook of behavioral and cognitive therapies with older adults*. Springer New York, 2008. 233-248.
- [79] Wade, DT, Langton Hewer, R, Skilbeck, CE, & David, RM. *Stroke: A critical approach to diagnosis, treatment and management*. London: Chapman and Hall Medical. 1985.
- [80] Brennan J. Adjustment to cancer—coping or personal transition? *Psycho-Oncology*. 2001; 01:1:10: 1:1-8.
- [81] Kerr GD, Slavin H, Clark D, Coupar F, Langhorne P, Stott DJ. Do vascular risk factors explain the association between socioeconomic status and stroke incidence: a meta-analysis. *Cerebrovascular Diseases*. 2011;31(1):57-63.

- [82] Addo J, Ayerbe L, Mohan KM, Crichton S, Sheldenkar A, Chen R, Wolfe CD, McKeivitt C. Socioeconomic status and stroke. *Stroke*. 2012; 43(4):1186-91.
- [83] Mesquita B, Walker R. Cultural differences in emotions: A context for interpreting emotional experiences. *Behaviour Research and Therapy*. 2003 07; 41(7):777-93.
- [84] Champion AJ. *Neuropsychological Rehabilitation: A resource for group-based education and intervention*. Wiley: Chichester. 2006.
- [85] Gould RA, Safren SA, Washington DO, Otto MW. A meta-analytic review of cognitive-behavioral treatments. *Generalized anxiety disorder: Advances in research and practice*. 2004:248-64.
- [86] Work SS, Colamonic JA, Bradley WG, Kaye RE. Pseudobulbar affect: an under-recognized and under-treated neurological disorder. *Advances in therapy*. 2011 07; 28(7):586-601.

Appendix E: WoSRES and NHS Lothian ethical approval letters

WoSRES

West of Scotland Research Ethics Service



Miss Niamh McAleese
 Trainee Clinical Psychologist
 University of Edinburgh/NHS Scotland Clinical
 Psychology Training Programme
 School of Health in Social Sciences, Doorway
 6,
 Old Medical School
 Teviot Place, Edinburgh
 EH8 9 AG

West of Scotland REC 4
 West Ambulatory Care Hospital
 Dalnair Street
 Yorkhill
 Glasgow
www.nhsqgc.org.uk

Date 03 May 2016
 Direct line 0141-232-1807
 e-mail Wosrec4@ggc.scot.nhs.uk

V2: Reissued 03 May 2016 – Participant consent form not included

Dear Miss McAleese

Study title: The Post-Stroke Emotionalism Cognition Questionnaire (PEC-Q): A development study
REC reference: 16/WS/0071
Protocol number: AC16017
IRAS project ID: 200114

Thank you for your email of 28 April. I can confirm the REC has received the documents listed below and that these comply with the approval conditions detailed in our letter dated 28 April 2016.

Documents received

The documents received were as follows:

| Document | Version | Date |
|---|-----------|---------------|
| Participant information sheet (PIS) [Updated Participant Information Sheet] | Version 4 | 28 April 2016 |

Approved documents

The final list of approved documentation for the study is therefore as follows:

| Document | Version | Date |
|---|-----------|---------------|
| Covering letter on headed paper [Cover letter] | | 10 March 2016 |
| Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [PL confirmation letter] | | 13 July 2015 |
| GP/consultant information sheets or letters [GP letter] | Version 2 | 10 March 2016 |
| Interview schedules or topic guides for participants [Sample interview schedule] | Version 2 | 10 March 2016 |

| Document | Version | Date |
|---|-----------|------------------|
| IRAS Checklist XML [Checklist_28042016] | | 28 April 2016 |
| Non-validated questionnaire [TEARS-Q] | Version 2 | 10 March 2016 |
| Other [EL Certificate] | | 31 August 2015 |
| Other [Clinical Trial Liability Insurance] | | 28 July 2015 |
| Other [Professional Indemnity Insurance] | | 28 July 2015 |
| Other [CV Academic co-supervisor] | | 11 November 2015 |
| Other [Unfavourable opinion letter] | | 23 December 2015 |
| Other [Debrief letter] | Version 2 | 10 March 2016 |
| Other [Staff information sheet] | version 1 | 19 April 2016 |
| Other [Updated Research proposal] | version 3 | 19 April 2016 |
| Other [Updated Flowchart] | version 3 | 19 March 2016 |
| Participant consent form [Participant consent form] | Version 2 | 10 March 2016 |
| Participant information sheet (PIS) [Updated Participant Information Sheet] | Version 4 | 28 April 2016 |
| REC Application Form [REC_Form_10032016] | | 09 March 2016 |
| Research protocol or project proposal [Research Proposal] | Version 2 | 10 March 2016 |
| Summary CV for Chief Investigator (CI) [CV_PI] | | 10 March 2016 |
| Summary CV for supervisor (student research) [CV for main supervisor] | | 10 March 2016 |

You should ensure that the sponsor has a copy of the final documentation for the study. It is the sponsor's responsibility to ensure that the documentation is made available to R&D offices at all participating sites.

16/WS/0071

Please quote this number on all correspondence

Yours sincerely



Sophie Bagnall
Assistant Coordinator

Copy to: *Mrs Jo-Anne Robertson, The University of Edinburgh*
Mr Gavin Robertson, NHS Lothian Research & Development Office

University Hospitals Division

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

FM/GM/approval

5th May 2016

Miss Niamh McAleese
Trainee Clinical Psychologist
School of Health in Social Sciences
Old Medical School, Teviot Place
Edinburgh
EH8 9AG

Research & Development
Room E1.12
Tel: 0131 242 3330

Email:
R&DOffice@nhslothian.scot.nhs.uk

Director: Professor David E Newby



Dear Miss McAleese,

Lothian R&D Project No: 2016/0088

Title of Research: The Post-Stroke Emotionalism Cognition Questionnaire (PEC-Q): A development study

REC No: 16/WS/0071

Participant Information Sheet:
Version 4 Dated 28th April 2016

Consent Form:
Version 2 Dated 10th March 2016

Protocol: Version 3 Dated 19th April 2016

I am pleased to inform you that this study has been approved for NHS Lothian and you may proceed with your research, subject to the conditions below. This letter provides Site Specific approval for NHS Lothian.

Please note that the NHS Lothian R&D Office must be informed if there are any changes to the study such as amendments to the protocol, recruitment, funding, personnel or resource input required of NHS Lothian.

Substantial amendments to the protocol will require approval from the ethics committee which approved your study and the MHRA where applicable.

Please inform this office when recruitment has closed and when the study has been completed.

I wish you every success with your study.

Yours sincerely

A handwritten signature in black ink that reads 'Fiona McArdle'.

Ms Fiona McArdle
Deputy R&D Director

cc: Ms Sheena Muir, Assistant General Manager, Astley Ainsley Hospital
Mr Chris Stirling, Hospital Director, WGH
Dr Andrew Flapan, Associate Medical Director - Medicine Services, RIE

Appendix F: International Classification of International Statistical Classification of Diseases and Related Health Problems 10th Edition (ICD-10)

F06 OTHER MENTAL DISORDERS DUE TO BRAIN DAMAGE AND DYSFUNCTION AND TO PHYSICAL DISEASE

F06.6 Organic emotionally labile (asthenic) disorder

A disorder characterised by marked and persistent emotional incontinence or lability, fatigability or a variety of unpleasant physical sensations and pains regarded as being due to the presence of an organic disorder. This disorder is thought to occur in association with cerebrovascular disease or hypertension more often than other causes.

Excludes: somatoform disorders, nonorganic or unspecified (F45. -)

- A. The general criteria for F06 must be met.
- B. The clinical picture is dominated by emotional lability (uncontrolled, unstable, and fluctuating expression of emotions).
- C. There is a variety of unpleasant physical sensations such as dizziness or pains and aches.

Comments: Fatigability and listlessness (asthenia) are often present but are not essential for the diagnosis.

- G1. Objective evidence (from physical and neurological examination and laboratory tests) and/or history of cerebral disease, damage or dysfunction, or of systemic physical disorder known to cause cerebral dysfunction, including hormonal disturbances (other than alcohol or other psychoactive substance-related) and non-psychoactive drug effects.
- G2. A presumed relationship between the development (or marked exacerbation) of the underlying disease, damage or dysfunction, and the mental disorder, the symptoms of which may have immediate onset or may be delayed.
- G3. Recovery or significant improvement of the mental disorder following removal
or improvement of the underlying presumed cause.
- G4. Absence of sufficient or suggestive evidence for an alternative causation of
the mental disorder, e.g. a highly loaded family history for a clinically similar or related disorder.

If criteria G1, G2, and G4 are met, a provisional diagnosis is justified; if, in addition, there is evidence of G3, the diagnosis can be regarded as certain.



Information for participants

You are invited to part in a research study. Before you make your decision about taking part, it's important that you fully understand this study and what it involves. Please read through this sheet carefully and talk about it with your family or support staff if you wish.

What is the purpose of the study?

This study is targeted at those who have problems controlling laughing and crying after having a stroke. This is sometimes called 'Post-Stroke Emotionalism' (PSE). Even though it is common, there is very little research asking about what people think about PSE and how it affects their everyday lives. This study aims to find out more about this and develop a questionnaire that can help guide treatments for people with these problems.

Why have I been invited?

We are looking for people who have had a stroke who have difficulties with controlling laughing or crying. We are inviting you to participate as you are receiving treatment at Western General Hospital, Royal Infirmary of Edinburgh, Astley Ainslie Hospital or St John's Hospital (*delete as appropriate*). Your clinical team has given you this information sheet as they thought that you may be interested. You are very welcome to involve a family member, friend or carer in this study if you wish.

Do I have to take part?

No. Participation is entirely voluntary. You should not feel under any pressure to be involved. If you do decide to take part, you'll be asked to sign a consent form. Even after signing the form you are still free to withdraw at any point and without giving a reason, this will not affect any healthcare that you may receive now or in the future. If you do wish to take part, we will ask you for your permission to inform your GP to let them know that you are participating in the study.

What would I be asked to do?

This study would ask you to meet with the main researcher who will ask you questions about what it is like to have problems with controlling how you express emotions after stroke. We can show you the questions in advance of the meeting if you wish. The general topics talked about in the meeting will be used to create a questionnaire.

The meeting will last up to 60 minutes and will be based at your home or at the NHS Lothian Royal Infirmary of Edinburgh/ St John's Hospital/ Western General/ Astley Ainslie Hospital inpatient stroke unit (*delete as appropriate to participant*).

At the end of the meeting the main researcher will ask to arrange a time to meet with you again or to speak with you over the telephone in a few weeks time. This will be to talk with you about how the questionnaire looks and whether it reflects what was talked about in the meeting. This should take 15-20 minutes.

The main researcher will make sure to give or post you this questionnaire before speaking with you.

How do I take part?

If you would like to take part, we would ask that you let a member of your clinical team know. We will then arrange to meet with you to explain the study in more detail and answer any questions. If you remain interested, we would ask you to sign the consent form and return this to any member of your clinical team.

We will ask for your permission to request information from your clinical team from via your medical notes about your stroke and about the impact that your stroke has had on your mood and physical or thinking abilities. We will not request this without your permission. Once we have this information and your completed questionnaires we will agree a date and time with you for the meeting.

What are the possible benefits of taking part?

The information that you provide will help us, as healthcare professionals, to better understand the impact that PSE symptoms can have. This could lead to new ways of identifying those most affected who are struggling and to help to develop or improve future therapies.

What are the possible risks of taking part?

Talking about the effects of PSE in the group could potentially lead you to become upset. If this does occur, please do contact us or a member of your stroke care team so that they can help you. Contact details of additional support services are also included with this information sheet.

Who will have access to my details?

The information that you provide will be as confidential as your medical records. This includes your consent form, completed questionnaires and your responses in the meeting. The information that you provide to the researcher from the questionnaires will not be shared with other individuals. The only instance in which information that you provide may be shared is if you disclose information that indicates that either yourself or another person is at risk of danger. In this instance, the main researcher would have a duty of care to share this information with your stroke clinician or your GP. However, we would always discuss this with you beforehand.

What happens to the information?

All data gathered from this study will be kept confidential and stored securely. The meeting will be audio recorded and responses will be typed out. These responses will be coded and your name would not be used in any written documents or any computer

files. If you do wish to discontinue it is important that you let us know before we begin to analyse the data as otherwise it will not be possible to identify you from other participants at that stage. This 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.

What if I have further concerns?

If you have queries about any aspect of the research, you can ask to speak to the main researcher who will try to address your questions. In the very unlikely event that something goes wrong and you are harmed during the research and this is due to someone 's negligence then you may have grounds for a legal action for compensation against NHS Lothian but you may have to pay your legal costs. The normal National Health Service complaints mechanisms will still be available to you (if appropriate).

If you wish to make a complaint about the study please contact: *The NHS Lothian Complaints Team, 2nd Floor, Waverley Gate, 2-4 Waterloo Place, Edinburgh, EH1 3EG* or Telephone: 0131 536 3370 and Email: feedback@nhslothian.scot.nhs.uk.

Who has reviewed the study?

All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee. A favourable ethical opinion has been obtained from NHS Lothian REC. NHS management approval has also been obtained.

What will happen to the results of the study?

The results of this study will be written up as a Clinical Psychology thesis and it will be submitted for publication in a peer reviewed scientific journal. As already indicated, all information will be kept confidential and no names will be included in the final report. If you wish to know the outcome of our research, please indicate this on the attached consent form and we can send you the findings once the study is completed.

If you would like further information, please contact:

Main researcher:

Niamh McAleese (Trainee Clinical Psychologist)

University of Edinburgh/NHS Scotland Clinical Psychology Training Programme

School of Health in Social Science, the University of Edinburgh, Medical School (Doorway 6)
Teviot Place, Edinburgh, EH8 9AG

T: +44 (0)131 650 3889

E: niamh.mcaleese@nhslothian.scot.nhs.uk or s1051471@sms.ed.ac.uk

| | | |
|---|--|--|
| <p>Clinical Supervisor: Dr. David Gillespie <i>Consultant Clinical Neuropsychologist</i></p> <p>NHS Lothian Western General Hospital T: 0131 537 2099 david.gillespie@nhslothian.scot.nhs.uk</p> | <p>Academic Supervisor: Dr. Suzanne O'Rourke <i>Lecturer in Clinical Psychology</i></p> <p>University of Edinburgh T: 0131 650 4272 suzanne.o'rourke@ed.ac.uk</p> | <p>Academic Supervisor: Dr. Azucena Guzman <i>Lecturer in Clinical Psychology</i></p> <p>University of Edinburgh T: 031 651 51 62 azucena.guzman@ed.ac.uk</p> |
|---|--|--|

For general queries regarding research within the University of Edinburgh please contact:

Dr. Emily Newman

Clinical Psychology Research Director

University of Edinburgh

T: +44 (0) 131 651 3945

emily.newman@ed.ac.uk

<http://www.ed.ac.uk/schools-departments/health/clinical-psychology/about/contact>

Thank you for your time and for any further involvement with this study.

Additional support services

Stroke Association

Web: www.stroke.org.uk

Tel: 0303 3033 100

Chest, Heart & Stroke Scotland

Web: www.chss.org.uk

Tel: 0845 077 6000

Edinburgh Headway Group

Web: www.edinburghheadway.org.uk

Tel: 0131 5379116

Vocals Carers Centre

Web: www.centre@vocal.org.uk

Tel: 0131 622 6666

Different Strokes

Web: www.differentstroke.co.uk

Tel: 01908 317618



Participant consent form

Please initial the box if you agree:

- 1 I have read and understood the participant information sheet and the researcher has answered my queries.

- 2 I understand that taking part is voluntary and that I am free to change my mind and withdraw at any time, without giving any reason. This will not affect the care that I receive in any way

- 3 I understand that relevant sections of my medical notes may be looked at by the researcher via my stroke team and by individuals from the regulatory authorities or by Sponsors (NHS Lothian and the University of Edinburgh) where it is relevant to my participation. I give permission for those individuals to have access to my records

- 4 I consent to my stroke care clinicians being informed that I am taking part in this study

- 5 I understand that if the researcher is worried about a risk of harm to myself or someone else during the study, then they will speak to a healthcare professional involved in my care.

- 6 I would like to be posted the results of this study.

- 7 I agree to the interview being audio recorded

- 8 I agree to the use of anonymised quotes in publications

I agree to take part in this study.

Printed name of Participant

Date

Signature

Appendix H: Research protocol



Thesis Research Proposal

This form is for methodological review of projects that are **not** being submitted as assessed work for Research 1. (e.g. where a trainee has already received a pass mark for Research 1, but subsequently changed the intended thesis project, or for trainees who started training in 2009 or earlier and thus did not need to complete Research 1 and have not previously had university approval for their study).

In such circumstances the form will be reviewed by a member of the academic team and will receive detailed feedback, but will not be graded. The feedback will include an evaluation of the viability of the project and any recommendations. If there are significant concerns about viability, the project will be flagged to the research director and the research committee will decide whether the project can proceed in its current form.

| | |
|---|----------------------|
| Trainee Name | |
| Niamh McAleese | |
| Provisional Thesis Title | |
| <i>The Post-Stroke Emotionalism cognition questionnaire (PEC-Q): A development study</i> | |
| Proposed Setting | |
| NHS Lothian | |
| Allocated Thesis Project Supervisors | |
| Clinical | Dr. David Gillespie |
| Academic 1 | Dr. Suzanne O'Rourke |
| Academic 2 | Dr. Azucena Guzman |
| Anticipated Month / Year of Submission | |
| Must be May of final year. Trainees from 2011 intake onwards must submit in May. Trainees who started in 2010 or earlier are advised to submit in May to reduce potential for HCPC registration difficulties. | |
| May 2017 | |
| Date Form Submitted / Version | |
| 28-04-2016 | |

Please Note: Whilst this is not an ethics review process, where questions have some similarities to questions contained in the NHS IRAS Research Ethics form, the corresponding IRAS question numbers are given in parentheses. This is intended to facilitate completion of NHS ethics where such approval is needed.

Section 1: Introduction

1.1 Provide a brief critical review of relevant literature, which should clearly demonstrate the rationale and scientific justification for the research

1000 – 1500 words

Relevant to IRAS A12

Post-stroke Emotionalism (PSE) is a disturbance in the ability to control emotional expression and is a common consequence of stroke (Carota & Calabrese, 2013). It is a disorder of affect and is characterized by transient, exaggerated and uncontrolled laughter or crying that occurs frequently and spontaneously (House *et al.*, 1989). At times these responses are incongruent to the individual's underlying emotional state, on other occasions they are an exaggerated response to emotional stimuli (House & Hosker, 2013). Onset of symptoms is usually at one-week post stroke with their frequency and severity gradually reducing over time (House *et al.*, 2008; Kim & Choi-Kwon, 2013).

The prevalence rates of PSE are unclear and vary between individual studies (Hackett, Kohler, O'Brien & Mead, 2014). Nonetheless it is estimated that the condition affects approximately one in four stroke survivors and ranges from 17-34%, dependent on time since stroke (House *et al.*, 2008). Given the similar symptom profiles of those with PSE compared to those with clinical mood disorders, great difficulty exists in detecting and diagnosing PSE (Cummings *et al.*, 2006). Furthermore, the inconsistent use of screening tools and terminologies within the literature only serves to exacerbate this (House & Hosker, 2013). The most frequently used terms to describe PSE include post-stroke emotionalism, pathological laughing and crying, emotional lability, emotional incontinence and involuntary emotional expression disorder (Allman *et al.*, 1990; Kim & Choi-Kwon, 2013; Rosen & Cummings, 2007). Those with PSE are quantitatively different from those with depression; most noticeably by the brevity of their symptoms, the dissociation between the expression of emotion and mood and their lack of depressive automatic thoughts (Picton, in preparation).

PSE is considered to occur secondary to a wide range of neurological conditions such as stroke, multiple sclerosis, dementia, amyotrophic lateral sclerosis and traumatic brain injury (Colamonico, Formella, & Bradley, 2012). Research suggests that PSE symptoms may be caused by lesions to the frontal lobes or descending corticobulbar and cerebellar pathways that regulate motor control and the co-ordination of emotional expression (Engleman, Hammond & Malec, 2013; Parvizi, Anderson, Martin, Damasio & Damasio, 2001). These impaired communicating pathways from frontal and cortical motor inputs are thought to disrupt the cerebellum's ability to modulate the motor expression of emotion (Miller, Pratt & Schiffer, 2011). The neurochemical hypothesis further argues that PSE arises from dysfunctional serotonergic neurotransmission in the cerebellum, thought to play an important role in emotion processing (Maruzairi & Koh, 2015). This is theorised to lead to a loss of a patient's voluntary control over their emotional expression in non-emotive situations (Miller, Pratt & Schiffer, 2011). Despite ongoing advances, the pathophysiology of PSE is not fully understood and there is no conclusive evidence demonstrating definitive links between specific lesion

locations and emotionalism (Carota & Calabrese, 2013; Miller, Pratt & Schiffer, 2011). Nonetheless PSE is largely regarded to be an organic disorder. This is reflected by current guidelines that recommend pharmacology as the first line of treatment in clinical practice (Intercollegiate Stroke Working Party, 2010; SIGN, 2010). However, the evidence base for this is tenuous and often PSE symptoms are left untreated (Hackett *et al.*, 2008).

There has been little investigation into the psychological mechanisms underlying PSE and the sparse existing research has relied heavily on individual case studies (Sacco *et al.*, 2008; Tateno, Jorge, & Robinson, 2004). The incongruence between a patient's behaviour and their emotional responses has been linked with significant distress, embarrassment and social avoidance (Calvert, Knapp & House, 1998; Maruzairi & Koh, 2015). It has been found that those with PSE report a reduced quality of life and are more likely to use avoidant coping strategies compared to those without the condition (Eccles, House & Knapp, 1999; Wei *et al.*, 2015). It is well known within the cognitive-behavioural literature that avoidance can serve as a maintenance factor for mood disorders (Picton, in preparation). Indeed those with PSE are theorized to be at an increased risk of developing depression and anxiety (House & Hosker, 2013). There is no research investigating the associations between PSE and clinical mood disorders, very little is known about maintenance factors and interestingly most patients with PSE are not depressed (House, Knapp & Calvert, 1989; Kneebone & Lincoln, 2012).

In efforts to construct a psychological model of PSE, past theorists hypothesized that it is a manifestation of a more general disorder of emotional control, likening it to that of Post Traumatic Stress Disorder (Carota & Calabrese, 2013; Calvert, 1998). However many of the key indicators of PTSD such as flashbacks, persistent re-experiencing of traumatic events, hyper vigilance and nightmares are not reported in those with PSE (Engleman, Hammond & Malec, 2013). Those with PTSD experience outbursts of emotion in response to triggers that remind them of the traumatic event, but often those with PSE report no emotive reason for their outbursts of laughter or crying (Carota & Calabrese, 2013).

Cognitive theorists emphasize the importance of key cognitions in the precipitance of emotional difficulties (House & Hosker, 2013). Indeed automatic negative thoughts and dysfunctional assumptions play a pivotal role in anxiety and depression (Williams & Garland, 2002). Often it is the *meaning* that an individual applies to an event rather than the *nature* of the event itself that determines their ability to cope (House & Hosker, 2013). Lincoln and colleagues (2002) investigated the cognitions of patients with post stroke depression (PSD) to find that they experienced a greater level of depressive cognitions about their stroke relative to other stroke patients without depression. Based on these findings the authors recommended the use of psychologically informed treatments for those with PSD based on cognitive-behavioural principles.

The impact of patients' illness cognitions on their psychological functioning is well established in other medical conditions (Leventhal, Nerenz & Steele, 1984). Illness perceptions are under-researched in the stroke population (Townend, 2010) and have never been explored before in those with PSE. PSE is linked to poor psychosocial outcomes, like reduced quality of life, avoidance or lowered social participation (Colamonico & Formella, 2012). PSE is also a risk factor for post-stroke depression and this is further associated with poor rehabilitation outcomes and mortality (Kootker *et al.*, 2016). Thus, there exists a need to explore the psychological factors that potentially precipitate the emergence of some of these functional difficulties in PSE.

The development of a measure that could capture negative and possibly dysfunctional perceptions about PSE could be used by clinicians to detect patients “at risk”, who may be more vulnerable to mood difficulties. If an important role for these psychological factors can be demonstrated, then this may provide the basis for the application of psychologically informed interventions in the PSE population.

| |
|---|
| Section 2: Research Questions / Objectives |
| 2.1 What is the principal research question / objective? |
| <i>IRAS A10</i> |
| To develop a questionnaire that identifies cognitions in individuals with PSE. It will be entitled the “Post-stroke Emotionalism Cognitions Questionnaire” (PEC-Q). |
| 2.2 What are the secondary research questions / objectives, if applicable? |
| Keep these focused and concise, with a maximum of 5 research questions |
| <i>IRAS A11</i> |
| N/A |

| |
|---|
| Section 3: Methodology |
| 3.1 Give a full summary of your design and methodology |
| It should be clear exactly what will happen at each stage of the project |
| <i>IRAS A13</i> |
| <i>Design:</i> |
| The project will use a qualitative research design. This is based on previous questionnaire development studies in the stroke population (Lincoln et al., 2002; Streinger & Norman, 2008). Given that there are no existing measures of cognitions in those with PSE, the methodology for this project is adapted from researched conducted by Lincoln and colleagues (2002) who investigated similar constructs but with a different population (stroke patients with depression). |
| Semi-structured interviews with patients with PSE will be facilitated and information derived from these will be used to develop a self-report outcome measure. This will follow several steps adhering to protocols grounded in qualitative literature (Braun & Clarke, 2006; Silverman, 2000). Semi-structured interviews in qualitative research are a useful tool for gaining unique insights into the experiences and perspectives of a specific population (Bender & Ewbank, 1994). Semi-structured interviews have been widely used within the stroke literature to explore un-researched areas and inform clinical practice guidelines (Hawkins et al., 2015). The use of a semi-structured approach in this study will allow control of the interview so that the purpose of the study can be achieved and the research questions explored (Holloway & Wheeler, 2010). Interviews will focus on eliciting appraisals directly from patients about PSE and objectively quantify those as statements to generate a questionnaire format. |
| This study will use a purposive sampling design, whereby participants will be deliberately selected based on similar or shared characteristics. By using this approach it is thought that these individuals could provide the depth and |

diversity of responses needed for interview data, as recommended for qualitative research by Tong *et al.* (2007).

Participants:

Participants will be inpatient and recently discharged stroke patients with a diagnosis of PSE, as judged by their clinical teams from the NHS Lothian Astley Ainslie, Royal Infirmary, St John's Hospital and Western General acute or rehabilitation stroke units.

Recruitment strategy:

- The PI will approach the clinical teams from the Astley Ainslie, Royal Infirmary Edinburgh, Western General Hospital and St Johns Hospital inpatient acute/rehabilitation stroke units. The PI will ask staff to identify medically stable patients with a diagnosis of PSE (using the ICD-10 criteria). Staff will provide a formal confirmation that these patients have the capacity to consent to the study.
- A member of the treating clinical team will approach patients to introduce the study. They will give patients the participant information sheet about the interview and ask for their permission for the PI to speak with them further about the study.
- The PI will then approach potential participants to explain the details of the information sheet and answer any questions about the study. The PI will explain that additional information is needed for this study and that they will need specific details from their medical records (i.e. side and location of stroke, time since stroke) and about their functional or cognitive impairments (as measured on the Barthel Index and Montreal Cognitive Assessment which are routinely collected measures). The PI will ask for their permission to access these details from their clinical team (detailed in consent form).
- Those patients that consent to take part will indicate their willingness to participate on the consent form. This will be returned to the clinical team to be collected by the PI after one week.
- Staff will give participants the HADS to complete independently (or with support from staff or family).
- The PI will return one week later to collect these questionnaires and to arrange a time for the interview. They will also inform participants of the non-cash incentive for taking part.
- The interviews will be held in booked clinic rooms on the stroke wards at AA, SJH, WGH and RIE. It is anticipated that the interviews will be facilitated in the evening to avoid any disruptions to patient's rehabilitation programmes or routine medical procedures. Stroke ward nurses will be present on the ward should any issues arise.
- Upon completion of the interviews the PI will verbally debrief participants and give them the debrief letter signposting additional support services.

The PI will arrange a follow-up 1:1 meeting or contact via telephone to review the PEC-Q to test its content validity.

Procedure:

(1) Expert consultation

The researcher will generate interview questions based on the existing PSE literature and bring these to a panel of NHS Lothian stroke specialist staff. From this meeting, a series of open ended questions regarding the impact of PSE will be constructed.

(2) Semi-structured interviews

Each participant will take part in an hour interview based at a booked clinic room located at their NHS Lothian Hospital. Participants will be encouraged to invite a family member or carer for additional support if wished. Participants may see questions in advance of the interview should they request this. The PI will explain the structure of the interview as well as boundaries of confidentiality. (Please refer to interview sample questions).

Upon completion of the interview the PI will explain that in order to test the face validity of the measure they would ask that they meet or speak with the participant again in a couple of weeks. If the participant consents to this the PI will arrange a time and make sure to give or post them the measure in advance of this. During this follow up the participant will be asked to make judgments on whether items are relevant, unambiguous and written in clear language. Participants will be asked to comment on the general design of the PEC-Q and on the wording of questions. This is to ensure that the questionnaire items reflect importance as judged by the target population.

(3) Qualitative reviewer

After transcription, both the PI and an additional researcher (volunteer trainee clinical psychologist) will independently rate themes within the data. The additional researcher will be blind to any participant identifiable information. The will be a method used to reduce the potential for researcher bias during analysis. The additional researcher and PI will then meet and agree on a mutually identified themes that will be used to generate a list of potential items to include on the PEC-Q.

(4) Consensus panel

An expert consultation meeting will be arranged with up to 5 stroke specialist staff to review the list of statements generated from the interviews. During this meeting professionals will be asked to review the domains identified, the language used and for their judgements of relevance to professional person centred care in stroke. Items will be removed or amended based on the panel responses and this will determine the major domains of the PEC-Q. Feedback from both the interviews and the consensus will be amalgamated to create the first draft of the PEC-Q. This will use a 5-point Likert scale asking participants to indicate their agreement to statements varying from "not at all" = 1 to "all of the time" =5. Higher scores will

indicate more negative cognitions about PSE. The face validity of the PEC-Q will be examined based on feedback from the consensus panel and interview feedback.

3.2 List the principal inclusion and exclusion criteria

IRAS A17-1 and IRAS A17-2

Inclusion criteria

- Diagnosis of ischemic or haemorrhagic stroke acute at the post-acute stage (maximum 1 year post stroke onset). This will be confirmed by the stroke clinical team.
- A probable diagnosis of emotionalism as decided by their clinical team based on (a) meeting criteria for symptoms listed on the ICD-10 Diagnostic Criteria.
- Adults aged 18 to 90 years
- English speaking and with no more than mild levels of language disturbance (as identified by their clinical team)
- Able to give informed consent. This will be based on formal assessment and the opinion of the stroke team about each participant's capacity and cognitive abilities.

Exclusion criteria

- Those with significant communication deficits that would prevent them from contributing to an interview (confirmed by the clinical team)
- Severe cognitive impairment, a previous psychiatric history or those with current substance or alcohol dependence. Patients with pre-morbid cognitive deficits such as dementia, head injury or a learning disability. Those who have sustained a Transient Ischemic Attack (indicated by their medical notes)
- Those with a severe concurrent medical condition that would prevent participation in study procedures (i.e. paralysis or severely limited mobility, indicated as a "severe ADL problem" on the Barthel Index meaning that these patients would have reduced opportunities to avoid social situations)
- Patients with a life expectancy of <3 months determined by their clinical team

Demographic information regarding patients' age, side of stroke and time since stroke will be taken from routinely collected information. On admission patients are typically screened for mood using the Hospital Anxiety and Depression Scale (HADS), levels of cognitive impairment using the Montreal Cognitive Assessment (MoCA) and functional ability using the Barthel Index (BI). This information will be sought to satisfy the inclusion criteria and will only be accessed once the researcher has gained consent from the participant to do this.

3.3 How will data be collected?

If quantitative, list proposed measures and justify the use of these measures. If qualitative, explain how data will be collected, giving reasonable detail (don't just say "by interviews".)

Qualitative data:

Semi-structured interviews will be conducted at the NHS WGH, RIE, SJH and will be facilitated by the researcher with support from nursing staff if needed. The content of these sessions will be audio recorded and data will be transcribed using the *Audacity Software* and *nVivo* software, which are freely accessible audio editor resources. The researcher will also document written accounts of participants' non-verbal behaviours that will further contribute to the interpretation of the data at the analysis phase.

Section 4: Sample Size

4.1 What sample size is needed for the research and how did you determine this?

For quantitative projects, outline the relevant Power calculations and the rationale for assuming given effect sizes. For qualitative projects, outline your reasoning for assuming that this sample size will be sufficient to address the study's aims

IRAS A59 and IRAS A60

There is no formula for theoretical sampling within qualitative research (Galvin, 2014). Given that illness representations are thought to change over the lifespan (Petrie & Weinman, 1997) and taking into account the wide age range for this study (18 to 90 years), the PI proposes to split the participants into 3 cohorts. These will roughly be 18-30 years, 31-64 and 65+. Studies indicate that sample sizes should be dependent upon reaching "saturation" within the data where after a number of interviews has been performed, it is unlikely that performing further interviews will reveal new information that hasn't already emerged in a previous interview (Guest, Bunce, and Johnson, 2006). The PI proposes to adapt the methodology from the study used by Lincoln et al. (2002) which investigated cognitions of depressed stroke patients where they analysed 9 CBT transcripts. Given this in conjunction with the principles of saturation, the PI aims to recruit approximately 10 participants with PSE per group or until the data reaches informational redundancy, equating to 30 participants overall.

4.2 Outline reasons for your confidence in being able to achieve a sample of at least this size

Give details of size of known available sample(s), percentage of this type of sample that typically participate in such studies, opinions of relevant individuals working in that area

The following Hospitals are proposed recruitment sites:

1. The Royal Infirmary Acute Stroke Unit –22 beds with an additional 41 beds off the acute site.
2. Western General Acute Stroke Unit - 40 beds and 317 discharged in 2015
3. St John's Hospital Acute Stroke Unit - 22 beds
4. Astley Ainslie Rehabilitation Unit – 11 wards

Last year a total of 1,450 stroke patients were discharged collectively from the above hospitals (Stroke Care Audit, 2015) with an average admission time of up

to 5 weeks. The proposed time period for data collection is estimated to be approximately eight months. As the current prevalence rate of PSE is about 25% at the acute stage it is predicted that of this figure up to 362 of these patients will have the condition. After the inclusion and exclusion criteria it is believed that at least 70% of this sample should be eligible to participate in this study. Given that approximately 12-19 participants are required for the success of the project, the researcher and thesis supervisors have expressed confidence in the ability to recruit the sample required. Should the researcher encounter difficulties with attaining numbers, an additional recruitment method via the Edinburgh community stroke services will be considered.

Section 5: Analysis

5.1 Describe the methods of analysis (statistical or other appropriate methods, e.g. for qualitative methods) by which the data will be evaluated to meet the study objectives

IRAS A62

The Framework Method is considered to be a form of thematic analysis and identifies commonalities within qualitative data before focusing on relationships to draw descriptive conclusions clustered around themes (Gale *et al.*, 2013). It is a widely-used methodology within health research and is unique in that it uses a “matrix” method for categorizing summarized data within a dataset. This means that interviewees are arranged into cases (rows) and codes as columns, allowing a systematic structure for which the researcher can analyse data across cases and categories (Gale *et al.*, 2013).

This method was chosen for this study for several reasons; it is most frequently used in research to thematically analyse content from semi-structured interviews (Gale *et al.*, 2013) and it can be adapted based either on data-driven or theory-driven theoretical perspectives (Braun and Clarke, 2006). Its systematic approach lends well to a mixed methods design. Finally, as this project aims to explore patient’s experiences with PSE using a semi-structured approach an approach flexible enough to analyse a combined inductive/deductive approach is warranted.

Procedure for analysis:

- 1. Transcription by researcher**
- 2. Familiarisation with the interview**
- 3. Coding** – allocation of an initial paraphrase of each sentence. This process will classify all of the data so that it can be compared systematically with other parts of the data set. This process will be repeated by a second separate researcher.
- 4. Develop a working analytic framework** – group initial codes into categories which are clearly defined
- 5. Applying framework** – index subsequent codes using existing categories (n-Vivo software).
- 6. Charting data into framework matrix** - summarizing/charting the data by case and category from each transcript into a spreadsheet to form a matrix
- 7. Interpreting the data** – identify characteristics and differences between the data to form typologies and map connections between categories to explore relationships and causality.

A significant threat in qualitative research is that of reliability arising from the wide variety of possible interpretations of the same data set. To mitigate these concerns a number of procedures will be adhered to. A separate rater will analyze

the data independently and simultaneously to the primary researcher. Mutually agreed themes will then be transformed into statements and shared with patients for their feedback and confirmation that these statements are accurate reflection of their experiences. The researcher will also keep a research journal to record reflexive notes, impressions of the data and thoughts about analysis throughout the process. Finally, the Framework Method facilitates an audit trail which will allow for greater scientific rigor and transparency within the analyzed data.

Section 6: Project Management / Timetable

6.1 Outline a timetable for completion of key stages of the project

E.g. ethics submission, start and end of data collection, data analysis, completion of systematic review

| | |
|--|-------------------------------------|
| Ethics submission | March 2016 |
| Consultation with NHS Staff | April 2016 |
| Preparation for interviews | |
| Recruitment and Data collection | May 2016 –February 2017 |
| Data analysis | May 2016– February 2017 |
| Completion of write up & systematic review | March 2017 |
| Thesis submission | Submission 1 st May 2017 |
| Viva | June 2017 |
| Corrections & Dissemination | July- Aug 2017 |

Section 7: Management of Risks to Project

7.1 Summarise the main potential risks to your study, the perceived likelihood of occurrence of these risks and any steps you will or have taken to reduce these risks. Outline how you will respond to identified risks if they should occur

1. *Difficulties that may be encountered as part of running focus interviews as a result of dynamics or symptoms experienced by population as a result of their condition*

PSE symptoms like uncontrollable crying or laughing can be intensely embarrassing for patients thus it is important to consider the impact that this may serve on the individual's readiness to engage. As a means of managing this risk, the facilitator will ensure that participants are fully informed of the content to be discussed, confidentiality will be assured and of their right to withdraw at any point without prejudice. They will also inform patients of the opportunity to arrange a debriefing session with the researcher should they wish to discuss issues that they may have become distressed by. Information about further support services will also be provided. The researcher has experience conducting interviews with stroke patients and so will use this knowledge to provide support when needed. Furthermore, these interviews will be facilitated NHS Lothian Hospital sites where experienced staff will be at hand should further assistance be required.

2. *The researcher's subjective bias in selecting items to include on the questionnaire*

Whilst subjectivity is often unavoidable in qualitative research, measures can be taken in order to minimise the impact of this. An additional independent researcher will be asked to analyse the data and mutually identified items will then be

incorporated into the scale. Subsequent to the interviews patients will be asked for feedback about the items added to the scale providing them with the opportunity to voice differences in opinion regarding the accuracy of statements in reflecting key cognitions. This measure will then undergo an additional consultation process with NHS clinicians to further mitigate any potential subjective bias.

3. *Data protection*

The data recorded will be frequently duplicated onto an external hard-drive that will be encrypted and stored in separate locations to minimise the risk of data loss. As this study will be a pilot the generalizability of results is extremely limited due to the small and heterogeneous participant sample. Nonetheless the researcher's strict adherence to qualitative and quantitative design and analysis protocols will aim to ensure the replicability and validity of the data obtained.

Section 8: Knowledge Exchange

8.1 How do you intend to report and disseminate the results of the study?

IRAS A51

As part of the requirements of the Clinical Psychology Doctorate thesis part of the reporting and dissemination of this project will take the form of a systematic review and journal article. These will be submitted to an appropriately identified relevant peer review journal so as to contribute to the current evidence base. The researcher intends to provide a verbal presentation of the results of this study to all NHS staff involved and other interested parties such as carers or other healthcare professionals. The researcher will also inform the participants of the study findings through means of a written letter or telephone call.

8.2 What are the anticipated benefits or implications of the project?

E.g. If this is an NHS project, in what way(s) is the project intended to benefit the NHS?

Despite the significant prevalence rates of PSE research into the psychological outcomes of those with the condition is in its infancy (Kneebone & Lincoln, 2012). Although studies make reference to the disabling impact it must have on an individual's functional abilities and emotional wellbeing, confirmation of this is entirely lacking (Hackett, Kohler, O'Brien & Mead, 2014). Theoretically patients may avoid situations in response to their appraisals of PSE symptoms further predisposing them to poor psychological outcomes (Picton, in preparation). The development of a measure that could illustrate these cognitions may be used as a predictive tool for future psychological distress and help to target treatments to those who need it.

This project may benefit the NHS in a number of ways. It could be used to assist staff in the early detection and monitoring of those with PSE who may be more vulnerable to psychological difficulties. These individuals could then be directed towards appropriate support at the early stages ensuring the timely delivery of psychological services and reducing the potential financial burden on the NHS. This study would further corroborate current SIGN 118 clinical practice guidelines that advocate for the early screening of suspected mood difficulties and the provision of information services to those affected by PSE (Gillespie, Joice, Lawrence & Whittick, 2011).

With the validation of this questionnaire future studies could then be facilitated to investigate causal links between patients' cognitions and rehabilitation or

psychosocial outcomes. Ultimately this study aims to extend the existing evidence base on PSE and further contribute towards a psychological understanding of this pervasive neurological condition.

8.3 Are there any potential costs for the project?

Outline any potential financial costs to the project, including the justification for the costs (why are these necessary for the research project?) and how funding will be obtained for these costs (how will they be met?) Please separate these into potential costs for the University and potential costs for your NHS Board and note that you should ask your NHS Board to meet stationery, printing, postage and travel costs.

Potential costs for University of Edinburgh:

The clinical tools listed above are routinely administered measures and so no financial cost will be required in gaining the rights to use these for the study. Interviews are time consuming and may be more effortful for those who have recently sustained a stroke (due to fatigue, increased cognitive load or emotional effort) therefore permission has been sought from the University of Edinburgh to fund a non-casher voucher as an incentive to take part.

Potential costs for NHS Lothian Health Board:

The costs to the NHS Lothian Health Board will involve printing and photocopying costs for the information and consent sheets and travel expenses for the researcher to the site of the interviews, this is currently being discussed with the researcher's line manager.

Section 11: Confirmation of Supervisors' Approval

"I confirm that both my Academic and Clinical Supervisors have seen and approved this research proposal and have both completed the supervisors' appraisal forms below."

| | |
|-----|----|
| Yes | No |
|-----|----|

Appendix I: Example of the process of coding

| Interview transcript | Line-by-line coding | Preliminary notes/ ideas | Initial codes |
|--|---|--|--|
| <p><u>Participant:</u></p> <p>“Very soon after my first stroke... it was about four months ago. It would just em... come on all of a sudden ... without any warning [pause] ... and I have no control. Any time someone spoke to me, I would just cry [pause] for no reason, if somebody tried to em ... comfort me [pause] I would cry. I feel like just crying all the time. [pause] for absolutely no reason... I don’t know why I cried.”</p> <p><u>Interviewer:</u></p> <p>“Ok and what was that like for you?”</p> <p><u>Participant:</u></p> <p>“Quite em ... confusing and worrying and em... strange eh.. I didn’t understand why it was happening. Sorta-bit frightened.”</p> <p>“My life will never be the same again. Em... my life is over [pause] em.. I won’t be able to go back to work.. em and that was really hard. Just about how em.. things have changed.”</p> | <p><i>Come on all of a sudden ... without any warning... for no reason... no control</i></p> <p><i>Any time someone spoke to me ... comfort me ... I would cry. Even when somebody said “hello” I would cry.</i></p> <p><i>Crying all the time...</i></p> <p><i>Confusing and worrying... strange... Sorta-bit frightened... Scared.</i></p> <p><i>I don’t know why I cried. I didn’t understand why it was happening...</i></p> <p><i>Never be the same again ... I won’t be able to go back to work..</i></p> <p><i>Nothing would’ve helped...</i></p> <p><i>Things have changed.</i></p> | <p>Sudden onset of emotional reactions. Lack of control.</p> <p>Other people can trigger emotionalism.</p> <p>The emotional impact of symptoms.</p> <p>Questioning/ trying to understand emotional reactions.</p> <p>Grieving the loss of old life</p> <p>Changes since the stroke</p> | <p>Emotional reactions are sudden and uncontrollable</p> <p>Triggers</p> <p>Enduring nature of emotional reactions</p> <p>The negative emotional impact of symptoms</p> <p>Uncertainty of the causes of emotionalism</p> <p>Loss</p> <p>Feeling hopeless</p> <p>Reflecting on changes since the stroke</p> |

Appendix J: The process of developing the analytical framework

| Initial codes | Initial themes | Charting themes | Global themes |
|---|---|---|---|
| Uncertainty about the causes for emotionalism Loss of control over reactions Easily emotional Emotional reactions are sudden and unpredictable Unexpected emotional reactions The unusual nature of emotional reactions Exaggerated emotional responses Enduring nature of emotional reactions | The nature of emotional reactions after stroke | <i>Making sense of emotionalism</i> | The spontaneous and uncontrollable nature of emotional reactions |
| Absence of triggers Emotive triggers Triggers The impact of others | Triggers | | |
| Lack of understanding from other people Fearing becoming emotional in public Gender differences in emotional expression Concern about what others think Worrying about upsetting others Fears of burdening people | Concern about others and their views | <i>Sociocultural views on emotion</i> | The stigma of expressed emotions |
| Mismatch with past emotional identity Incongruence Wishing to return to past self Self-criticism Not normal Feeling weak | Changes to self-concept after stroke | <i>The personal impact of stroke and emotionalism</i> | Incongruence (Subtheme: social incongruence) |
| Feeling disabled The negative emotional impact of symptoms Loss of confidence Feeling helpless | The emotional impact of stroke and emotionalism | | |
| People misunderstand reactions The impact of emotionalism on other people Embarrassment and guilt in public | Misinterpreting emotional reactions | | |

| | | | |
|---|---|---|--|
| <p>Making sense of emotional reactions Determined to overcome adversity Enthusiastic to get better Positive lifestyle changes Taking responsibility for own recovery Accepting limitations Sense of control Signs of recovery Maintaining optimism Humour Acceptance Self-encouragement Living in the moment Maintaining hope Ways of managing difficulties Avoiding</p> | <p>Ways of managing difficulties</p> | <p><i>Responding and adjusting to changes</i></p> | <p>Convalescence (Subtheme: Loss)</p> |
| <p>Stage of recovery Expectations about recovery Assumptions about stroke Uncertainty about the future</p> | <p>Assumptions around recovery</p> | | |
| <p>Reflecting on emotional changes since the stroke Reflecting on challenges Loss of sense of agency Grieving the loss of old roles Loss of independence Loss of emotional control Hopelessness Despair</p> | <p>Reflecting on changes since the stroke</p> | <p><i>Grieving the loss of old roles</i></p> | |



POST-STROKE EMOTIONALISM COGNITIONS QUESTIONNAIRE (PEC-Q)

Name: _____

Date: _____

| Please indicate the extent to which you agree or disagree with the following statements by ticking a box on the right column. | Strongly Disagree | Disagree | Neither agree or disagree | Agree | Strongly Agree |
|---|--------------------------|--------------------------|---------------------------|--------------------------|--------------------------|
| 1. I have no control over my laughter or crying. (T) | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 2. I worry that my laughter or crying will upset other people. (S) | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 3. I feel weak when I am emotional. (I) | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 4. When I'm emotional I feel unable to cope. (T) | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 5. I feel embarrassed when I become emotional in public. (S) | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 6. Being emotional just isn't me. (I) | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 7. I have ways to manage my emotionalism. (C) | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 8. I worry that other people will think that there is something wrong with me if I become emotional. (S) | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 9. I believe that emotionalism will improve over time. (C) | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 10. My inability to control my emotions is a sign that I am losing my mind. (T) | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 11. I try to avoid people or situations that might trigger my laughter or crying. (I) | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |

Appendix K: The Post-stroke Emotionalism Cognition Questionnaire

Dimensions:

1. The uncontrollable and spontaneous nature of emotional reactions (T)
2. incongruence (I)
3. Stigma of expressed emotion (S)
4. Convalescence (C)

Scoring

Higher scores = greater negative perceptions

Reverse score: 7, 9

| Rating | Score |
|-----------------------|--------------|
| Strongly disagree | 0 point |
| Disagree | 1 points |
| Unsure | 2 points |
| Agree | 3 points |
| Strongly agree | 4 points |
| Interpretation | |
| High range | 41-60 |
| Medium range | 21- 40 |
| Low range | 0-20 |
| Total score | |

Appendix L: Reflective journal entry

Reflections on the research process

The researcher was mindful of their dual role as qualitative researcher and trainee clinical psychologist. For the most part these roles worked harmoniously, for example the doctorate training equipped the researcher with clinical interviewing competencies. On occasion, the researcher needed to inhibit the desire to jump between roles into “therapist” mode in response to participant’s reactions or to the emotive nature of the interviews. These experiences were documented in the reflective diary.

Journal extract 21-07-2016:

“I find it difficult not to reflect, summarise or interpret what the participant is saying. I’m learning to remain open and curious and fighting the urge to give advice or suggestion.”

Another area for reflection was the researcher’s responses to the participant’s emotionalism, which often presented as intense and prolonged sobbing. The researcher struggled at times to remember that this was a symptom of emotionalism and not (always) of significant distress.

Journal extract 07-10-2017:

“I noticed that when the participant began to cry, my immediate urge was to comfort, reassure and try to minimise their distress. Yet they weren’t in distress. I smiled and laughed along when the participant wasn’t laughing out of happiness. I found that I reacted in these ways without realising it. I became aware that I was responding to the participant’s emotions as though they were congruent to their mood. I must make sure to ask participants how best to respond to their emotionalism.”

Appendix M: Participant debrief letter and questionnaire evaluation form



THE UNIVERSITY of EDINBURGH



Research study:

*The Post-Stroke Emotionalism Cognitions Questionnaire:
A development study.*

Dear

Thank you for taking part in my research project. I very much appreciate the time that you've taken the time to speak with me and share your experiences. During our meeting, we talked about what it is like to struggle with controlling your laughter or crying after stroke. I met with other individuals who also struggle with these difficulties. From these interviews, people listed 4 themes as being the most important for them when talking about their emotionalism.

Since their stroke, people described suddenly bursting into crying or laughing for very little reason. Everyone mentioned that they feel like they're not in control of their reactions when they become emotional.

Many people would not have described themselves as being an emotional person before their stroke. For them, this is a change of their character.

People described feeling embarrassed about being emotional in front of other people. The general public don't seem to understand emotionalism

Most people described helpful ways of coping with emotionalism like distraction, using humour or just letting it run it's course. Some people avoid certain situations or talking about things that may trigger their emotionalism.

Given that you have already contributed a lot to this project please do not feel under any obligation to respond. However, I would appreciate your feedback on a questionnaire that I've developed based on what was discussed during these interviews. Your feedback will be completely anonymous. Thank you very much for your time and I wish you all the best in your future.

Kind Regards,

A handwritten signature in black ink, appearing to be 'N. A.', with a horizontal line extending to the right.

Feedback on the questionnaire:

Are the statements clear and easy to understand? Yes No

If "No" how could the wording be improved?

**Do the statements accurately reflect the beliefs that Yes No
people may have about emotionalism?**

If "No" how could the statements be made more relevant?

How satisfied are you with the content of this questionnaire?

Not satisfied Unsure Satisfied

How could I improve this questionnaire?

Any further comments or suggestions?

Please send this page in the pre-stamped envelope that I've included in this letter to the following address:

Niamh McAleese
Trainee Clinical Psychologist
Department of Clinical Neurosciences (DCN)
Western General Hospital
Edinburgh
EH4 2XU

Thesis portfolio references

- Aben, I., Verhey, F., Lousberg, R., Lodder, J., & Honig, A. (2002). Validity of the Beck Depression Inventory, Hospital Anxiety and Depression Scale, SCL-90, and Hamilton Depression Rating Scale as screening instruments for depression in stroke patients. *Psychosomatics*, *43*(5), 386-393.
- Addo, J., Ayerbe, L., Mohan, K. M., Crichton, S., Sheldenkar, A., Chen, R., Wolfe, C.D. & McKeivitt, C. (2012). Socioeconomic status and stroke. *Stroke*, *43*(4), 1186-1191.
- Ahmed, A., & Simmons, Z. (2013). Pseudobulbar affect: prevalence and management. *The Clin Risk Manag*, *9*, 483-489.
- Alsén, P., Brink, E., Person, L. O., Brändström, Y., & Karlson, B. W. (2010). Illness perceptions after myocardial infarction: relations to fatigue, emotional distress, and health-related quality of life. *Journal of Cardiovascular Nursing*, *25*(2), 1-10.
- Avison, R. (2009). Discrepant illness perceptions in stroke survivor-partner dyads: relationship to psychological adjustment and expressed emotion. *Doctoral dissertation, University of Hull*, 53-100.
- Beck, J. S. (1979). *Cognitive therapy*. John Wiley & Sons, Inc..In Leahy, R. L., Holland, S. J., & McGinn, L. K. (2011). *Treatment plans and interventions for depression and anxiety disorders*. Guilford press.
- Benyamini, Y. (2011). Health and illness perceptions. *The Oxford Handbook of Health Psychology*, 281-314.

- Bergman, S., Jacobsson, L. T., Herrström, P., & Petersson, I. F. (2004). Health status as measured by SF-36 reflects changes and predicts outcome in chronic musculoskeletal pain: a 3-year follow up study in the general population. *Pain*, *108*(1), 115-123.
- Berry, E. (1998). Post-traumatic stress disorder after subarachnoid haemorrhage. *British Journal of Clinical Psychology*, *37*(3), 365-367.
- Bingeliene, A., Shapiro, C. M., Sockalingam, S., & Hawa, R. J. (2016). Pathological Laughing and Crying Post-stroke: Liaison Psychiatrist Beware. *Journal of Neuropsychiatry*.
- Black, N. (2013). Patient reported outcome measures could help transform healthcare. *BMJ (Clinical research ed)*, *346*-67.
- Bowlby J. Processes of mourning. (1961). The International journal of psychoanalysis. 1, 42,317 In Nelson, J. K. (1998). The meaning of crying based on attachment theory. *Clinical Social Work Journal*, *26*(1), 9-22.
- Brédart, A., Marrel, A., Abetz-Webb, L., Lasch, K., & Acquadro, C. (2014). Interviewing to develop Patient-Reported Outcome (PRO) measures for clinical research: eliciting patients' experience. *Health and Quality of Life Outcomes*, *12*(1), 15.
- Brennan, J. (2001). Adjustment to cancer—coping or personal transition? *Psycho-Oncology*, *10*(1), 1-18.
- Broadbent, E., Petrie, K. J., Main, J., & Weinman, J. (2006). The brief illness perception questionnaire. *Journal of Psychosomatic Research*, *60*(6), 631-637.

- Broadbent, E., Niederhoffer, K., Hague, T., Corter, A., & Reynolds, L. (2009). Headache sufferers' drawings reflect distress, disability and illness perceptions. *Journal of psychosomatic research*, 66(5), 465-470.
- Broadbent, E., Wilkes, C., Koschwanez, H., Weinman, J., Norton, S., & Petrie, K. J. (2015). A systematic review and meta-analysis of the Brief Illness Perception Questionnaire. *Psychology & health*, 30(11), 1361-1385.
- Burton, C. R. (2000). Re-thinking stroke rehabilitation: The Corbin and Strauss chronic illness trajectory framework. *Journal of Advanced Nursing*, 32(3), 595-602.
- Butler, E. A., Lee, T. L., & Gross, J. J. (2007). Emotion regulation and culture: are the social consequences of emotion suppression culture-specific?. *Emotion*, 7(1), 30.
- Cacioppo, J. T., & Hawkley, L. C. (2003). Social isolation and health, with an emphasis on underlying mechanisms. *Perspectives in Biology and Medicine*, 46(3), S39-S52.
- Calvert, T., Knapp, P., & House, A. (1998). Psychological associations with emotionalism after stroke. *Journal of Neurology, Neurosurgery & Psychiatry*, 65(6), 928-929.
- Campbell Burton, C., Murray, J., Holmes, J., Astin, F., Greenwood, D., & Knapp, P. (2013). Frequency of anxiety after stroke: a systematic review and meta-analysis of observational studies. *International Journal of Stroke*, 8(7), 545-559.
- Carota, A., Berney, A., Aybek, S., Iaria, G., Staub, F., Ghika-Schmid, F., Bogousslavsky, J. (2005). A prospective study of predictors of poststroke depression. *Neurology*, 64(3), 428-433.

- Carota, A., & Calabrese, P. (2013). Post stroke Emotionalism. *Journal of Neurological Disorders*. 1(4), 1-4.
- Carron, S. F., Alwis, D. S., & Rajan, R. (2016). Traumatic brain injury and neuronal functionality changes in sensory cortex. *Frontiers in Systems Neuroscience*, 10.
- Centre for reviews, dissemination (CRD). Systematic reviews: CRD's guidance for undertaking reviews in health care. Centre for Reviews and Dissemination; 2009. Accessed May 2017 from: https://www.york.ac.uk/media/crd/Systematic_Reviews.pdf.
- Champion, A. J. (2006). *Neuropsychological rehabilitation: a resource for group-based education and intervention*. John Wiley & Sons.
- Chowdhury, R. N., Hasan, A. H., Rahman, Y. U., Khan, S. I., Hussain, A. R., & Ahsan, S. (2014). Pattern of neurological disease seen among patients admitted in tertiary care hospital. *BMC research notes*, 7(1), 202.
- Coetzer, R. (2006). *Traumatic brain injury rehabilitation: A psychotherapeutic approach to loss and grief*. Nova Publishers.
- Colamonic, J., Formella, A., & Bradley, W. (2012). Pseudobulbar affect: burden of illness in the USA. *Advances in Therapy*, 1-24.
- Comper, P., Bisschop, S. M., Carnide, N., & Tricco, A. (2005). A systematic review of treatments for mild traumatic brain injury. *Brain Injury*, 19(11), 863-880.
- Coole, C., Radford, K., Grant, M., & Terry, J. (2013). Returning to work after stroke: perspectives of employer stakeholders, a qualitative study. *Journal of Occupational Rehabilitation*, 23(3), 406-418.

- Cooper, A. F., Jackson, G., Weinman, J., & Horne, R. (2002). Factors associated with cardiac rehabilitation attendance: a systematic review of the literature. *Clinical Rehabilitation*, 16(5), 541-552.
- Corrigan, J. D., Selassie, A. W., & Orman, J. A. L. (2010). The epidemiology of traumatic brain injury. *The Journal of Head Trauma Rehabilitation*, 25(2), 72-80.
- Crowe, C., Coen, R.F., Kidd, N., Hevey, D., Cooney, J., Harbison, J. (2016). A qualitative study of the experience of psychological distress post-stroke. *Journal of Health Psychology*. 21(11), 2572-9.
- Cummings, J. L., Arciniegas, D. B., Brooks, B. R., Herndon, R. M., Lauterbach, E. C., Piro, E. P., ... & Weintraub, D. (2006). Defining and diagnosing involuntary emotional expression disorder. *CNS spectrums*, 11(S6), 1-11.
- Dempster, M., McCorry, N. K., Brennan, E., Donnelly, M., Murray, L. J., & Johnston, B. T. (2011). Do changes in illness perceptions predict changes in psychological distress among oesophageal cancer survivors? *Journal of Health Psychology*, 16(3), 500-509.
- Dinsmore, J.G. (2010). Quality of life and illness perception in Stroke Patients: A 12 Month Study. *Doctoral dissertation, Queen's University Belfast*.
- di Ruffano, L. F., Dinnes, J., Sitch, A. J., Hyde, C., & Deeks, J. J. (2017). Test-treatment RCTs are susceptible to bias: a review of the methodological quality of randomized trials that evaluate diagnostic tests. *BMC medical research methodology*, 17(1), 35.

- Dowswell, G., Lawler, J., Dowswell, T., Young, J., Forster, A., & Hearn, J. (2000). Investigating recovery from stroke: a qualitative study. *Journal of Clinical Nursing*, 9(4), 507-515.
- Ellis-Hill, C. S., Payne, S., & Ward, C. (2000). Self-body split: issues of identity in physical recovery following a stroke. *Disability and rehabilitation*, 22(16), 725-733.
- Engelman, W., Hammond, F. M., & Malec, J. F. (2014). Diagnosing pseudobulbar affect in traumatic brain injury. *Neuropsychiatric disease and treatment*, 10, 190-3.
- Evans, S. (2008). Stroke, psychological outcomes and the self-regulatory model. *Doctoral dissertation, Lancaster University*, 53-100.
- Foxwell, R., Morley, C., & Frizelle, D. (2013). Illness perceptions, mood and quality of life: A systematic review of coronary heart disease patients. *Journal of Psychosomatic Research*, 75(3), 211-222.
- Friedman, H. S. (2011). Personality, disease, and self-healing. *The Oxford handbook of health psychology*, pg.215-240.
- Gale, N. K., Heath, G., Cameron, E., Rashid, S., & Redwood, S. (2013). Using the framework method for the analysis of qualitative data in multi-disciplinary health research. *BMC*, 13(1), 117.
- Gibbs, G. R. (2008). *Analysing qualitative data*. Sage Publishers.
- Gilbert, P. (2009). Introducing compassion-focused therapy. *Advances in psychiatric treatment*, 15(3), 199-208.
- Gilbert, P., & Procter, S. (2006). Compassionate mind training for people with high shame and self-criticism: Overview and pilot study of a

- group therapy approach. *Clinical Psychology & Psychotherapy*, 13(6), 353-379.
- Gillespie, D. C., Cadden, A. P., Lees, R., West, R. M., & Broomfield, N. M. (2016). Prevalence of Pseudobulbar Affect following Stroke: A Systematic Review and Meta-Analysis. *Journal of Stroke and Cerebrovascular Diseases*, 25(3), 688-694.
- Goldstein, L. H., Holland, L., Soteriou, H., & Mellers, J. D. (2005). Illness representations, coping styles and mood in adults with epilepsy. *Epilepsy Research*, 67(1), 1-11.
- Gómez-de-Regil, L., Kwapil, T. R., & Barrantes-Vidal, N. (2014). Predictors of expressed emotion, burden and quality of life in relatives of Mexican patients with psychosis. *Journal of psychiatric and mental health nursing*, 21(2), 170-179.
- Gould, R. A., Safren, S. A., Washington, D. O., & Otto, M. W. (2004). A meta-analytic review of cognitive- behavioural treatments. *Generalized anxiety disorder: Advances in research and practice*, 248-264.
- Granerod, J., Ambrose, H. E., Davies, N. W., Clewley, J. P., Walsh, A. L., Morgan, D. & Ward, K. N. (2010). Causes of encephalitis and differences in their clinical presentations in England: a multicentre, population-based prospective study. *The Lancet*, 10(12), 835-844.
- Grice, S. J., Kuipers, E., Bebbington, P., Dunn, G., Fowler, D., Freeman, D., & Garety, P. (2009). Carers' attributions about positive events in psychosis relate to expressed emotion. *Behaviour research and therapy*, 47(9), 783-789.

- Hackett, M. L., Yang, M., Anderson, C. S., Horrocks, J. A., & House, A. (2010). Pharmaceutical interventions for emotionalism after stroke. *Stroke*, *41*(7), e460-e461.
- Hackett, M. L., Anderson, C. S., House, A. O., & Xia, J. (2009). Interventions for treating depression after stroke. *Stroke*, *40*(7), 487.
- Hagger, M. S., & Orbell, S. (2003). A meta-analytic review of the common-sense model of illness representations. *Psychology and health*, *18*(2), 141-184.
- Hale, E. D., Treharne, G. J., & Kitas, G. D. (2007). The Common-Sense Model of self-regulation of health and illness: how can we use it to understand and respond to our patients' needs? *Rheumatology*, *46*; 904-906.
- Harris, C. (2014). Return to Work After Stroke. *Stroke*, *45*(9), 174-6.
- Harman, G., & Clare, L. (2006). Illness representations and lived experience in early-stage dementia. *Qualitative Health Research*, *16*(4), 484-502.
- Hartling, L., Milne, A., Hamm, M. P., Vandermeer, B., Ansari, M., Tsertsvadze, A., & Dryden, D. M. (2013). Testing the Newcastle Ottawa Scale showed low reliability between individual reviewers. *Journal of Clinical Epidemiology*, *66*(9), 982-993.
- Higgins, J. P., & Green, S. (Eds.). (2011). *Cochrane Handbook for Systematic Reviews of Interventions* (Vol. 4). John Wiley & Sons.

- Horrocks, J. A., Hackett, M. L., Anderson, C. S., & House, A. O. (2004). Pharmaceutical interventions for emotionalism after stroke. *Stroke*, 35(11), 2610-2611.
- House, A. Knapp, P. & Calvert, T. (1989). Emotionalism after stroke. *BMJ*. 298, 991-994.
- House, A. & Hosker, C. (2013). Emotional disorders in neurological rehabilitation. *Handbook of Clinical Neurology*. 10, 110:389.
- Hou, R., Moss-Morris, R., Peveler, R., Mogg, K., Bradley, B. P., & Belli, A. (2012). When a minor head injury results in enduring symptoms: a prospective investigation of risk factors for post-concussional syndrome after mild traumatic brain injury. *Journal of Neurology, Neurosurgery & Psychiatry*, 83(2), 217-223.
- Jain, S. (2014). Pseudobulbar affect: No laughing matter. *Current Psychiatry*, 13(4), 66.
- Jones, K. M., Kydd, R., Broadbent, E., Theadom, A., Barker-Collo, S., Edwards, H., & Feigin, V. L. (2016). Brain drawings following traumatic brain injury (TBI) and links to illness perceptions and health outcomes—Findings from a population-based study. *Psychology & Health*, 31(10), 1182-1202.
- Jonsbu, E., Martinsen, E. W., Morken, G., Moum, T., & Dammen, T. (2013). Change and impact of illness perceptions among patients with non-cardiac chest pain or benign palpitations following three sessions of CBT. *Behavioural and Cognitive Psychotherapy*, 41(04), 398-407.

- Kaptein, A. A., Scharloo, M., Helder, D. I., Snoei, L., Van Kempen, G. M. J., Weinman, J., Houwelingen, J.C. & Roos, R. A. C. (2007). Quality of life in couples living with Huntington's disease: the role of patients' and partners' illness perceptions. *Quality of Life Research*, 16(5), 793-801.
- Kasprisin, Arlene. (2004). "Alternative cognitive therapy for emotional instability (pathologic laughing and crying)." *Physical medicine and rehabilitation clinics of North America* 15,4, 883-917.
- Kerr, G.D, Slavin, H., Clark, D., Coupar, F., Langhorne, P.& Stott, D.J (2011). Do vascular risk factors explain the association between socioeconomic status and stroke incidence: a meta-analysis. *Cerebrovascular Diseases*.31(1), 57-63.
- Keltner, D. & Gross, J.J. (1999). Functional accounts of emotions. *Cognition & Emotion*. 13(5), 467-80.
- King, N. S. (2003). Post-concussion syndrome: clarity amid the controversy?
- King, N. S., & Kirwilliam, S. (2011). Permanent post-concussion symptoms after mild head injury. *Brain injury*, 25(5), 462-470.
- Kirkevold, M. (2002). The unfolding illness trajectory of stroke. *Disability and Rehabilitation*, 24(17), 887-898.
- Kneebone, I. I., & Lincoln, N. B. (2012). Psychological problems after stroke and their management: state of knowledge. *Neuroscience and Medicine*, 3(1), 83.
- Kreutzer, J. S., Rapport, L. J., Marwitz, J. H., Harrison-Felix, C., Hart, T., Glenn, M., & Hammond, F. (2009). Caregivers' well-being after

- traumatic brain injury: a multi-center prospective investigation. *Archives of Physical Medicine and Rehabilitation*, 90(6), 939-946.
- Laidlaw, K. (2008). Post-stroke depression and CBT with older people. In *Handbook of behavioral and cognitive therapies with older adults* (pp. 233-248). Springer New York.
- Lazarus, R. S. (1993). Coping theory and research: past, present, and future. *Psychosomatic medicine*, 55(3), 234-247.
- Leahy, R. L., Holland, S. J., & McGinn, L. K. (2011). *Treatment plans and interventions for depression and anxiety disorders*. Guilford press.
- Leventhal H, Meyer D, Nerenz D. (1980). The common-sense representation of illness danger. *Contributions to Medical Psychology*. 2, 7-30.
- Leventhal, H., Benyamini, Y., Brownlee, S., Diefenbach, M., Leventhal, E. A., Patrick-Miller, L., & Robitaille, C. (1997). Illness representations: theoretical foundations. *Perceptions of Health and Illness*, 2, 19-46.
- Leventhal, H., Leventhal, E. A., & Contrada, R. J. (1998). Self-regulation, health, and behavior: A perceptual-cognitive approach. *Psychology and Health*, 13(4), 717-733.
- Lincoln, N. B., & Flannaghan, T. (2003). Cognitive behavioral psychotherapy for depression following stroke. *Stroke*, 34(1), 111-115.
- MacInnes, J. D. (2006). The illness perceptions of women following symptoms of acute myocardial infarction: a self-regulatory

- approach. *European Journal of Cardiovascular Nursing*, 5(4), 280-288.
- Maruzairi, H., & Koh, C. H. (2015). Pseudobulbar affect post stroke. *Brunei Int.Med.J.* 11(1), 59-61
- Mathuranath, P. S., Nestor, P. J., Berrios, G. E., Rakowicz, W., & Hodges, J. R. (2000). A brief cognitive test battery to differentiate Alzheimer's disease and frontotemporal dementia. *Neurology*, 55(11), 1613-1620.
- 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 Neurological Sciences*, 169(1), 43-48.
- McCullagh, S., & Feinstein, A. (2003). Outcome after mild traumatic brain injury: an examination of recruitment bias. *Journal of Neurology, Neurosurgery & Psychiatry*, 74(1), 39-43.
- Meares, S., Shores, E. A., Batchelor, J., Baguley, I. J., Chapman, J., Gurka, J., & Marosszeky, J. E. (2006). The relationship of psychological and cognitive factors and opioids in the development of the post-concussion syndrome in general trauma patients with mild traumatic brain injury. *Journal of the International Neuropsychological Society*, 12(06), 792-801.
- Medley, A. R., Powell, T., Worthington, A., Chohan, G., & Jones, C. (2010). Brain injury beliefs, self-awareness, and coping: a preliminary cluster analytic study based within the self-regulatory model. *Neuropsychological rehabilitation*, 20(6), 899-921.

- Mesquita, B., & Walker, R. (2003). Cultural differences in emotions: A context for interpreting emotional experiences. *Behaviour Research and Therapy*, 41(7), 777-793.
- McAleese N, Gillespie D, O'Rourke S & Guzman A. A systematic review of illness perceptions, coping and health outcomes in adults with sudden onset neurological conditions. PROSPERO 2017: CRD42017054487 Available from http://www.crd.york.ac.uk/PROSPERO/display_record.asp?ID=CRD42017054487
- Miller, L. (1993). The "trauma" of head trauma: Clinical, neuropsychological, and forensic aspects of posttraumatic stress syndromes in brain injury. *Journal of Cognitive Rehabilitation*.
- Mioshi, E., Dawson, K., Mitchell, J., Arnold, R., & Hodges, J. R. (2006). The Addenbrooke's Cognitive Examination Revised (ACE-R): a brief cognitive test battery for dementia screening. *International journal of geriatric psychiatry*, 21(11), 1078-1085.
- Modabbernia, A., Poustchi, H., & Malekzadeh, R. (2013). Neuropsychiatric and psychosocial issues of patients with hepatitis C infection: a selective literature review. *Hepatitis monthly*, 13(1).
- Morris, J. C., Storandt, M., Miller, J. P., McKeel, D. W., Price, J. L., Rubin, E. H., & Berg, L. (2001). Mild cognitive impairment represents early-stage Alzheimer disease. *Archives of Neurology*, 58(3), 397-405.
- Moss-Morris, R., Weinman, J., Petrie, K., Horne, R., Cameron, L., & Buick, D. (2002). The revised illness perception questionnaire (IPQ-R). *Psychology and health*, 17(1), 1-16.

- Nasreddine, Z. S., Phillips, N. A., Bédirian, V., Charbonneau, S., Whitehead, V., Collin, I., Cummings, J.L & Chertkow, H. (2005). The Montreal Cognitive Assessment, MoCA: a brief screening tool for mild cognitive impairment. *Journal of the American Geriatrics Society*, 53(4), 695-699.
- National Institute for Clinical Health and Excellence. Clinical guideline CG176: Head injury: assessment and early management. 2014. Accessed May 2017 from: <https://www.nice.org.uk/guidance/cg176/chapter/3-Otherinformation>.
- Nelson, J. K. (1998). The meaning of crying based on attachment theory. *Clinical Social Work Journal*, 26(1), 9-22.
- Nicholl, C. R., Lincoln, N. B., Muncaster, K., & Thomas, S. (2002). Cognitions and post-stroke depression. *British Journal of Clinical Psychology*, 41(3), 221-231.
- Nogueira, M., & Teixeira, M. J. (2012). Central pain due to stroke: cognitive representation and coping according to gender. *Arquivos de neuro-psiquiatria*, 70(2), 125-128.
- O'Donovan, C. E., Painter, L., Lowe, B., Robinson, H., & Broadbent, E. (2016). The impact of illness perceptions and disease severity on quality of life in congenital heart disease. *Cardiology in the Young*, 26(01), 100-109.
- Parkinson, S., Eatough, V., Holmes, J., Stapley, E., & Midgley, N. (2016). Framework analysis: A worked example of a study exploring young people's experiences of depression. *Qualitative Research in Psychology*, 13(2), 109-129.

- Patchick, E. L., Horne, M., Woodward-Nutt, K., Vail, A., & Bowen, A. (2015). Development of a patient-centred, patient-reported outcome measure (PROM) for post-stroke cognitive rehabilitation: qualitative interviews with stroke survivors to inform design and content. *Health Expectations, 18*(6), 3213-3224.
- Perotti, L. P., Cummings, L. D., & Mercado, J. (2016). Behavioral Treatment of Pseudobulbar Affect: A Case Report. *Perspectives in Psychiatric Care, 52*(2), 82-87.
- Petrie, K. J., & Weinman, J. (1997). *Perceptions of health and illness: current research and applications*. Taylor & Francis.
- Petrie, K. J., Cameron, L. D., Ellis, C. J., Buick, D., & Weinman, J. (2002). Changing illness perceptions after myocardial infarction: an early intervention randomized controlled trial. *Psychosomatic Medicine, 64*(4), 580-586.
- Petrie, K. J., Jago, L. A., & Devcich, D. A. (2007). The role of illness perceptions in patients with medical conditions. *Current Opinion in Psychiatry, 20*(2), 163-167.
- Petrie, K. J., & Weinman, J. (2012). Patients' perceptions of their illness: The dynamo of volition in health care. *Current Directions in Psychological Science, 21*(1), 60-65.
- Picton, H. (2014). 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' conceptualisation of PSEL. Edinburgh Research Archive. Accessed May 2017 from: <https://www.era.lib.ed.ac.uk/handle/1842/9760>

- Powell, J., Kitchen, N., Heslin, J., & Greenwood, R. (2002). Psychosocial outcomes at three and nine months after good neurological recovery from aneurysmal subarachnoid haemorrhage: predictors and prognosis. *Journal of Neurology, Neurosurgery & Psychiatry*, 72(6), 772-781.
- Ritchie, J., Lewis, J., Nicholls, C. M., & Ormston, R. (Eds.). (2013). *Qualitative research practice: A guide for social science students and researchers*. Sage.
- Rogan, C., Fortune, D. G., & Prentice, G. (2013). Post-traumatic growth, illness perceptions and coping in people with acquired brain injury. *Neuropsychological Rehabilitation*, 23(5), 639-657.
- Ryan, L. M., & Warden, D. L. (2003). Post-concussion syndrome. *International review of psychiatry*, 15(4), 310-316.
- Sacco, S., Sarà, M., Pistoia, F., Conson, M., Albertini, G., & Carolei, A. (2008). Management of pathologic laughter and crying in patients with locked-in syndrome: a report of 4 cases. *Archives of Physical Medicine and Rehabilitation*, 89(4), 775-778.
- Salisbury, L., Wilkie, K., Bulley, C., & Shiels, J. (2010). 'After the stroke': patients' and carers' experiences of healthcare after stroke in Scotland. *Health & social care in the community*, 18(4), 424-432.
- Saltapidas, H., & Ponsford, J. (2008). The influence of cultural background on experiences and beliefs about traumatic brain injury and their association with outcome. *Brain Impairment*, 9(01), 1-13.

- Salter, K., Hellings, C., Foley, N., & Teasell, R. (2008). The experience of living with stroke: a qualitative meta-synthesis. *Journal of Rehabilitation Medicine, 40*(8), 595-602.
- Sangha, H., Lipson, D., Foley, N., Salter, K., Bhogal, S., Pohani, G., & Teasell, R. W. (2005). A comparison of the Barthel Index and the Functional Independence Measure as outcome measures in stroke rehabilitation: patterns of disability scale usage in clinical trials. *International Journal of Rehabilitation Research, 28*(2), 135-139.
- Sarre, S., Redlich, C., Tinker, A., Sadler, E., Bhalla, A., & McKeivitt, C. (2014). A systematic review of qualitative studies on adjusting after stroke: lessons for the study of resilience. *Disability and Rehabilitation, 36*(9), 716-726.
- Satink, T., Cup, E. H., Ilott, I., Prins, J., de Swart, B. J., & Nijhuis-van der Sanden, M. W. (2013). Patients' views on the impact of stroke on their roles and self: a thematic synthesis of qualitative studies. *Archives of Physical Medicine and Rehabilitation, 94*(6), 1171-1183.
- Schoormans D, Mulder BJ, van Melle JP, Pieper PG, van Dijk AP, Sieswerda GT, Hulsbergen-Zwarts MS, Plokker TH, Brunninkhuis LG, Vliegen HW, Sprangers MA. (2014). Illness perceptions of adults with congenital heart disease and their predictive value for quality of life two years later. *European Journal of Cardiovascular Nursing. 13*(1), 86-94.

Scottish Intercollegiate Guidelines Network (SIGN). SIGN 50: a guideline developer's handbook. Edinburgh: SIGN; 2015. (SIGN pub.no. 50).

Accessed May 2017 from:

<http://www.sign.ac.uk/pdf/sign50.pdf>.

Scottish Intercollegiate Guidelines Network (SIGN). SIGN 118:

Management of patients with stroke: Rehabilitation, prevention and management of complications, and discharge planning A national clinical guideline 2010. Retrieved on 9th May from:

<http://www.sign.ac.uk/pdf/sign118.pdf>

Searle, A., Norman, P., Thompson, R., & Vedhara, K. (2007). A prospective examination of illness beliefs and coping in patients with type 2 diabetes. *British journal of health psychology*, 12(4), 621-638.

Schneider, M. A., & Schneider, M. D. (2017). Pseudobulbar Affect: What Nurses, Stroke Survivors, and Caregivers Need to Know. *Journal of Neuroscience Nursing*, 49(2), 114-117.

Shamir M, Travis J. Boys don't cry? (2002). *Rethinking narratives of masculinity and emotion in the US*. Columbia University Press.

Sheldrick, R., TARRIER, N., BERRY, E., & KINCEY, J. (2006). Post-traumatic stress disorder and illness perceptions over time following myocardial infarction and subarachnoid haemorrhage. *British Journal of Health Psychology*, 11(3), 387-400.

Siemonsma, P. C., Stuive, I., Roorda, L. D., Vollebregt, J. A., Walker, M. F., Lankhorst, G. J., & Lettinga, A. T. (2013). Cognitive treatment

of illness perceptions in patients with chronic low back pain: a randomized controlled trial. *Physical Therapy*, 93(4), 435.

Silverberg, N. D., & Iverson, G. L. (2011). Etiology of the post-concussion syndrome: physiogenesis and psychogenesis revisited. *Neuro Rehabilitation*, 29(4), 317-329.

Sjölander, M., Eriksson, M., & Glader, E. L. (2013). The association between patients' beliefs about medicines and adherence to drug treatment after stroke: a cross-sectional questionnaire survey. *BMJ open*, 3(9), e003551.

Smith, J. A., and V. Eatough. (2007) *Analysing qualitative data in psychology*. London: Sage, pg.35-50.

Smith, J., & Firth, J. (2011). Qualitative data analysis: the framework approach. *Nurse Researcher*, 18(2), 52-62.

Snell, D., & Surgenor, L. (2006). An analysis of referees and referrals to a specialist concussion clinic in New Zealand. *The New Zealand Medical Journal (Online)*, 119(1231).

Snell, D. L., Siegert, R. J., Hay-Smith, E. J. C., & Surgenor, L. J. (2011). Associations between illness perceptions, coping styles and outcome after mild traumatic brain injury: preliminary results from a cohort study. *Brain Injury*, 25(11), 1126-1138.

Snell, D. L., Hay-Smith, E. J. C., Surgenor, L. J., & Siegert, R. J. (2013). Examination of outcome after mild traumatic brain injury: The contribution of injury beliefs and Leventhal's Common Sense Model. *Neuropsychological Rehabilitation*, 23(3), 333-362.

- Snell, D. L., Surgenor, L. J., Hay-Smith, E. J. C., Williman, J., & Siegert, R. J. (2015). The contribution of psychological factors to recovery after mild traumatic brain injury: Is cluster analysis a useful approach? *Brain Injury, 29*(3), 291-299.
- Soo, C., & Tate, R. L. (2007). Psychological treatment for anxiety in people with traumatic brain injury. *The Cochrane Library*.
- Stack, R. J., Simons, G., Kumar, K., Mallen, C. D., & Raza, K. (2013). Patient delays in seeking help at the onset of rheumatoid arthritis: the problem, its causes and potential solutions. *Aging Health, 9*(4), 425-435.
- Streiner, D. L., Norman, G. R., & Cairney, J. (2014). *Health measurement scales: a practical guide to their development and use*. Oxford University Press, USA.
- Stroke Association. State of the Nation: stroke statistics. 2017. Retrieved 05 May 2017 https://www.stroke.org.uk/sites/default/files/state_of_the_nation_2017_final_1.pdf
- Stowd, R. E., Cartwright, M. S., Okun, M. S., Haq, I., & Siddiqui, M. S. (2010). Pseudobulbar affect: prevalence and quality of life impact in movement disorders. *Journal of Neurology, 257*(8), 1382-1387.
- Svedbo Engström, M., Leksell, J., Johansson, U. B., & Gudbjörnsdóttir, S. (2016). What is important for you? A qualitative interview study of living with diabetes and experiences of diabetes care to establish a basis for a tailored Patient-Reported Outcome Measure for the Swedish National Diabetes Register. *BMJ open, 6*(3).

- Tateno, A., Jorge, R. E., & Robinson, R. G. (2004). Pathological laughing and crying following traumatic brain injury. *The Journal of Neuropsychiatry and Clinical Neurosciences*, 16(4), 426-434.
- Taylor, G. H., Todman, J., & Broomfield, N. M. (2011). Post-stroke emotional adjustment: A modified social cognitive transition model. *Neuropsychological Rehabilitation*, 21(6), 808-824.
- Teoh, V., Sims, J., & Milgrom, J. (2009). Psychosocial predictors of quality of life in a sample of community-dwelling stroke survivors: a longitudinal study. *Topics in Stroke Rehabilitation*, 16(2), 157-166.
- Thomas, S. A., & Lincoln, N. B. (2006). Factors relating to depression after stroke. *British Journal of Clinical Psychology*, 45(1), 49-61.
- Twiddy, M. (2008). *Beliefs about stroke: negotiating shared understandings* (Doctoral dissertation, University of Leeds).
- Twiddy, M., House, A., & Jones, F. (2012). The association between discrepancy in illness representations on distress in stroke patients and carers. *Journal of Psychosomatic Research*, 72(3), 220-225.
- Van Mierlo, M. L., Van Heugten, C. M., Post, M. W., De Kort, P. L., & Visser-Meily, J. M. (2015). Psychological factors determine depressive symptomatology after stroke. *Archives of Physical Medicine and Rehabilitation*, 96(6), 1064-1070.
- van Mierlo, M. L., van Heugten, C. M., Post, M. W., Hajós, T. R., Kappelle, L. J., & Visser-Meily, J. M. (2016). Quality of life during the first two years post stroke: the Restore4Stroke cohort study. *Cerebrovascular Diseases*, 41(1-2), 19-26.

- Var, F. A., & Rajeswaran, J. (2012). Perception of illness in patients with traumatic brain injury. *Indian Journal of Psychological Medicine*, 34(3), 223.
- Var FA, Rajeswaran J. (2013). Quality of life and perception of illness in patients with traumatic brain injury. *The Indian Journal of Neurotrauma*. 10(1), 115-119.
- Vaughan, R., Morrison, L., & Miller, E. (2003). The illness representations of multiple sclerosis and their relations to outcome. *British Journal of Health Psychology*, 8(3), 287-301.
- Vestling, M., Tufvesson, B., & Iwarsson, S. (2003). Indicators for return to work after stroke and the importance of work for subjective well-being and life satisfaction. *Journal of Rehabilitation Medicine*, 35(3), 127-131.
- Viswanathan, M., Ansari, M. T., Berkman, N. D., Chang, S., Hartling, L., McPheeters, M., Santaguida, P.L, Shamliyan, T., Singh, K., Tsertsvadze, A. & Treadwell, J. R. (2012). Assessing the risk of bias of individual studies in systematic reviews of health care interventions. Accessed May 2017 from www.effectivehealthcare.ahrq.gov/
- Wade, DT., Langton Hower, R., Skilbeck, CE., & David, RM. (1985). Stroke: A critical approach to diagnosis, treatment and management. London: Chapman and Hall Medical.
- Wade, D. T., & Collin, C. (1988). The Barthel ADL Index: a standard measure of physical disability? *International Disability Studies*, 10(2), 64-67.
- Wade DT. (1992). Measurement in neurological rehabilitation. *Current*

Opinion in Neurology. 5(5), 682-6.

Wade, D. (2015). Rehabilitation—a new approach. Overview and Part One: the problems. *Clinical Rehabilitation*. 29(11):1041-50

Waldron, B., Casserly, L. M., & O'Sullivan, C. (2013). Cognitive behavioural therapy for depression and anxiety in adults with acquired brain injury. What works for whom? *Neuropsychological Rehabilitation*, 23(1), 64-101.

Wei, N., Yong, W., Li, X., Zhou, Y., Deng, M., Zhu, H., & Jin, H. (2015). Post-stroke depression and lesion location: a systematic review. *Journal of Neurology*, 262(1), 81-90.

Weinman, J., Petrie, K. J., Moss-Morris, R., & Horne, R. (1996). The illness perception questionnaire: a new method for assessing the cognitive representation of illness. *Psychology and Health*, 11(3), 431-445.

Weiss, J. J., Konstantinidis, I., Boueilh, A., Fierer, D. S., Gardenier, D., Barber, M. G., ... & Yostos, M. P. (2016). Illness perceptions, medication beliefs, and adherence to antiretrovirals and medications for comorbidities in adults with HIV infection and hypertension or chronic kidney disease. *JAIDS Journal of Acquired Immune Deficiency Syndromes*, 73(4), 403-410.

Wells, G.A., Shea B., O'Connell D., Peterson, J., Welch, V., Losos, M. & Tugwell, P. The Newcastle-Ottawa Scale (NOS) for assessing the quality of non-randomised studies in meta-analyses. Ottawa HospitalResearchInstitute. Accessed May 2017: http://www.ohri.ca/programs/clinical_epidemiology/oxford.asp .

- Whitmarsh, A., Koutantji, M., & Sidell, K. (2003). Illness perceptions, mood and coping in predicting attendance at cardiac rehabilitation. *British Journal of Health Psychology, 8*(2), 209-221.
- Whittaker, R., Kemp, S., & House, A. (2007). Illness perceptions and outcome in mild head injury: a longitudinal study. *Journal of Neurology, Neurosurgery & Psychiatry, 78*(6), 644-646.
- Work, S. S., Colamonico, J. A., Bradley, W. G., & Kaye, R. E. (2011). Pseudobulbar affect: an under-recognized and under-treated neurological disorder. *Advances in Therapy, 28*(7), 586-601.
- World Health Organization. The world health report 2002: reducing risks, promoting healthy life. World Health Organization. 2002. Accessed May2017from:
http://www.who.int/whr/2002/en/whr02_en.pdf?ua=1
- Zigmond, A. S., & Snaith, R. P. (1983). The hospital anxiety and depression scale. *Acta Psychiatrica Scandinavica, 67*(6), 361-370.

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| | brain. <i>J Integr Neurosci</i> . 2004;3(3):253–266. |
| Two authors | Prokai-Tatrai K, Prokai L. Modifying peptide properties by prodrug design for enhanced transport into the CNS. <i>Prog Drug Res</i> . 2003;61:155–188. |
| Three authors | Veronesi U, Maisonneuve P, Decensi A. Tamoxifen: an enduring star. <i>J Natl Cancer Inst</i> . 2007;99(4):258–260. |
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