Post-stroke emotionalism (PSE): Making sense of neurological, psychological and experiential factors

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Thesis portfolio abstract

Background: Emotionalism is a condition which arises following a range of neurological disorders including multiple sclerosis, amyotrophic lateral sclerosis, traumatic brain injury, and stroke. It is characterised by episodes of uncontrollable crying or laughter, not under usual control and which are disproportionate or inappropriate to the social context.

Aim: This thesis aimed to explore the neurological, psychological and experiential factors of emotionalism in neurological disorders.

Method: Firstly, a systematic review was conducted to investigate the neurophysiological, neuropsychological and psychological predictors and correlates of emotionalism across neurological disorders. A narrative synthesis and additional analyses were completed to explore differences between predictors and correlates across neurological disorders. Secondly, an empirical paper explored individuals experience of emotionalism following stroke, over time. A qualitative longitudinal analysis of interview data was completed two-weeks, six-months and 12-months post-stroke.

Results: The systematic review highlighted predictors and correlates found across several neurological disorders including bulbar networks, serotonergic pathways, genetics and psychological impact. However, the majority of research focused on stroke populations whereby further research is required across neurological disorders to enable definitive answers. In the empirical paper, three main experiential themes of post-stroke emotionalism were identified: '*In the moment*', '*Ways of coping*' and '*Impact*'. Participants with more negative experiences of emotionalism described higher frequencies of avoidance, negative beliefs, distress, embarrassment and found it hard to describe their experience.

Conclusions: This thesis portfolio highlights the neurological, psychological and experiential factors of emotionalism, which helps to further our understanding of emotionalism and presents a framework/model, which can shape clinical practice.

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Introduction to Thesis Portfolio

This thesis portfolio presents two main papers: a systematic review investigating the neurophysiological, neuropsychological and psychological predictors and correlates of emotionalism across a range of neurological disorders (Chapter one) and an empirical paper exploring individuals lived experience of post-stroke emotionalism (PSE) over time (Chapter three). A bridging chapter outlines the association between the two main papers (Chapter two). An extended methodology chapter discusses ontology, epistemology, rationale for chosen qualitative methods and a review of alternative qualitative methods for the empirical paper (Chapter four). Additionally, an extended results section in relation to the empirical paper is presented, which helps to further explore the research questions (Chapter five). Finally, an overall discussion and critical review chapter summarises the thesis portfolio and discusses strengths and weaknesses and implications for future research and clinical practice (Chapter six).

This thesis portfolio aimed to explore the neurological, psychological and experiential factors of emotionalism in neurological disorders. Emotionalism is a condition characterised by episodes of uncontrollable crying or more rarely laughter, not under usual control and which are disproportionate or inappropriate to the social context (Ahmed & Simmons, 2013). Across the literature the terminology of emotionalism has varied such as pseudobulbar affect (PBA), involuntary crying or laughing, emotional lability and involuntary emotional expression disorder (IEED). Throughout this thesis portfolio the term emotionalism will be used and PSE, when referring specifically to emotionalism after stroke. Emotionalism has been found to occur in a range of neurological disorders including multiple sclerosis, amyotrophic lateral sclerosis (ALS), traumatic brain injury (TBI) and stroke (Schiffer & Pope, 2005).

The neurobiology of emotionalism is still relatively unknown. Emotionalism can occur as a consequence of both bilateral and unilateral strokes (Allman, 1991). It is believed to arise from lesions to the frontal lobes and descending corticobulbar-cerebellar circuits, which regulate motor control and the co-ordination of emotional expression (Engelman et al., 2014). Additionally, disruptions to pathways in frontal, parietal and brainstem regions may cause involuntary crying or laughter episodes (Colamonico et al., 2012).

Evidence of the effectiveness of pharmacological interventions is mixed. A Cochrane systematic review found antidepressants have been effective in reducing the frequency of crying episodes following stroke (Hackett et al., 2010). However, this review also found that antidepressants have only a small positive effect, which is not specific to one drug or one class of drugs. Additionally, they found that drug treatments have been ineffective in many cases. Importantly, the review concluded that the studies that have been conducted to date are of low to moderate quality and comprising only small-scale trials, whereas larger scale high quality research to provide reliable data is required to provide recommendations for treatment. The Cochrane review was recently updated and again supported the previous conclusions on intervention evidence for PSE. Moreover, it was suggested that longitudinal research is required to assess emotionalism over time, that could help to gain an understanding in terms of severity and factors which could aid remission (Allida et al., 2019).

An individual's appraisal about the causes or meaning of emotionalism may influence their approach to coping and recovery however, there is no psychological model to account for the diverse experience of individuals with emotionalism. According to an attribution model of illness, those who believe that illness chronicity and severity is due to psychological causes express higher levels of distress and poorer psychosocial outcomes

(King, 1983). The benefit of investigating patient interpretations of their illness is recognised in other health populations, though less so in stroke.

Additionally, bereavement models have been proposed as a way of understanding how stroke survivors adjust and respond psychologically. Wade et al. (1985) utilised a four-stage model of psychological adjustment to stroke developed by Holbrook (1982). The first stage of this model is categorised by crisis, which consists of high levels of anxiety and shock. The following stage is treatment whereby high expectations of recovery are established as well as periods of grieving. Realisation of the impact of the stroke in terms of disability is the third stage where individuals may experience feelings of frustration, anger and despair. The final stage is adjustment or acceptance of a new reality following their stroke. However, models such as these address general adaptions to life post-stroke but to date there is little in the literature regarding adaption to PSE specifically. Further research is required to explore stroke survivors experience of emotionalism longitudinally and how this may impact rehabilitation and coping with PSE.

Rationale for main papers

To date there is no systematic review which has explored neurophysiological, neuropsychological and psychological predictors and correlates of emotionalism across neurological disorders. Research has suggested emotionalism is common and a comprehensive investigation of predictors and correlates is required to enhance theoretical knowledge and clinicians' understanding of emotionalism. This systematic review could help provide further knowledge about emotionalism, which could shape clinical practice and contribute to the development of a model to guide medical and psychological assessment, management and treatment of emotionalism across neurological disorders.

Qualitative research can be beneficial to explore individuals' lived experience of a condition whereby rich data can be captured (Maxwell, 2012[a]). Only one previous

qualitative research study has been conducted exploring stroke survivors' lived experience of emotionalism (McAleese et al., 2019). However, this study had a small sample size with participants interviewed on average 4.3 months' post-stroke, so there was no consideration of the lived experience of PSE at the acute stage, and longitudinally, over time. Further larger scale longitudinal research is important to further explore experience, ways of coping and the impact of emotionalism. The empirical paper is the first to date to explore lived experience of emotionalism over time specifically following stroke, using a longitudinal qualitative approach. This research is clinically important, which could help develop a framework/model to shape psychological and behavioural interventions for this common disorder.

Chapter One: Systematic Review

Prepared for submission to Neuropsychological Rehabilitation

Author Guidelines available in Appendix A.

Predictors and correlates of emotionalism across acquired and progressive neurological conditions: A Systematic Review

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Abstract

Background: Emotionalism can develop following a range of neurological disorders however the aetiology of emotionalism is still unclear.

Objectives: To identify neurophysiological, neuropsychological and psychological predictors and correlates of emotionalism across neurological disorders: stroke, Parkinson's disease, multiple sclerosis, traumatic brain injury, Alzheimer's disease, vascular dementia and amyotrophic lateral sclerosis. To explore if these predictors and correlates of emotionalism differ across neurological disorders.

Methods: A comprehensive systematic search was completed of four databases: MEDLINE, CINAHL Complete, PsycINFO and EMBASE. Methodological quality was assessed using the Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies and each study graded according to the level of evidence using the Scottish Intercollegiate Guidelines Network.

Results: 50 papers (participants N= 1922) were included. 25 studies were rated as 'Fair', 21 studies rated as 'Good' and four studies rated as 'Poor'. The review identified predictors and correlates found in several neurological disorder such as bulbar networks, serotonergic pathways, genetics and female gender. Psychological predictors were only explored in participants who had experienced a stroke with a correlation between emotionalism and avoidant coping style reported in two studies.

Conclusions: Due to the disproportionate number of studies identified across neurological disorders it is difficult to draw definitive answers. Further research is required across neurological disorders to explore similarities and differences in neurophysiological, neuropsychological and psychological predictors and correlates.

Keywords: Emotionalism; predictor; correlate; systematic review; neurological disorders

Systematic Review Registration ID: CRD42020159413

Introduction

Emotionalism, also known as emotional incontinence, pseudobulbar affect (PBA), emotional lability, pathological laughing and crying or involuntary emotional expression disorder (IEED) is a condition which arises following a range of neurological disorders including multiple sclerosis, amyotrophic lateral sclerosis (ALS), traumatic brain injury (TBI) and stroke (Schiffer & Pope, 2005). The term emotionalism will be used for this review although there is not a universally agreed term within the literature. Emotionalism produces a lessening of the ability to control emotional expression (House et al., 1989). There is a consensus regarding the diagnostic criteria of emotionalism usually referred to as the House diagnostic criteria (House et al., 1989; Calvert et al., 1998; Eccles et al., 1999). According to this criteria emotionalism is characterised by (i) increased tearfulness or laughter, (ii) episodes occurring suddenly, with no warning, (iii) crying/laughter not under usual control and (iv) episodes occurring at least once a week. Crying episodes are more common with approximately 82% of individuals with emotionalism following a stroke experience crying episodes only and 2% experience laughing episodes only (Calvert et al., 1998).

Emotionalism may lead to negative consequences in terms of social and occupational functioning such as a reduction in work productivity or activities of daily living, potentially increasing the burden which already exists due to the primary neurological disorder (Colamonico et al., 2012). Individuals with emotionalism have higher Barthel Index scores (Choi et al., 2013) and a higher degree of disability (Choi-Kwon et al., 2012). Research has found emotionalism can lead to embarrassment, increased levels of distress and social withdrawal (Wortzel et al., 2008). Additionally, emotionalism may interfere with rehabilitation

and could cause a lack of willingness to engage with services (Allman, 1991; Sacco et al., 2008).

The prevalence of emotionalism varies considerably across the neurological disorders dependent on the criteria and terminology used. A systematic review and meta-analysis of 15 post-stroke emotionalism prevalence studies found 17% of stroke survivors suffer from PSE acutely, 20% at six months and 12% beyond six months (Gillespie et al., 2016). Research has found a prevalence rate for emotionalism in patients with multiple sclerosis to be between 10-46.2% (Vidović et al., 2015). Additionally, in a sample of patients with TBI the prevalence of emotionalism was between 5-11% and approximately 49% in a sample of patients with ALS (Zeilig et al., 1996; Parvizi et al., 2006).

Emotionalism has been found to be co-morbid with psychiatric disorders with research suggesting an increased likelihood for depression in individuals with emotionalism (Tang et al., 2004). Emotionalism is also under-recognised and can be mis-diagnosed for depression due to the co-occurrence of both disorders (Wortzel et al., 2008) and because of the tearful aspects central to both. An important difference is noted for depression whereby affect is proportionate and consistent with prolonged feelings of sadness and hopelessness. In contrast crying or laughing episodes associated with emotionalism are usually brief, subjectively uncontrollable and could be triggered by an emotional event rather than an individual's mood (Poeck, 1969; Cummings et al., 2006). Therefore, although emotionalism and mood disorders can be co-morbid, they are different clinical entities in terms of duration and context and require different treatment strategies (Colamonico et al., 2012).

Despite the high prevalence of emotionalism across neurological disorders, the aetiology of emotionalism and underlying mechanisms remains unclear. The release hypothesis proposes that emotionalism occurs as a result of disrupted cortical inhibition to the upper brainstem centre and release of the lower bulbar nuclei (Wilson, 1924). Other theories suggest disruptions of neurotransmitters such as serotonin or dopamine may lead to changes in emotional expression (Rabins & Arciniegas, 2007). More recently, a gate control theory proposes that damage to the corticobulbar/cerebellar pathways that regulate motor control and co-ordination of emotional expression or lesions in the frontal lobes may contribute to the development of emotionalism (Parvizi et al., 2009). Due to the limited understanding of the aetiology of emotionalism, a systematic review is required to explore predictors and correlates across neurological disorders, which could help to enhance theoretical understanding and shape clinical practice.

Multiple methods have been used to investigate the pathophysiology of emotionalism. Earlier theories or hypotheses of the pathophysiology of emotionalism were based on post-mortem studies (Bede & Finegan, 2018). More recent theories have deployed in vivo investigation methods including neuroimaging techniques, electrophysiological responses studying event-related potentials and exploration of neurochemistry (Floeter et al., 2014). The development of more modern technology has enabled further investigations of biological predictors and correlates of emotionalism and to validate previous theories or propose alternative hypotheses.

There is a lack of reviews specifically investigating the aetiology of emotionalism, with only a few published to date. A narrative review of emotionalism explored an overview of PSE in terms of epidemiology,

pathophysiology, clinical features and therapeutic options (Girotra et al., 2018). Additionally, a literature review of the epidemiology and pathophysiology of emotionalism was completed (King & Reiss, 2013). However, these reviews have only provided an overview and lacked a predefined protocol and not completed quality checks or assessment of bias, which highlights the methodological limitations of previous reviews meaning the results/conclusions may not be reliable or valid. Furthermore, these reviews have not explored emotionalism across neurological disorders to enable a greater understanding of this condition.

This systematic review is the first to investigate neurophysiological, neuropsychological and psychological predictors and correlates of emotionalism across neurological disorders: stroke, Parkinson's disease, multiple sclerosis, TBI, Alzheimer's disease, vascular dementia and ALS. This review is important to provide further knowledge, which could inform clinical practice and treatment whereby education could be provided to clients and families about emotionalism. Therefore, this review is clinically important to help contribute to the development of a model to guide medical and psychological assessment, prevention and management of emotionalism across neurological disorders.

Objectives

The systematic review aimed to explore the following questions:

1. What are the neurophysiological, neuropsychological and psychological predictors and correlates of emotionalism across neurological disorders: stroke, Parkinson's disease, multiple sclerosis, TBI, Alzheimer's disease, vascular dementia and ALS?

2. Do neurophysiological, neuropsychological and psychological predictors and correlates of emotionalism differ across neurological disorders?

Methods

Protocol and registration

The systematic review protocol was registered with PROSPERO: International prospective register of systematic reviews (Registration ID CRD42020159413, Appendix B) outlining rationale, aims, search strategy and data synthesis plans. The review conforms to the guidelines outlined by the Preferred Reporting Items for Systematic Reviews and Meta-Analyses statement (PRISMA; Moher et al., 2009).

Eligibility criteria

For this review the eligibility and inclusion criteria were outlined using the PICOS (Participants, Interventions, Comparisons, Outcomes and Study design) framework (Tacconelli, 2010). For this review 'comparisons' was not applicable due to the type of review questions. Due to the breath of study designs included in this review 'intervention' was extended to 'independent variable' to include interventions, predictors or correlates. Articles were included for review if they met the following eligibility criteria below.

Participants

Inclusion criteria

- Studies of emotionalism with adults (18 years or over) with a neurological disorder; stroke, Parkinson's disease, multiple sclerosis, TBI, Alzheimer's disease, vascular dementia and ALS. The seven neurological disorders

were chosen based on prevalence research which has shown that emotionalism is most commonly associated with these neurological disorders (Wortzel et al., 2008).

- No restrictions on time since onset of emotionalism.

Exclusion criteria

- Any neurological disorder not included in the inclusion criteria.

Independent variable – Intervention, predictors or correlates

For this review predictors were defined as variables used in regression analyses which provide information on an associated dependent variable regarding a particular outcome (Salkind, 2010) and correlates were defined as a measure of the strength of the relationship/association between two variables (Bobko, 2001).

Inclusion criteria

- Biological variables (neurophysiological, anatomical, neuropsychological)
- Psychological variables

Outcome

Inclusion criteria

- Measure of emotionalism such as standardised Kim's criteria (Kim & Choi-Kwon, 2000), House's criteria (House et al., 1989), Center for Neurological Study – Lability Scale (CNS-LS; Moore et al., 1997), Pathological Laughing and Crying Scale (PLACS; Robinson et al., 1993), interviews or self-report questionnaires.

Study Design and Publication Type

Inclusion criteria

- Quantitative studies
- Cross sectional studies

- Observational
- Cohort studies
- Case-control.

Exclusion criteria

- Qualitative studies
- Reviews
- Dissertations
- Unpublished 'grey' literature
- Studies not published in English language

Context

No limits in terms of context. Studies across different settings such as hospital, residential nursing home, supported living and independent living in the community were included.

Information sources

A comprehensive systematic search of MEDLINE, Cumulative Index to Nursing and Allied Health Literature (CINAHL Complete), PsycINFO and EMBASE databases were completed for this review.

Search Strategy

Boolean operators (OR, AND) were used to search each neurological disorder ('participant') with the search terms for emotionalism ('outcome') individually. For example, stroke OR "cerebr* accident" OR "cva" OR "apoplexy" AND emotionalism OR "emotional lability" OR "emotional dysregulation" OR "involuntary emotional expression disorder" OR "involuntary crying" OR "involuntary laughing" OR "lability of mood" OR "pathological laughing" OR "pathological crying" OR "pseudobulbar affect" OR "emotional incontinence" OR

"pathological display of affect" OR "inappropriate laughing" OR "inappropriate crying". It was decided not to include search terms for predictors and correlates ('independent variable') as this resulted in a limited number of results in the pilot search whereby studies could be unintentionally missed or this could increase bias where certain independent variables were selected.

Keywords and Medical Subject Headings (MeSH; Rogers, 1963) were also used when completing the search strategy for each neurological disorder such as "Stroke" [Mesh]. See Appendix C for the full search strategy for each database.

Once the searches were completed, title and abstracts were screened according to eligibility criteria. If a decision for eligibility was not able to be made at title and abstract screening stage due to insufficient information, the full article was reviewed. Following this the full texts of identified studies were further screened with reasons for exclusion noted. Reference lists of studies were hand-searched to check if any potential studies were not captured by the search strategy. A total of 25% of papers were checked by a second independent reviewer, a trainee clinical psychologist at title and abstract stage and at full text stage.

The final search was conducted on 12th February 2021, therefore only research published up to this point was included in the review.

Data Extraction

Once searches were completed relevant data were extracted from the full papers and summarised. A data extraction template was designed to include a descriptive summary of the studies included in the review (cf. Centre for Reviews and Dissemination, 2008). This included study characteristics; authors, year, country/setting, neurological disorder ('participant'), sample size and makeup, independent variables/predictors/correlates ('independent variable'), measures of

emotionalism used (‘outcome’), research design (‘study’), age range and study findings in relation to the review question, see Table 1 to 4.

Due to the significant heterogeneity in how emotionalism was measured and small sample sizes, a narrative synthesis was completed rather than a meta-analysis. The systematic review followed the narrative synthesis framework of Popay et al. (2006) to describe the neurophysiological, neuropsychological and psychological predictors and correlates of emotionalism across each neurological disorder. The narrative synthesis adopted a textual approach to summarise and explain the findings of the synthesis, explore relationships in the data and assess the robustness of the synthesis.

Assessment of Methodological Quality

The Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies (QATOCCS; National Heart, Lung and Blood Institute, NHLBI, 2014) was used to rate the methodological quality of the studies. The researcher was aware there were alternative tools available but the QATOCCS was chosen as this closely aligns with the type of study designs included in the review, the range of domains assessed in the checklist to assess the quality of the studies, the detailed guidance provided and the ability to calculate inter-rater reliability when using a second independent rater. This tool examined researcher bias, sample bias, sample size, time effects, accuracy and reliability of outcome measures, drop-out rates and if confounding variables were accounted for. The tool consists of 14 questions with each element rated using ‘yes’, ‘no’, ‘cannot determine’, ‘not reported’ or ‘not applicable’. Each study was summarised and critically appraised following the rating for each item to provide an overall rating of ‘good’, ‘fair’ or ‘poor’. For each study if less than seven items were rated yes this was classed as ‘poor’, seven

or above items rated yes this was classed as ‘fair’ and if 10 items were rated as yes or nine with additional reasons such as not applicable this was classed as ‘good’.

A random 25% of papers were independently reviewed by a second rater, a trainee clinical psychologist, to increase the rigour of the quality ratings. There was strong agreement between the two reviewers’ judgements, $\kappa = .83$, $p<0.001$. Any discrepancies between ratings were resolved through discussions and review of the QATOCCS guidance document leading to full agreement between reviewers.

No studies were excluded based on the quality rating, see Table 2.

Assessment of Risk of Bias

The Scottish Intercollegiate Guidelines Network (SIGN; Miller, 2002) was used to grade each study according to the level of evidence. The grades range from 1++ for high quality meta-analyses with very low risk of bias to 4 for expert opinion and formal consensus. This tool examines the quality of evidence whereby greater weight is given to studies which have controlled for biases or design limitations.

Table 1: Summary of study characteristics and data extraction for neurophysiological variables

Authors	Aim	Country / Setting	Neurological disorder/s	Total number of participants (N) and number in subgroups (n)	Mean age (SD)	IV - Predictors or correlates in relation to SR question	Measure of emotionalism	Study design	Findings in relation to SR question
Lebert et al. 1994	Examine the relationship between hemispheric asymmetry and DAT-specific emotionalism using SPECT	France - Outpatient	Alzheimer's disease	N = 34 Emotionalism n = 8 Non-emotionalism n = 26	71.4 ± 10.8	Hemispheric asymmetry	Semi-structured interview with participant and carer and carer questionnaire	Cross-sectional	Frontolateral asymmetry indices were significantly lower in emotionalism-positive group compared with emotionalism-negative group.
Liu et al. 2017	Examine the global spontaneous brain activity in individuals exhibiting PLC after stroke	China - Hospital	Stroke	N= 36 PLC n= 12 Non-PLC n = 12 Healthy control n = 12	57.42 ± 5.71	ALFF and ReHo	PLACS	Case-control	ALFF in the right anterior cingulate cortex, middle temporal gyrus, parahippocampal gyrus and bilateral medial prefrontal cortex was significantly greater and ALFF in left precentral gyrus and right superior frontal gyrus was significantly lower with patients with PLC. ReHo in the left inferior temporal gyrus, middle temporal gyrus and bilateral anterior cingulate cortex was significantly greater and ReHO in the left precentral gyrus, superior frontal gyrus/supplementary motor area and right inferior parietal lobule was significantly lower with patients with PLC. In patients with PLC ALFF in the right anterior cingulate cortex/medial prefrontal cortex and parahippocampal gyrus was significantly greater and the left cerebellum posterior lobe was significantly lower. Patients with PLC ReHo in the right anterior cingulate cortex/medial prefrontal cortex and inferior temporal gyrus was significantly greater and the left dorsolateral prefrontal cortex was significantly lower.

Kim et al. 2012	Investigate whether the 5-HTT and 5-HTR2a genes are associated with PSEI independently and/or interactively	Korea - Hospital	Stroke	N= 276 PSEI n = 37 Non-PSEI n = 239	64.8 ± 10.3	Polymorphisms of 5-HTT and serotonin 2a receptor (5-HTR2a) genes	Participants and caregivers asked specified questions using Kim's criteria	Cross-sectional	Patients with PSEI were more likely to have an anterior stroke location (borderline statistical significance). Patients with PSEI had a significantly higher frequency of the 5-HTTLPR 5 allele compared with those without PSEI. The association between 5-HTTLPR genotype and PSEI strengthened progressively with increasing number of 5 alleles and remained significant in patients with 5/5 genotype.
Haiman et al. 2008	To characterise the electrophysiological activity and the brain structures involved in response to subjectively significant and neutral auditory stimuli, to indicate whether PBA in MS patients is limited to the motor system or involves other cortical areas associated with emotional and sensory processing	Israel -MS Clinic	Multiple sclerosis	N= 33 Emotionalism n = 11 Non-emotionalism n = 11 Healthy controls n = 11	46.6 ± 9.6	Electrophysiological activity and brain structures	CNS-LS	Case-control	Significantly distinct activation in MS with emotionalism group in the vicinity of the somatosensory and motor arears in response to neutral stimuli and at pre-motor and supplementary motor arears in response to subjectively significant stimuli. Subjectively significant and neutral stimuli evoked higher current density in MS and emotionalism group.
Andersen et al. 1993	To investigate the effects of the selective serotonin reuptake inhibitor citalopram on uncontrolled crying	Denmark – Hospital	Stroke	N = 13	58.5	Citalopram	Semi-structured interview	Double-blind placebo-controlled	Number of daily crying episodes decreased by at least 50% in all cases during citalopram treatment compared with patients during placebo treatment. The effects were rapid and pronounced in 11 patients (73%).
Prokšelj et al. 2014	Determine platelet 5-HT concentration in AD patients with or without aggression or IEED	Slovenia	Alzheimer's disease	N = 49 IEED n = 16 Aggressive behaviour n = 14 Controls n = 19	79.3 ± 4.5	Platelet 5-HT concentrations	PLACS	Cross-sectional	Platelet 5-HT concentrations were significantly lower (2.9 times and 2.6 times) in patients with Alzheimer's Disease and co-existing IEED compared to control and patients with aggressive behaviours.

MacHale et al. 1998	Investigate the relation between lesion location and psychiatric illness after stroke	Edinburgh – Hospital	Stroke	N = 55 Emotionalism n = 26 Non-emotionalism n = 29	66	Lesion location	Short emotionalism questionnaire as defined in the Oxfordshire Community Stroke Project	Part of an RCT study	Patients with lesions in the right anterior region had a higher frequency of emotionalism at the time of psychiatric assessment when compared with any other region.
Haiman et al. 2009	To assess the effects of DM/Q by comparing the electrophysiological activity and the brain structures involved in MS patients with PBA before and after administration of DM/Q	Israel - MS Clinic	Multiple sclerosis	N = 12 PBA n = 6 Healthy controls n = 6	48.6 ± 9.5	Brain activity and cortical structures	CNS-LS	Case-control	Comparisons between PBA-Pre DM/Q treatment and post treatment indicated distinct activations in areas involved in emotional processing and high-level and associative visual processing in response to neutral stimuli. Distinct activations in areas involved in emotional processing in response to subjectively significant stimuli.
Tang et al. 2004	To examine the frequency of PSEI according to two sets of diagnostic criteria and to determine the clinical and radiological correlates of PSEI in a cohort of Chinese stroke survivors	Hong Kong – Hospital	Stroke	N = 84 PSEI n = 15 Non-PSEI n = 69	69	Radiological correlates	Psychiatric interviews based on Kim's criteria	Cross-sectional	PSEI was associated with a younger age, previous TIA, total National Institute of Health Stroke Scale and cortical infarcts. Cortical infarcts were independent predictors of PSEI.
Morris et al. 1993	To determine the frequency of emotional lability in this population and to identify factors associated with this condition	New Zealand – Hospital	Stroke	N = 66 EL n = 12 Non-EL n = 54	70 ± 11	Lesion location	Psychiatric assessment based on House's criteria	Cross-sectional	Marginally higher frequency of EL among patients with left hemisphere lesions than those with right hemisphere lesions. Frequency of EL increased from more posterior to more anterior brain regions, a significant association that was apparent in both hemispheres. Single lesions located in anterior regions of the cerebral hemispheres had four times the odds of EL than lesions located elsewhere.
Brown et al. 1998	To explore the effectiveness of fluoxetine in the treatment of post-stroke emotionalism	UK – Hospital	Stroke	N = 19 Emotionalism n = 9 Controls n = 10	61.4 ± 8.6	Fluoxetine	Semi-structured interview and modification of Lawson and MacLeod rating scale	Double-blind placebo-controlled	A clinically significant improvement in participants who received fluoxetine treatment compared to the placebo group.

Floeter et al. 2014	To determine the prevalence and characteristics of pseudobulbar affect in patients with primary lateral sclerosis and amyotrophic lateral sclerosis and to test the hypothesis that damage of inputs to the cerebellum, leading to cerebellar dysfunction is associated with PBA	America – Clinics	PLS and ALS	N = 47 PBA n = 22 Non-PBA n = 25	56.9 ± 7.1	Clinical variables and white matter tracts	Psychiatric interview with family	Cross-sectional	ALSFRS-R score and finger-tapping rates were significantly lower for patients with PBA suggesting more impaired motor function. Patients with PBA had increased mean diffusivity of white matter tracts underlying the frontotemporal cortex, the transverse pontine fibers and the middle cerebellar peduncle.
Choi et al. 2018	To examine the relationship between sex hormones and post-stroke emotional disturbances in patients with a history of stroke especially AP and EI and to investigate whether statins affect sex hormone levels or the presence of post-stroke AP/EI	Korea - Medical Centre	Stroke	N = 40 EI n = 16 Non-EI n = 24	61.6 ± 11.3	Hormone levels (testosterone)	Interviews with patients and caregivers	Prospective observational pilot	Total testosterone levels were significantly lower in the AP/EI group. Testosterone was independently associated with the presence of AP/EI.
Tang et al. 2009[a]	To assess the relationship between MBs and PSEI in stroke survivors	Hong Kong – Acute Stroke Unit	Stroke	N = 519 PSEL n = 74 Non-PSEL n = 445	65.6 ± 9.9	Number and location of microbleeds	Psychiatric interviews based on Kim's criteria	Case control	Patients with PSEL group were more likely to have microbleeds in the thalamus as a whole, its anterior and paramedian territories and a higher number of microbleeds in the entire brain. Multivariate analysis indicated microbleeds in the thalamus and MMSE were significant independent predictors of PSEL.
Ghaffar et al. 2008	To address the pathogenesis of PBA using quantitative MRI brain analyses in multiple sclerosis patients with and without PBA	Canada – Outpatient	Multiple sclerosis	N = 28 PBA n = 14 Non-PBA n = 14	46.6 ± 9.8	Lesion volume	Screened using Poeck's definition	Case-control	Those with PBA showed discrete differences in hyperintense lesion volume in five regions: right medial inferior frontal, right inferior parietal, left medial inferior frontal and left inferior parietal. Brainstem hypointense lesion volume was also significantly higher in PBA group. A Logistic regression model identified brainstem hypointense, left inferior parietal hyperintense and left and right medial inferior frontal hyperintense lesion volumes accounted for 70% of the variance when it came to explaining the presence of PBA.

Murai et al. 2003	To clarify whether differences in serotonin neurotransmission explain the differences between PC patients with unilateral cerebral lesions and non-PC patients with similar lesions	Germany - Stroke Unit	Stroke	N = 15 PC n = 6 Non-PC n = 9	60.2 ± 11	SERT densities	CNS-LS	Pilot	Midbrain/pons [I]β-CIT binding ratios of the PC group were significantly lower than those of the non-PC group.
Andersen et al. 1994	To correlate the severity of post-stroke PC with lesion size and location as demonstrated by MRI paying particular attention to brain areas involved in serotonergic neurotransmission	Denmark	Stroke	N = 12	51.5	Lesion location	Based on clinical judgement	Double-blind placebo-controlled trial	Those classed as clinically most severe pathological crying had relatively large bilateral pontine lesions without lesions in the hemispheres. Intermediate group had bilateral central hemispheric lesions. Clinically least affected group had mainly unilateral subcortical lesions.
Kim, 2002	Investigate the factors related to PSEI including detailed lesion location	Korea - Medical Centre	Stroke	N = 25 EI n = 13 Non-EI n = 12	58.5	Lesion location – lower (L) level and upper (U) level	Assessment	Prospective	At the L level the lesions of EI-present patients were more localised dorsally than those of PSEI-absent patients. The right sided lesions were significantly more frequent in the PSEI-present group than PSEI-absent group at this level. At the U level the difference between groups was not distinct. Lesions involving mainly the globus pallidus, dorsally located lesions were more often associated with PSEI than ventrally located ones.
Hübers et al. 2016	To explore the pathogenesis of PLC by exposing ALS patients to simultaneously presented visual and auditory stimuli, which were either emotionally congruent or incongruent	Germany - Neurology Department	ALS	N = 20 PBA n = 10 Controls n = 10	64 ± 12.2	Physiological parameters (heart rate, galvanic skin response, activity of facial muscles)	CNS-LS	Case-control	Group-differences in electrophysiological data were explained by frontal cortex functioning as expressed by the ECAS score: EMG activity of the orbicularis ois muscle, the orbicularis oculi muscle, a non-significant trend for heart rate and GST between the two musical conditions.

Christidi et al. 2018	To examine the neuroanatomical substrate of PLC in ALS without dementia by simultaneously evaluating grey matter and white matter changes in ALS patients with and without PLC	Greece	ALS	N = 81 PLC n = 56 Healthy controls n = 25	60.43 ± 10.06	Grey matter and white matter	CNS-LS	Case-control	ALS-PLC patients showed decreased grey matter volume in left orbitofrontal cortex, frontal operculum and putamen and bilateral frontal poles compared to ALS non-PLC. ALS-PLC patients had decreased fractional anisotropy in left cingulum bundle and posterior corona radiata. White matter abnormalities were additionally detected in white matter associative and ponto-cerebellar tracts.
Burns et al. 1999	To assess whether a selective serotonin reuptake inhibitor is effective in treatment of stroke-associated lability of mood	UK – hospital	Stroke	N = 28	73.4 ± 9.1	Sertraline	Interviewer rated participants using House criteria	Double-blind placebo-controlled trial	Statistically significant improvements in lability score, tearfulness significantly diminished after 2 weeks and remained significantly lower throughout the trial. Independent association for the improvement in tearfulness with length of time since stroke and score on the Frenchay aphasia battery.
Luhoway et al. 2019	To explore the relationship between posterior fossa lesions and PLC in people with MS	England and Canada – MS Clinics	Multiple sclerosis	N = 77 PLC n = 22 Non-PLC n = 55	39.3 ± 11	Posterior fossa lesions	CNS-LS	Retrospective	A significant inverse relationship was identified for patients with MS and without evidence of depression such that fewer posterior fossa lesions on automated magnetic resonance imaging was associated with the presence of PLC.
Kim & Choi-Kwon, 2000	To correlate the location of stroke with post-stroke depression and emotional incontinence	Korea – Outpatient	Stroke	N = 148 PSEI n = 50 Non-PSEI n = 98	62 ± 9	Lesion location, demographic variables and characteristics	Assessment with patients and relatives	Prospective cohort	Female gender, ischemic stroke and severe motor dysfunction were found to be related to PSEI. Anterior cortical stroke was more associated with PSEI than was the posterior cortical lesion.
Lopez et al. 2001	To examine the hypothesis that depression, emotional lability and apathy in AD patients are associated with cortical-subcortical dysfunction	America – Research Centre	Alzheimer's disease	N = 8 EL n= 1 Controls n = 7	84	Cortical-subcortical dysfunction – (rel-CBF)	Psychiatric assessment	Part of a longitudinal study	Patient with EL showed decreased re-CBF in the anterior cingulate and dorsolateral prefrontal cortices, bilaterally, and in the left basal ganglia. Showed increased rel-CBF in the right middle temporal area.

Tateno et al. 2004	To examine the clinical correlates and course of PLC following TBI	America - Hospital and Clinics	TBI	N = 103 PLC n = 34 Non-PLC n = 69	30.4 ± 11.5	Neuropsychiatric assessment variables and severity and localisation of brain injury	PLACS	Longitudinal cohort	Patients with PLC had a greater frequency of frontal lobe injury than patients without PLC. Significant difference between PLC group and non-PLC group in frequency of diffuse lesions. At three months after TBI there was a significant difference in the frequency of frontal lobe lesions between patients with and without PLC. Logistic regression analysis showed lateral aspect of left frontal lobe was associated with the presence of PLC.
Ko et al. 2017	To investigate whether TPH2 gene polymorphisms were associated with PSED	Korea - Medical Centre	Stroke	N = 383 PSEI n = 41 Non-PSEI n = 342	60 ± 10	TPH2 genes, NIHSS and lesion location	Interview based on Kim's criteria	Secondary analysis	PSEI was associated with high NIHSS score at admission and severe mRS score at 3 months. TPH2 SNP rs4641528 differed significantly between patients with and without PSEI. TT homozygote of rs4641528 was less common in patients with PSEI. PSEI was associated with NIHSS score at admission and with TPH2 rs4641528 allele carriers.
Starkstein et al. 1995	To examine the prevalence and correlates of pathological affect	Argentina	Alzheimer's disease	N = 103 Pathological crying n = 26 Pathological mixed n = 14 Non-PBA n = 63	72.5 ± 7.1	Neuroradiological variables and neurological findings	PLACS	Cross-sectional	Patients with mixed pathological affect had a significantly larger left lateral ventricle than patients with either pathological affect crying or no pathological affect. Patients with pathological laughing or crying had a significantly longer duration of illness and significantly higher anosognosia scores.

Table 2: Summary of study characteristics and data extraction for neuropsychological variables

Authors	Aim	Country / Setting	Neurological disorder/s	Total number of participants (N) and number in subgroups (n)	Mean age (SD)	IV - Predictors or correlates in relation to SR question	Measure of emotionalism	Study design	Findings in relation to SR question
Hanna et al. 2016	Determine the association between PLC and CI in an MS cohort	Canada - Cognitive Clinic	Multiple sclerosis	N = 153 PLC n = 58 Non-PLC n = 95	45.6 ± 8.1	Characteristics and MACFIMS performance variables	CNS-LS	Retrospective cohort	CNS-LS was found to be significantly correlated with years of education. CNS-LS scores were negatively correlated with performance on the COWAT, BVMTR immediate recall, BVMTR delayed recall, PASAT, DKEFS card sort, DKEFT card sort description and Stroop score. Comparing non-PLC group to the PLC group a difference was identified on the COWAT, CVLT2-IR, CVLT2-DR.
McCullagh et al. 1999	Explore a possible role for the prefrontal cortex in the syndrome of PLC using novel neuropsychological measures to probe its functional integrity	Canada – Clinic	ALS	N = 28 PLC n = 10 Non-PLC n = 8 Healthy controls n = 10	63.5 ± 6.7	WCST and novel 'Gambling task' psychometric variables	Poeck's criteria	Case-control	PLC group made significantly more total errors on the WCST than either the non-PLC or healthy controls group. A trend in the same direction was found for perseverative errors. WSCT total errors predicted the presence or absence of PLC with 75% accuracy.
Feinstein et al. 1999	To explore a putative role for the prefrontal cortex in the pathogenesis of PLC	Canada – Outpatient	Multiple sclerosis	N = 24 PLC n = 11 Non-PLC n = 13	43.7 ± 8.3	Cognitive indices	PLACS	Case-control	PLC group generated significantly less words on the COWAT while also taking significantly longer to perform the Stroop test. A trend was found for PLC patients to make more (total) errors on the WCST.
Feinstein et al. 1997	To define associated neurological, emotional and cognitive correlates of PLC	Canada – Outpatient	Multiple sclerosis	N = 24 PLC n = 11 Controls n = 13	43.7 ± 8.3	WAIS subtests	PLACS	Case-control	Patients with PLC had lower performance and full-scale but not verbal, IQ scores on the WAIS-R. PLC group were more impaired on a single verbal subscale, namely Arithmetic, and on two of the performance tasks, namely Digit Symbol and Picture Arrangement. Significant differences were identified on the Digit Backwards scores.
Patel et al. 2018	To describe the neuropsychiatric correlates of self-reported PBA symptom severity	America – tertiary care Centre	Parkinson's disease, ALS and aPD	N = 108 PBA n = 31 Non-PBA n = 77	PD 63.39 aPD 68.08 ALS 62.01	Characteristics and MoCA scores	CNS-LS	Cross-sectional	PBA patients were significantly younger than PBA negative patients in the aPD and ALS groups. CNS-LS score was significantly related to younger age and education and corrected lower MoCA scores for participants in the aPD group but not for the other clinical groups.

Fitzgerald et al. 2018	To determine the prevalence of pseudobulbar affect and assess its association with disability and symptom severity	America – MS Registry	Multiple sclerosis	N = 8136 PBA n = 133 Non-PBA n = 8003	56.9	MS symptoms, cognitive impairment and characteristics	CNS-LS	Retrospective Cohort	PBA group tended to be younger, non-white and have less education and income compared with non-PBA group. Those with PBA had on average greater disease severity across a range of symptoms. PBA was associated with increased odds of moderate vs mild self-reported cognitive impairment in multivariate models. Multivariate models adjusted for depression severity identified PBA was associated with increased odds of severe vs mild self-reported impairment in cognition, fatigue, hand function, vision, sensory and spasticity domains.

Table 3: Summary of study characteristics and data extraction for psychological variables

Authors	Aim	Country / Setting	Neurological disorder/s	Total number of participants (N) and number in subgroups (n)	Mean age (SD)	IV - Predictors or correlates in relation to SR question	Measure of emotionalism	Study design	Findings in relation to SR question
Eccles et al. 1999	To determine whether patients with emotionalism differed from patients without emotionalism in their psychological reactions to stroke or in the coping strategies reported	UK - Hospital	Stroke	N = 65 Emotionalism n = 19 Non-emotionalism n = 46	71.8	IES and MASS items	Interview	Cross-sectional	Association between emotionalism and the impact of events subscales intrusion and avoidance. Association between emotionalism and the mental adjustment to stroke scale subscales helplessness/hopelessness and anxious preoccupation.
Calvert et al. 1998	To identify psychological symptoms, other than those that define emotionalism which are associated with the condition	UK	Stroke	N = 448 Emotionalism n = 101 Non-emotionalism n = 347	68.5	Psychological variables	Standardised set of questions	Interview data from an RCT study	Multivariate logistic regression identified irritability and ideas of reference were associated with emotionalism.

Table 4: Summary of study characteristics and data extraction for mixed variables

Authors	Aim	Country / Setting	Neurological disorder/s	Total number of participants (N) and number in subgroups (n)	Mean age (SD)	IV - Predictors or correlates in relation to SR question	Measure of emotionalism	Study design	Findings in relation to SR question
Wei et al. 2016	Evaluate PSD and PSEI at different stages to correlate their symptoms with lesion location, coping styles and other variables	China – Hospital	Stroke	N = 378 PSEI n = 40 Non-PSEI n = 328	61.3 ± 9.4	Lesion location, coping styles and characteristics	Interview based on Kim's criteria	Longitudinal cohort study	Patients with both motor and sensory dysfunctions were more susceptible to PSEI at admission compared to individuals with pure motor or sensory dysfunction. Avoidance, acceptance-resignation and low objective support were predisposing factors for PSEI. Anterior cortex, pons and midbrain infarction, bilateral lesion location, severe white matter change, avoidance and acceptance-resignation were all significant risk factors associated with PSEI 3 months later. Multivariate analysis indicated PSEI at admission was associated with acceptance-resignation, whereas it was related to anterior cortex infarction and acceptance-resignation 3 months later.
Choi-Kwon et al. 2012	To investigate the characteristics and prevalence of post-stroke depression and post-stroke emotional incontinence and the factors related to these conditions at admission and 3 months after stroke	Korea - Medical Centre	Stroke	N = 508 PEI n = 48 Non-PEI n = 460	63.2 ± 10.3	5-HTTLPR, number of tandem repeats within intron 2 (Stin2VNTR), social support and lesion location	Interview based on set criteria	Longitudinal cohort	At admission lesion location (basal ganglia, corona radiata and internal capsule), presence of microbleeds and NIHSS score were related to PSEI. Lesion location, motor dysfunction at admission, mRS score and low social support were related to PSEI 3 months after stroke. Significant difference in the genotype frequencies of Stin2 VNTR polymorphism at 3 months after stroke between patients with and without PSEI. Stin2 10/10 and 12/10 genotypes more common among patients with PSEI. Multivariate logistic regression indicated lesion location (pons and midbrain) was the only independent factor associated with PSEI at admission whereas mRS score, Stin2 VNTR and low social support were independently associated with PSEI at 3 months after stroke.

Choi et al. 2013	To investigate the association of post-stroke emotional incontinence with various psychiatric symptoms and quality of life independent of potential covariates	Korea – Hospital	Stroke	N = 432 PSEI n = 51 Non-PSEI n = 381	64.6 ± 10	Sociodemographic and clinical variables	Patients and caregivers interviewed based on Kim's criteria	Secondary analysis	PSEI group were significantly more likely to have a previous history of stroke, higher NIHSS and lower BI scores.
Wang et al. 2016	To study the clinical features of and to identify the factors associated with PSPLC, to correlate PSPLC with lesion location and to analyse the difference between patients with and without pseudobulbar signs	China – Hospital	Stroke	N = 112 PSPLC n = 56 Non-PSPLC n = 56	62.4 ± 6.8	Characteristics, cognitive impairment, lesion location	PLACS	Retrospective case-control	Lesion located at the pons was related to PSPLC. PSPLC was independently related to pontine lesion. PSPLC group had a higher frequency of bilateral lesions. PSPLC was significantly related to Mild Cognitive Impairment. Significantly more patients had at least two strokes in the last two years in the group with PSPLC with pseudobulbar signs. Percentage of patients with severe neurological deficits at the onset of PLC was higher in the group with pseudobulbar signs. Those with PLC and pseudobulbar signs showed lesions with both bilateral basal ganglia plus bilateral subcortical white matter than those without pseudobulbar signs.
Tang et al. 2009[b]	To assess the relationship between executive functions, PSEI, and frontal and basal ganglia infarcts in stroke survivors	China - Research Clinic	Stroke	N = 78 PSEI n = 39 Controls n = 39	63.8 ± 8.7	MRI variables, executive function, location of acute infarcts	Psychiatric interviews based on Kim's criteria	Case-control	PSEI group had significantly more infarcts in the frontal and/or basal ganglia. There was a trend whereby the involvement of the middle cerebral artery territory was more frequent in PSEI group. PSEI group had significantly lower Chinese Frontal Assessment Battery scores. A significant correlation between frontal infarct and severity of PSEI (.420).
Vidović et al. 2015	To determine the prevalence of pseudobulbar affect in patients with MS and to analyse the link between PBA and patient age, sex, clinical course of MS, disease duration and degree of disability	Croatia – Inpatient	Multiple sclerosis	N = 79 PBA n = 33 Non-PBA n = 46	48.7 ± 10.96	Age, sex, clinical course of MS, disease duration and degree of disability	CNS-LS	Cross-sectional	Those with PBA showed a significantly higher female predominance.

Petracca et al. 2009	To examine the frequency and clinical correlates of IEED in Parkinson's disease	Argentina - Hospital	Parkinson's disease	N = 131 IEED n = 22 Non-IEED n = 109	63.8 ± 9.6	Parkinson's disease-related variables	PLACS	Cross-sectional	Patients with IEED had greater severity of Parkinson's disease compared with non-IEED patients; higher unified Parkinson's Disease Rating Scale salivation, axial rigidity, bradykinesia and gait disturbance scores.
Siddiqui et al. 2009	Examine whether PBA was associated with mood disturbances, motor disability, disease stage and quality of life	America - Movement Disorder Centre	Parkinson's disease	N = 719 PBA n = 37 Non-PBA n = 682	64.8 ± 12.2	Diagnosis, medication, VAMS score, severity of Parkinson's disease	Modified University of Florida Pseudobulbar affect screening questionnaire	Cross-sectional	Increased prevalence of PBA as PD/Parkinsonism patients approached higher levels of disability - stage 2.
Phuong et al. 2009	To examine frequency and correlates of IEED in PD using both the CNS-LS and new IEED diagnostic criteria, examine the overlap between IEED and depression, determine the discriminant validity of the CNS-LS in PD against a diagnostic interview applying the new IEED diagnostic criteria	America - Clinical and Medical Centres	Parkinson's disease	N = 193 IEED n = 100 Non-IEED n = 93	65.8 ± 10.7	Clinical variables and GDS-15	CNS-LS	Cross-sectional	Univariate analysis found female sex was associated with increasing CNS-LS score.
Thakore & Pioro, 2017	To examine the prevalence, associations and course of PBA in ALS, explore associations if any, that differentiate laughter from crying in PBA and examine the relationship of PBA and depression	America - Clinical Centre	ALS	N = 735 PBA n = 209 Non-PBA n = 526	60.8	Population characteristics and ALS related variables	CNS-LS	Exploratory observational study	PBA was significantly associated with female gender, bulbar onset, lower ALSFRS-R score and more rapidly progressive disease. Female gender, lower bulbar and gross motor ALSFRS-R sub-scores, lower age and shorter disease duration significantly increased odds of PBA. Model has an acceptable goodness of fit and predicts the presence of PBA with 74%.
Andersen et al. 1995	To explore possible relationship between post-stroke PC and depression	Denmark - Hospital	Stroke	N = 211 PC 1-month n = 24 PC 6 months n = 16 PC 1-year n = 33	69	Lesion site, lesion size, intellectual impairment, BI and Motricity Index	Interview	Longitudinal cohort	Patients with PC were significantly more physically impaired (BI and Motricity Index). PC was correlated to intellectual impairment at 6- and 12-months post stroke but not at 1 month. Lesion size were significantly larger in patients with PC.

Foley et al. 2015	To estimate the prevalence of PBA and examine the relationship between PBA symptoms and other clinical correlates	America - Nursing Home	Alzheimer's disease, stroke, Parkinson's disease, multiple sclerosis	N = 804 PBA n = 72 Non-PBA n = 732	79.6 ± 12.6	Demographic characteristics	CNS-LS	Retrospective observational	Patients with PBA symptoms were significantly more likely to be female compared to those without PBA.
Tortelli et al. 2016	To investigate using both a self-reported questionnaire and clinical examination the prevalence of pseudobulbar affect and define the ALS clinical phenotype associated with PBA at onset	Italy - Population based registry	ALS	N = 132 PBA n = 45 Non-PBA n = 87	62	Clinical phenotypes	CNS-LS	Prospective cross sectional	PBA group was characterised by shorter disease duration from symptom onset, onset-diagnosis interval (ODI) and lower ALSFRS-R bulbar sub-score. In patients with MS for less than two years pathological CNS-LS was associated with a shorter ODI and lower ALSFRS-R bulbar sub-score.
House et al. 1989	To estimate the prevalence of emotionalism after stroke, to assess its relation with other mood disorders, and to identify clinical variables with which its associated	England – Stroke Register	Stroke	N = 112 One-month n = 13 Six months n = 25 12 months n = 12	NR	Intellectual function and lesion location	Psychiatric assessment based on standardised questions	Longitudinal cohort study	Patients with emotionalism at one and six months had more intellectual impairments. At one month a significant trend for patient with emotionalism to show larger lesions. Lesions in the left frontal and temporal regions were associated with emotionalism at six months. Significant association at 12 months between emotionalism and anterior lesion locations. Patients with left anterior lesions compared with those with visible lesions in other parts of the brain showed a significant association with emotionalism at one, six and 12 months.
McGrath, 2000	To identify possible causal factors of emotionalism	England - Inpatient	TBI	N = 82 Emotionalism n = 43 Non-emotionalism n = 39	46.76	Lesion location and psychological variables	Structured interview	Retrospective design	Significant correlations were obtained between emotionalism-tearfulness ratings and ratings for other interview items: sadness, frustration, fear and worry. Independent variables which predicted crying behaviour were female gender and focal damage to the right cerebral hemisphere.

[123I]2 β -carbomethoxy-3 β -(4-iodophenyl)tropane ([I]β-CIT); 5-hydroxytryptamine (**5-HT**); ALS Functional Rating Scale-Revised (**ALSFRS-R**); Amplitude of low frequency fluctuation (**ALFF**); Amyotrophic Lateral Sclerosis (**ALS**); Anger Proneness (**AP**); Atypical Parkinson Disease (**aPD**); Barthel Index (**BI**); Center for Neurological Study - Lability Scale (**CNS-LS**); Controlled Oral Word Association Test (**COWAT**); Dementia of Alzheimer's type (**DAT**); Dextromethorphan/Quinidine (**DM/Q**); Emotional Lability (**EL**); Electromyography (**EMG**); Geriatric Depression Scale (**GDS-15**); Impact of Events Subscales (**IES**); Involuntary Emotional Expression Disorder (**IEED**); Pathological Laughing and Crying (**PLC**); Pathological Laughter and Crying Scale (**PLACS**); Post-stroke Emotional Dysfunction (**PSED**); Post-stroke Emotional Incontinence (**PSEI**); Post-stroke

Emotionalism Laughing (**PSEL**); Post-stroke Pathological Laughing and Crying (**PSPLC**); Primary Lateral Sclerosis (**PLS**); Pseudobulbar affect (**PBA**); Mental Adjustment to Stroke Scale (**MASS**); Minimal Assessment of Cognitive Function in MS (**MACFIMS**); Montreal Cognitive Assessment (**MoCA**); Mini-Mental State Examination (**MMSE**); Multiple Sclerosis (**MS**); National Institutes of Health Stroke Scale (**NIHSS**); Polymorphism of the Serotonin Transporter Protein (**STin2VNTR**); Regional homogeneity (**ReHo**); relative regional Cerebral Blood Flow (**rel-CBF**); Serotonin Transporter (**5-HTT**) Serotonin transporter (**SERT**); Serotonin transporter protein (**5-HTTLPR**); Traumatic Brain Injury (**TBI**); Tryptophan hydroxylase 2 (**TPH2**); Visual Analogue Mood Scales (**VAMS**); Wechsler Adult Intelligence Scale (**WAIS**); Wisconsin Card Sort Test (**WCST**)

Results

Study selection

Initial searches of the databases generated 3,238 studies with a total of 1,342 studies once duplicates were removed. The titles and abstracts of the studies from the search results were reviewed for eligibility and studies were excluded if they did not meet criteria (1203 studies). A total of 139 studies were reviewed at full text stage and reasons for excluding studies were recorded. Following eligibility checking a total of 50 studies were regarded as eligible for the review. See Figure 1 for PRISMA flow chart displaying the process of identifying final selection of studies to be included.

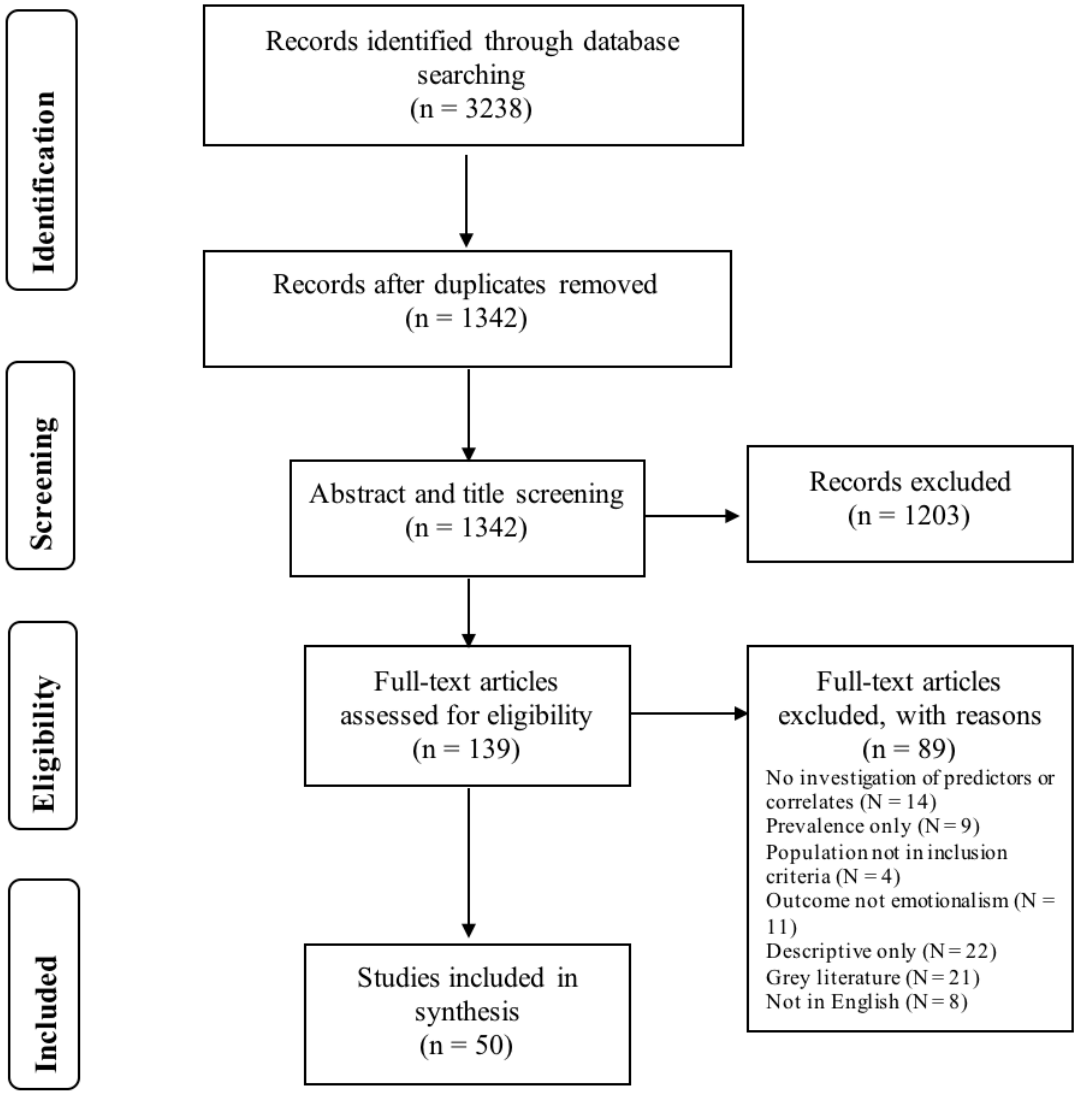

Figure 1: Flowchart displaying the process of identifying studies for inclusion in the review

Study Characteristics

The characteristics of the studies were extracted and have been outlined according to the PICOS criteria below.

Participants

A total of 1922 participants with emotionalism were included in the studies across all the papers. Of these participants 48% had a diagnosis of stroke, 12% ALS, 18% multiple sclerosis, 4% TBI, 6% Parkinson's disease, 8% Alzheimer's disease and 4% mixed with no breakdown of diagnoses. No studies were identified as appropriate that included participants with vascular dementia. Across the 50 studies the mean age of participants with emotionalism was 63.87 years.

The majority of studies had small sample sizes whereby the largest sample had 209 unique participants (Thakore & Pioro, 2017) and the smallest sample had one participant (Lopez et al., 2001) with emotionalism. The mean sample size was 38 participants.

Studies were conducted across 17 different countries. The largest number of studies were conducted in Europe (N=18) followed by 15 studies in Asia. Nine studies were completed in America, five in Canada, two in South America and one in New Zealand.

Predictors and correlates

Participant/disorder characteristics, neurophysiological, neuropsychological and psychological factors were investigated as possible predictors and correlates of emotionalism across neurological disorders. Neurophysiological factors were the most commonly explored across studies and included lesion location, number of lesions, lesion size, white matter changes and alleles/genes. Neuropsychological

factors were only investigated in stroke, multiple sclerosis and ALS. Additionally, only five studies explored psychological factors in a sample of stroke participants.

Outcome

Emotionalism was measured using a range of methods. The majority of studies (N=14) used the Center for Neurological Study – Lability Scale (CNS-LS; Moore et al., 1997). This is a self-report questionnaire, comprising seven questions across two subscales of laughter and labile tearfulness. The Pathological Laughing and Crying Scale (PLACS; Robinson et al., 1993) was used by eight studies. This is an interviewer-rated instrument, which consists of 16 items which are scored from zero (rarely or not at all) to three (frequently).

Eleven studies completed assessments for emotionalism using a psychiatric interview basing on Kim's criteria (N=7; Kim & Choi-Kwon, 2000; Appendix E). Three conducted a psychiatric assessment using pre-defined criteria (House et al., 1989; Lopez et al., 2001; Choi-Kwon et al., 2012) and two studies (Morris et al., 1993; Burns et al., 1999) used House's criteria (House et al., 1989; Appendix F).

Of the remaining studies, two studies (McCullagh et al., 1999; Ghaffar et al., 2008) screened patients using Poeck's criteria (Poeck, 1969), one study used the modified University of Florida PBA Screening Questionnaire (Siddiqui et al., 2009), one study used a short emotionalism questionnaire (MacHale et al., 1998) and one study assessed emotionalism based on clinical judgement (Andersen et al., 1994).

Design

The majority of included studies had cross-sectional designs (N=13) or case-control designs (N=11). Two included studies were part of an RCT (MacHale et al., 1998; Calvert et al., 1998) and four were from a double-blind placebo-controlled trial (Andersen et al., 1993; Andersen et al., 1994; Brown et al., 1998; Burns et al., 1999).

A longitudinal cohort design was implemented by five studies (House et al., 1989; Andersen et al., 1995; Tateno et al., 2004; Choi-Kwon et al., 2012; Wei et al., 2016) and one study was part of a longitudinal study (Lopez et al., 2001). Six studies were retrospective (McGrath, 2000; Foley et al., 2015; Hanna et al., 2016; Wang et al., 2016; Fitzgerald et al., 2018; Luhoway et al., 2019), three were prospective (Kim & Choi-Kwon, 2000; Kim, 2002; Choi et al., 2018).

Risk of bias within studies

Quality Assessment

The QATOCCS was completed for studies which were observational, cohort and cross-sectional in design, see Table 5. The majority of studies (N=25) were rated as 'Fair', 21 studies were rated as 'Good' and four studies were rated as 'Poor' (Andersen et al., 1994; Feinstein et al., 1999; Hübers et al., 2016; Liu et al., 2017). All the studies had clearly stated the research question and all but one study (Thakore & Pioro, 2017) had defined the study population sample. Additionally, all studies had clearly defined the exposure (predictors and correlates) and outcome (emotionalism) measures with the exception of one study where it was not possible to determine if the outcome measure was clearly defined (Andersen et al., 1994).

All the studies included in this review had elements of risk of bias and no studies were excluded from this review based on the quality assessments.

Level of evidence

The Scottish Intercollegiate Guidelines Network grading system (SIGN; Miller, 2002) was used to examine the level of evidence for each study. When grading the level of evidence, the ratings from the QATOCCS were considered. For this review the level of evidence ranged from '2++' for high quality case-control or cohort studies with a very low risk of confounders or bias, '2+' for well conducted

case-control or cohort studies with a low risk of confounders or bias or ‘2-’ for case-control or cohort studies with a high risk of confounders or risk. Only one study was classified as ‘3’ for non-analytic studies including case reports. Overall, the majority of the studies fell in the ‘2++’ and ‘2+’ level of evidence.

Table 5: Quality Assessment Ratings using the QATOOCS

Criteria	Lebert et al. 1994	Liu et al. 2017	Hanna et al. 2016	Kim et al. 2012	Haiman et al. 2008	Andersen et al. 1993	Wang et al. 2016	Prokšelj et al. 2014	MacHale et al. 1998	Haiman et al. 2009	Tang et al. 2009[a]	Tang et al. 2004
1. Clear research question?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
2. Study population defined?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
3. Participation rate at least 50%?	CD	CD	CD	CD	CD	CD	CD	CD	Yes	CD	CD	Yes
4. Inclusion and exclusion criteria prespecified?	Yes	CD	Yes	Yes	Yes	CD	Yes	NR	Yes	Yes	Yes	Yes
5. Sample size justification?	No	No	No	No	No	No	No	No	No	No	No	No
6. Exposure(s) of interest measured prior to the outcome?	No	No	Yes	No	No	Yes	No	No	Yes	Yes	Yes	Yes
7. Timeframe between measures sufficient?	Yes	Yes	Yes	Yes	Yes	CD	Yes	Yes	Yes	Yes	Yes	Yes
8. Different levels of the exposure?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
9. Exposure measures clearly defined?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
10. Exposure(s) assessed more than once?	No	No	No	No	No	Yes	No	No	No	No	No	No
11. Outcome measures clearly defined?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
12. Outcome assessors blinded?	No	No	No	No	No	Yes	No	Yes	Some	No	Yes	Some
13. Follow-up loss under 20%?	NA	NA	NA	NA	NA	Yes	NA	NA	NA	NA	NA	NA
14. Measurement of confounding variables?	NR	NR	Yes	NR	NR	NR	NR	Yes	Yes	Yes	NR	NR
Quality Rating	**Fair**	**Poor**	**Good**	**Fair**	**Fair**	**Good**	**Fair**	**Fair**	**Good**	**Good**	**Good**	**Good**

Criteria	Morris et al. 1993	House et al. 1989	Wei et al. 2016	Brown et al. 1998	Petracca et al. 2009	Floeter et al. 2014	Siddiqui et al. 2009	Christidi et al. 2018	Phuong et al. 2009	Thakore & Pioro, 2017	Choi et al. 2018	Tang et al. 2009[b]	Ghaffar et al. 2008
1. Clear research question?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
2. Study population defined?	Yes	Yes - details in previous study	Yes	Yes	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes
3. Participation rate at least 50%?	Yes	Yes	CD	CD	CD	CD	Yes	CD	Yes	Yes	Yes	CD	NR
4. Inclusion and exclusion criteria prespecified?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
5. Sample size justification?	No	No	No	No	No	No	No	No	No	No	No	No	No
6. Exposure(s) of interest measured prior to the outcome?	Yes	Yes	Yes	Yes	Yes	Some	No	Yes	No	No	No	Yes	Yes
7. Timeframe between measures sufficient?	Yes	Yes	CD	CD	Yes	Yes	No	Yes	No	CD	CD	Yes	Yes
8. Different levels of the exposure?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	NA	Yes	Yes
9. Exposure measures clearly defined?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
10. Exposure(s) assessed more than once?	No	Yes	Yes	Yes	No	No	No	No	No	No	No	No	No
11. Outcome measures clearly defined?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
12. Outcome assessors blinded?	Some	No	No	Yes	Yes	Yes	No	No	Yes	No	No	Yes	No
13. Follow-up loss under 20%?	NA	Yes	Yes	Yes	NA	NA	NA	NA	NA	NA	NA	NA	NA
14. Measurement of confounding variables?	Yes	NR	NR	NR	NR	NR	Yes	Yes	NR	Yes	Yes	NR	NR
Quality Rating	**Good**	**Good**	**Good**	**Good**	**Good**	**Good**	**Fair**	**Good**	**Fair**	**Fair**	**Fair**	**Good**	**Fair**

Criteria	Murai, et al. 2003	Andersen et al. 1994	Tateno et al. 2004	Hübers et al. 2016	McCullagh et al. 1999	Feinstein et al. 1999	Kim, 2002	Anders et al. 1995	Luhoway et al. 2019	Kim & Choi-Kwon, 2000	Choi-Kwon et al. 2012	Ko et al. 2017
1. Clear research question?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
2. Study population defined?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
3. Participation rate at least 50%?	NR	NR	CD	CD	CD	CD	CD	CD	CD	CD	No	Yes
4. Inclusion and exclusion criteria prespecified?	Yes	CD	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
5. Sample size justification?	No	No	No	No	No	No	No	No	No	No	No	No
6. Exposure(s) of interest measured prior to the outcome?	Yes	Yes	Some	No	No	No	Yes	Yes	CD	Yes	Yes	Yes
7. Timeframe between measures sufficient?	CD	CD	CD	CD	CD	CD	Yes	Yes	CD	Yes	Yes	CD
8. Different levels of the exposure?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
9. Exposure measures clearly defined?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
10. Exposure(s) assessed more than once?	No	No	Yes	No	No	No	No	Yes	No	No	Yes	No
11. Outcome measures clearly defined?	Yes	CD	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
12. Outcome assessors blinded?	No	No	No	No	No	No	No	No	No	No	No	No
13. Follow-up loss under 20%?	NA	NA	Yes	NA	NA	NA	NA	Yes	NA	NA	Yes	NA
14. Measurement of confounding variables?	CD	CD	No	NR	Yes	NR	NR	NR	Yes	NR	Yes	Yes
Quality Rating	**Fair**	**Poor**	**Good**	**Poor**	**Fair**	**Poor**	**Fair**	**Good**	**Fair**	**Fair**	**Good**	**Good**

Criteria	Starkstein et al. 1995	Feinstein et al. 1997	Foley et al. 2015	Tortelli et al. 2016	Patel et al. 2018	Vidović et al. 2015	Fitzgerald et al. 2018	Choi et al. 2013	Lopez et al. 2001	Eccles et al. 1999	Calvert et al. 1998	McGrath, 2000	Burns et al. 1999
1. Clear research question?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
2. Study population defined?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
3. Participation rate at least 50%?	CD	CD	CD	CD	Yes	CD	CD	NR	CD	Yes	NR	Yes	Yes
4. Inclusion and exclusion criteria prespecified?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
5. Sample size justification?	No	No	No	No	No	No	No	No	No	No	No	No	No
6. Exposure(s) of interest measured prior to the outcome?	No	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	No	No	Yes	Yes
7. Timeframe between measures sufficient?	Yes	Yes	CD	CD	CD	CD	Yes	Yes	Yes	CD	CD	CD	CD
8. Different levels of the exposure?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	NA	Yes	Yes	Yes	Yes
9. Exposure measures clearly defined?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
10. Exposure(s) assessed more than once?	No	No	No	No	No	No	No	No	No	No	No	No	Yes
11. Outcome measures clearly defined?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
12. Outcome assessors blinded?	Yes	No	No	No	No	No	No	No	No	No	No	No	Yes
13. Follow-up loss under 20%?	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	No
14. Measurement of confounding variables?	NR	CD	NR	NR	Yes	NR	NR	Yes	NR	NR	Yes	NR	NR
Quality Rating	**Fair**	**Fair**	**Fair**	**Fair**	**Good**	**Fair**	**Fair**	**Good**	**Fair**	**Fair**	**Fair**	**Fair**	**Good**

Results of individual studies

Participant/Disorder characteristics predictors and correlates

Overall, female gender was associated with emotionalism for participants with stroke, multiple sclerosis, TBI, ALS and Parkinson's disease (Kim & Choi-Kwon, 2000; McGrath, 2000; Phuong et al., 2009; Foley et al., 2015; Vidović et al., 2015; Thakore & Pioro, 2017). However, Kim (2002) found no relationship between gender and emotionalism in participants following a stroke. This study included 25 participants with emotionalism and was rated 'fair' in terms of methodological quality with the participation rate not able to be determined, which could decrease the power of the study. Lower education level (Hanna et al., 2016; Fitzgerald et al., 2018) and non-white ethnicity (Fitzgerald et al., 2018) were identified as predictors of emotionalism in a cohort of participants with multiple sclerosis.

Additionally, a correlation between emotionalism and a younger age was reported for participants with stroke and ALS (Tang et al., 2004; Thakore & Pioro, 2017). Patel et al. (2018) similarly found emotionalism was significantly correlated with younger age for individuals with ALS but no difference was identified for individuals with Parkinson's disease. This study had a fairly small sample size with only 31 participants with emotionalism whereas Thakore and Pioro (2017) included 209 participants with emotionalism.

Studies exploring predictors of emotionalism and stroke reported an association between a previous history of stroke (Tang et al., 2004; Choi et al., 2013), motor and sensory dysfunction (Kim & Choi-Kwon et al., 2000; Choi-Kwon et al., 2012; Wei et al., 2016), higher NIHSS scores (Tang et al., 2004; Choi-Kwon

et al., 2012; Choi et al., 2013; Ko et al., 2017), higher BI score (Andersen et al., 1995; Choi et al., 2013) and mRS score (Choi-Kwon et al., 2012; Ko et al., 2017).

Disease characteristics were investigated by only a small number of studies. Shorter disease duration (Tortelli et al., 2016) and rapidly progressive disease (Thakore & Pioro, 2017) in participants with ALS, greater disease severity in participants with multiple sclerosis (Vidović et al., 2015) and longer illness duration in participants with Alzheimer's disease (Starkstein et al., 1995) were associated with emotionalism. Furthermore, a higher level of disability was associated with emotionalism in participants with Parkinson's disease (Siddiqui et al., 2009).

Neurophysiological predictors and correlates

Out of the total studies identified for this review the majority of studies explored neurophysiological predictors and correlates of emotionalism in participants following a stroke. Therefore, a summary for the reader has been provided according to each neurological disorder.

Stroke

Lesion size was found to be significantly larger in participants with emotionalism post-stroke (Andersen et al., 1995). House et al. (1989) explored predictors longitudinally and revealed an association between larger lesions at one month, lesions in left frontal and temporal regions at 6 months and anterior lesions at 12 months. Additionally, the number of lesions was commonly investigated as a predictor of emotionalism in participants following a stroke, which could be an indication of the extent of damage to the brain. Higher frequency of lesions in right anterior region at assessment (MacHale et al., 1998), bilateral lesions (Wang et al., 2016) and more lesions in the globus pallidus and dorsally located (Kim, 2002)

were predictors of emotionalism. A further study found a significant correlation between more infarcts in frontal and/or basal ganglia and a significant correlation between frontal infarct and severity of emotionalism (Tang et al., 2009[b]).

Overall, there was a higher number of studies exploring lesion location as a predictor of emotionalism in participants who experienced a stroke. Lesions in the right frontal/anterior region featured more with an association with anterior cortical stroke (Kim & Choi-Kwon, 2000), cortical infarcts (Tang et al., 2004), lesion location at admission (Choi-Kwon et al., 2012; Ko et al., 2017) and at three months (Ko et al., 2017) associated with emotionalism. Evidence reporting single lesions in the anterior regions of cerebral hemisphere were four times the odds of emotionalism than lesions located elsewhere (Morris, Robinson & Raphael, 1993) and only left anterior lesions were significantly associated with emotionalism at one, six and 12 months (House et al., 1989). Three months post-stroke anterior cortex, pons and midbrain infarction, bilateral lesion location and severe white matter changes were also identified as significant risk factors associated with emotionalism (Wei et al., 2016).

Research also highlighted evidence of further brain areas involved in emotionalism specifically the brainstem and posterior structures. Andersen et al. (1994) classified participants with emotionalism in terms of severity. They found those classed as most severe had relatively large bilateral pontine lesions without lesions in the hemispheres, those classed intermediate had bilateral central hemispheric lesions and those classed least affected had mainly unilateral subcortical lesions. Furthermore, Liu et al. (2017) investigated specific brain networks and revealed differences in the amplitude of low frequency fluctuation and regional homogeneity in the default mode network, sensorimotor network,

affective network and cerebellar lobes. However, both these studies were rated as 'poor' for methodological quality as authors did not report if participation rate was above 50%, if confounding variables were accounted for and no sample size justification was provided.

The presence of microbleeds was investigated by several studies. Tang et al. (2009[a]) found individuals with PSE had higher frequency of microbleeds in the thalamus as a whole, anterior and paramedian areas and higher number in the entire brain. Only microbleeds in the thalamus was a significant independent predictor of emotionalism however. Furthermore, the presence of microbleeds was associated with emotionalism at admission (Choi-Kwon et al., 2012).

Studies exploring the serotonergic system as a predictor of emotionalism examined this by investigating genes and medication effectiveness. Disruptions to serotonergic pathways or abnormalities were implicated in a number of studies. Kim et al. (2012) found higher frequency of 5-HTTLPR 5 allele of participants with emotionalism. An association between 5-HTTLPR genotype and PSEI strengthened progressively with increasing number of 5 alleles and remained significant in participants with 5/5 genotype. Further studies found TPH2 rs4641528 allele carriers were associated with emotionalism at admission (Ko et al., 2017) and STin2 VNTR was one factor associated at 3 months (Choi-Kwon et al., 2012). Administration of Sertraline (Burns et al., 1999), Fluoxetine (Brown et al., 1998) and Citalopram medicines (Andersen et al., 1993) resulted in significant improvements in emotionalism scores compared with a placebo group. These studies were rated as 'good' for methodological quality with clear research questions, exposure measured prior to outcome and well-defined outcome measures. Taken together, this evidence of clinical improvement following the

administration of medication targeting serotonergic pathways, suggests the serotonin system may be involved in the pathophysiology of emotionalism.

Two studies rated as 'fair' according to QATOCCS found lower total testosterone levels were independently associated with emotionalism (Choi et al., 2018) and midbrain/pons [I]β-CIT binding ratios were significantly lower in those with emotionalism (Murai et al., 2003). These were both pilot studies with small sample sizes for participants with emotionalism (N= <6), exposure was not measured more than once and researchers were not blinded to outcomes.

Multiple sclerosis

Lesion volume and location was investigated in participants with multiple sclerosis. An inverse relationship for those with emotionalism and without depression was found with fewer posterior lesions associated with emotionalism (Luhoway et al., 2019). Ghaffar et al. (2008) found brainstem hypointense lesion volume was significantly higher in individuals with emotionalism and differences in hyperintense lesion volume in five regions: right medial inferior frontal, right inferior parietal, left medial inferior frontal and left inferior parietal. A logistic regression model, which accounted for 70% of the variance identified brainstem hypointense, left inferior parietal hyperintense and left and right medial inferior frontal hyperintense lesion volumes as factors in explaining the presence of emotionalism. This further supports the evidence found in stroke patients with damage to brainstem and posterior structures. Furthermore, frontal lobes are more dense with serotonin receptors and the raphe nuclei located in the brain stem are heavily implicated in serotonin production which is suggestive of a serotonin related network which, if disrupted in any specific location might increase vulnerability to emotional expression in response to a trigger/stressor.

Investigations of activations of brain areas in response to emotional and neutral stimuli were explored to identify differences in multiple sclerosis participants with emotionalism. Distinct activations in areas involved in emotional processing, high-level and associative visual processing in response to neutral stimuli (Haiman et al., 2009) and somatosensory and motor areas in response to neutral stimuli and higher current density were revealed (Haiman et al., 2008). This suggests that individuals with emotionalism show greater emotional reactivity to neutral stimuli in certain brain areas compared to individuals without emotionalism.

ALS

Overall, there was evidence of white and grey matter changes in participants with ALS and emotionalism. Evidence of disruptions to the corticobulbar/cerebellar pathways that regulate motor control and co-ordination of emotional expression were highlighted in this review. Floeter et al. (2014) found increased mean diffusivity of white matter tracts underlying the frontotemporal cortex, transverse pontine fibers and middle cerebellar peduncle in individuals with emotionalism. Christidi et al. (2018) found white matter abnormalities in associative and ponto-cerebellar tracts and decreased grey matter volume in the left orbitofrontal cortex, frontal operculum, putamen and bilateral frontal poles. Additionally, they found decreased fractional anisotropy in left posterior cingulum and posterior corona radiata.

Electrophysiological differences were explored to investigate the role of the frontal cortex as expressed by the ECAS score with participants with emotionalism. Hübers (2016) found changes in EMG activity of mimic muscles in individuals with emotionalism compared with controls. They concluded reduced inhibitory

activity in the frontal cortex could explain changes in physiological parameters in relation to emotionalism. However, it should be noted that the methodological quality of this paper was rated as 'poor' as the predictors were not measured prior to the outcome, the timeframe between measures could not be determined and the authors did not report whether confounding variables were controlled for.

Several studies found an association between bulbar onset, lower bulbar and gross motor ALSFRS-R sub-scores and emotionalism in a sample of participants with ALS (Floeter et al., 2014; Tortelli et al., 2016; Thakore & Pioro, 2017).

TBI

Only one study exploring neurophysiological predictors in patients with TBI was identified in this review providing evidence of damage to frontal lobes. In a sample of participants with TBI, greater frequency of frontal lobe injury, more diffuse lesions and lateral left frontal lobe were associated with emotionalism (Tateno et al., 2004). This further supports the evidence identified in other neurological disorders of a serotonergic hypothesis implicating the serotonergic pathways/networks involving raphe nuclei, frontal white matter projections including thalamus and frontal areas.

Alzheimer's disease

Studies investigating predictors of emotionalism varied in terms of variables explored in participants with Alzheimer's disease. Lebert et al. (1994) explored cerebral laterality and found frontolateral asymmetry indices were significantly lower in those with emotionalism. Significant differences in anatomical predictors were identified in several studies. Starkstein et al. (1995) found mixed pathological affect had significantly larger left ventricle compared with pathological crying affect or no pathological affect. Additionally, emotionalism was associated with

decreased rel-CBF in the anterior cingulate and dorsolateral prefrontal cortices bilaterally and in the left basal ganglia and increased rel-CBF in the right middle temporal area (Lopez et al., 2001).

Further implications of the serotonergic pathways were highlighted with one study which revealed significantly lower (2.9- and 2.6-times) platelet 5-HT concentrations in individuals with emotionalism (Prokšelj et al., 2014).

Neuropsychological predictors and correlates

General intellectual impairments or global functioning were assessed by seven studies with evidence that mild cognitive impairment (MCI) was significantly related to emotionalism in participants who had experienced a stroke (Wang et al., 2016) and MMSE scores were a significant predictor of post-stroke laughter (Tang et al., 2009[a]). Those with emotionalism were found to have greater intellectual impairments at one and six-months following a stroke (House et al., 1989). However, another study found an association between emotionalism and intellectual impairments at six and 12-months post-stroke but no association at one-month (Andersen et al., 1995).

Participants with emotionalism had lower performance and full-scale IQ scores on the Wechsler Adult Intelligence Scale-Revised (WAIS-R) whereby those with emotionalism were more impaired on a single verbal subscale, on the Digit Symbol and Picture Arrangement tests (Feinstein et al., 1997). Furthermore, emotionalism was associated with increased odds of moderate versus mild cognitive impairments in individuals with multiple sclerosis (Fitzgerald et al., 2018). This possibly suggests more global impairments reflect the extent of damage to the brain and could indicate a likely impact on network functioning rather than a specific lesion location for emotionalism.

A number of studies included measures of executive functioning to explore the cognitive correlates of emotionalism based on the hypothesis executive/inhibitory control might be implicated with emotionalism. The Wisconsin Card Sorting Task (WCST) is a measure of frontal lobe function whereby performance in this task is considered to be sensitive to the dorsolateral prefrontal function and lesions (Berman et al, 1995). Evidence suggested that those with emotionalism and multiple sclerosis generated significantly less words on the Controlled Oral Word Association Test (COWAT), took longer to perform the Stroop test and showed a trend for more total errors on the WSCT (Feinstein et al., 1999). Additionally, those with ALS and emotionalism made significantly more total errors on the WSCT and more perseverative errors (McCullagh et al., 1999). The authors also found that WSCT total errors predicted emotionalism with 75% accuracy. However, this research was rated as 'fair' as the participation rate and a sufficient timeframe between the measures were not able to be determined. These studies highlighted deficits in executive functioning but did not state which specific components of executive functioning were associated with greater emotionalism.

Frontal dysfunction was highlighted by a case-control study, which revealed participants with emotionalism had significantly lower Chinese Frontal Assessment Battery Scores (Tang et al., 2009[b]). Furthermore, evidence found a negative correlation with performance on several cognitive subtests in a sample of patients with emotionalism and multiple sclerosis. Hannah et al. (2016) revealed deficits in verbal fluency (COWAT), visual memory (Brief Visuospatial Memory Test-Revised; BVMTR immediate and delayed recall and California Verbal Learning Test-2 Immediate Recall; CVLT2-IR and California Verbal Learning Test-2 Delayed Recall; CVLT2-DR), slower processing speed (Paced Auditory Serial

Addition Test; PASAT) and executive dysfunction (Delis-Kaplan Executive Function System; D-KEFS card sort and card sort description). This study was rated as methodologically 'good' for this review as they controlled for variables such as years of education, had clear research questions and variables were clearly defined.

Psychological predictors and correlates

There was limited research exploring psychological predictors and correlates in participants with neurological disorders. In this review, psychological factors were only investigated in a stroke population and the psychological impact of emotionalism was investigated by several studies. There was evidence that irritability and ideas of reference were associated with emotionalism (Calvert et al., 1998). One study reported associations of emotionalism with the Impact of Events (IES) subscales of 'intrusion' and 'avoidance' and the Mental Adjustment to Stroke Scale (MASS) subscales 'helplessness/hopelessness' and 'anxious preoccupation' (Eccles et al., 1999).

Variables such as ways of coping or social support from others were investigated. Low social support was independently related to emotionalism three months after stroke (Choi-Kwon et al., 2012). Wei et al. (2016) found 'avoidance', 'acceptance-resignation' and low 'social support' subscales were predisposing factors for emotionalism. Additionally, 'acceptance-resignation' and 'avoidance' were associated with emotionalism three months after stroke. Both these studies were rated as methodologically 'good' for the purpose of this review with clearly defined research questions, measures and less than 20% follow-up dropout rate.

Additional analysis

As the majority of studies explored neurophysiological, neuropsychological and psychological predictors and correlates of emotionalism in a stroke population only tentative comparisons of predictors between neurological disorders were completed. As previously discussed, shared participant/disorder characteristic predictors of lower age and female gender were found in participants with stroke, multiple sclerosis, TBI, Parkinson's disease and ALS. Interestingly, two studies found emotionalism was associated with a shorter duration of ALS (Tortelli et al., 2016; Thakore & Pioro, 2017) whereas a longer illness duration was associated with emotionalism in participants with Alzheimer's disease (Starkstein et al., 1995).

Studies investigating neurophysiological predictors found similarities in terms of white matter changes in both stroke and ALS groups. A number of studies exploring predictors in a cohort of stroke participants revealed similar variables associated with emotionalism such as larger lesion sizes and presence of microbleeds. Similarly, level of cognitive impairment was associated with emotionalism in both participants with stroke and multiple sclerosis.

Synthesis of results

The majority of studies included in this review focused on neurophysiological predictors of emotionalism in participants following a stroke with only a limited number of psychological predictors explored and only with stroke participants. No studies were identified as appropriate for this review that included participants with vascular dementia. There are methodological variations in the studies included in this review with some rated as 'poor' for the purpose of this review so the findings need to be considered cautiously. Overall, this review

identified common predictors and correlates such as bulbar networks, serotonergic pathways, frontal areas, white matter, genetics, executive functioning, coping style and female gender as potentially involved in the pathophysiology of emotionalism. The evidence in the review emphasises the importance of serotonin which highlights any brain area that is relatively more involved in serotonin (production, modulation of function) might show up as more likely damaged across neurological populations. This could suggest there is not a specific anatomical or neuropsychological 'signature' because of the widespread presence of serotonin related mechanisms in the brain and beyond. However, findings from the review implicated bulbar and frontal areas as well as white matter tracts involved in connecting frontal, posterior/brain stem/midbrain regions. Potentially a diathesis-stress model of emotionalism could be tentatively proposed whereby if serotonin pathways are disrupted in any specific location this might increase vulnerability to emotional expression in response to a trigger/stressor and in turn facilitating avoidance. However, there are some stronger associations perhaps reflective of areas more heavily implicated in serotoninergic activity.

Table 6 summarises findings for all predictors and correlates across the neurological disorders. A number in brackets has been added to indicate the number of times findings have been found in different studies and predictors and correlates have been bolded to indicate these were found across different neurological disorders.

Table 6: Summary of predictors and correlates across neurological disorders

Neurological Disorder	Characteristics	Neurophysiological	Neuropsychological	Psychological
Stroke	Previous history of stroke (2) Higher NIHSS score (4) Higher BI score (2) mRS score (2) **Female gender** Motor (2) and sensory dysfunction **Younger age**	Higher 5-HTTLPR 5 allele ALDD and ReHo differences Lesion location (9) Higher frequency of lesions (2) Left hemisphere lesion Larger lesion size (4) **White matter changes (2)** Microbleeds (2) Lower testosterone levels Ischemic stroke Lower midbrain/pons [I]β-CIT binding ratios Pronounced hypoperfusion STin2 VNTR polymorphism TPH2 SNP rs4641528	Intellectual impairment (2) Lower Chinese Frontal Assessment Battery Scores **Mild Cognitive Impairment** MMSE	Avoidance (2) Acceptance-resignation Low social support (2) Irritability Ideas of reference Intrusion Helplessness/hopelessness Anxious preoccupation
Multiple sclerosis	Lower education level (2) **Female gender** **Younger age** Non-white ethnicity Greater disease severity	Somatosensory and motor areas Higher current density Activation of emotional processing areas Hyperintense lesion volume differences Fewer posterior fossa lesions	Negatively correlated with COWAT, BVMTR items, PASAT, DKEFS card sort, CVLT2-IR and CVLT2-DR Less words generated on COWAT and more total errors Longer to perform Stroop test (2) Lower performance and full-scale IQ scores **Increase odds of moderate cognitive impairment**	
TBI	**Female gender**	Greater frequency of frontal lobe injury, diffuse lesions Lateral left frontal lobe Damage to right cerebral hemisphere Bilateral lesions, unilateral lesions, brain cortex and basal ganglia, cortical and subcortical areas		
Alzheimer's disease	Longer illness duration	Frontal asymmetry indices lower Lower platelet 5-HT concentrations Larger left lateral ventricle Higher anosognosia scores Rel-CBF levels		
ALS	**Female gender** **Lower age** Shorter disease duration (2) Rapidly progressive disease	Decreased grey matter volume, fractional anisotropy **White matter abnormalities** Bulbar onset (2) Lower ALSFRS-R score (2) EMG activity	WSCT total errors and more preservative errors	
Parkinson's disease	**Female gender** Higher levels of disability	Higher unified Parkinson's disease rating scale salivation, axial rigidity, bradykinesia and gait disturbance scores		

** Predictors and correlates bolded indicate those found across different neurological*
** A number in brackets has been added to indicate the number of times findings have been found in different studies*

Discussion

Summary of evidence

To date this is the first systematic review that has provided a comprehensive narrative synthesis of the published research exploring the predictors and correlates of emotionalism across neurological disorders. A total of 50 studies were included in this review and overall the quality ratings of the studies ranged from 'good' to 'fair'. The largest amount of evidence revealed neurophysiological predictors and correlates of emotionalism across neurological disorders, however a large majority of these predictors were investigated in only stroke participants. Due to the disproportionate number of studies across the neurological disorders, the review provides patterns of predictors and correlates for each disorder and tentatively compares across disorders. The current review identified shared predictors and correlates that were found in several neurological disorders such as bulbar networks, serotonergic pathways, frontal areas, white matter, genetics, executive functioning, psychological impact, coping style and female gender. This review highlights the need for further high-quality research exploring emotionalism across neurological disorders to validate these findings and to enhance theoretical understanding.

Strengths and Limitations

Key strengths of this review were the use of a systematic approach, a clear predefined protocol and the inclusion of quality checks or assessment of biases. This meant that the methodological quality of studies could be appraised and researcher bias was less likely, which allowed for the review to summarise the evidence highlighting strengths and limitations of research. Additionally, the review explored predictors and correlates of emotionalism across neurological disorders, which enabled a greater understanding of this condition and for similarities and differences to be tentatively investigated.

This review has a number of limitations. Firstly, due to the significant heterogeneity in how emotionalism was measured and small sample sizes, a narrative synthesis was completed rather than a meta-analysis. It has been highlighted that narrative syntheses lack transparency, there is an increased potential for bias and conclusions are based on subjective interpretation (Valentine et al., 2017). To minimise researcher bias, a systematic review protocol was registered before commencing with aims, search strategy, data analysis plan and an assessment to measure risk of methodological bias in the studies outlined. The review conforms to the guidelines outlined by the Preferred Reporting Items for Systematic Reviews and Meta-Analyses statement (PRISMA; Moher et al., 2009).

A second limitation was that this review excluded studies not published in English and 'grey' literature, which could limit the generalisability of the findings. It has been highlighted that including 'grey' literature can minimise the effects of publication bias and provide a more balanced understanding of the evidence (McAuley et al., 2000). Although the inclusion criteria for this review included a number of neurological disorders, the majority of studies focused on predictors of emotionalism in participants following a stroke whereby there was limited research focusing on the other neurological disorders. For example, only three studies were identified that explored predictors in a sample of participants following a TBI and no studies investigated predictors and correlates in participants with vascular dementia. Therefore, further analysis of the results to compare predictors of emotionalism across neurological disorders was completed tentatively.

Additionally, the range of study designs varied with the majority of studies using a cross-sectional or case-control design and only a few RCT's. RCT's have high internal validity, minimise the risk of bias by controlling for confounding variables and participants are randomised, which allows for causation to be explored (Booth & Tannock, 2014). In contrast, cross-sectional studies measure exposure and outcome at the same time whereby

it is difficult to derive causal relationships (Wang & Cheng, 2020). The conclusions drawn from this review acknowledge that causation is difficult to determine and highlights the need for further RCT studies or longitudinal studies that use appropriate sampling and controls to be completed in the future.

The methodological quality of each study was rated using the QATOCCS. In this review there was variation in the overall ratings with the majority of the studies rated as 'good' or 'fair' and a few studies rated as 'poor'. For this review a total of 25% of studies were rated by a second independent rater. As only 25% of studies were reviewed independently this increases the risk of bias with ratings based on the interpretation of the rater.

A further limitation included the wide-ranging methods used in studies to measure emotionalism across neurological disorders. Some studies measured emotionalism based on clinical judgement or criteria assessed by a physician, which could increase bias and potential error whereby this is based on subjective interpretation. However, a number of studies used the Center for Neurological Study – Lability Scale (CNS-LS; Moore et al., 1997), which has been shown to have good test-retest reliability and internal consistency.

The systematic review identified a large number of studies to be included which helps to advance theoretical and clinical understanding of emotionalism across neurological disorders. However, due to the heterogeneity of conditions, diverse populations, range of independent variables and differences in how emotionalism was measured this resulted in a large number of studies (N=50) to be included in this review. This highlights challenges in aggregating the results and drawing specific conclusions or implications on specific predictors or correlates of emotionalism for neurological disorders.

Interpretation of findings

The findings from this review will be discussed further in relation to each predictor and/or correlate across the neurological disorders.

Participant/Disorder characteristic predictors and correlates

Evidence suggested that female gender (Kim & Choi-Kwon, 2000; McGrath, 2000; Phuong et al., 2009; Foley et al., 2015; Vidović et al., 2015; Thakore & Pioro, 2017) and a younger age (Tang et al., 2004; Thakore & Pioro, 2017; Patel et al., 2018) were associated with emotionalism in participants with stroke, multiple sclerosis and ALS. However, not all research controlled for confounding variables, which increases the risk of bias and could limit the generalisability of these findings. This research highlights important factors clinicians may consider in clinical practice whereby further support to aid prevention of emotionalism or psycho-education to help with treatment could be offered to individuals who are younger or female.

Interestingly, participant and stroke specific characteristic predictors and correlates suggested strong evidence for the association between emotionalism and history of previous strokes (Tang et al., 2004; Choi et al., 2013), higher NIHSS score (Tang et al., 2004; Choi-Kwon et al., 2012; Choi et al., 2013; Ko et al., 2017), motor and sensory dysfunction (Kim & Choi-Kwon et al., 2000; Choi-Kwon et al., 2012; Wei et al., 2016), higher BI score (Andersen et al., 1995; Choi et al., 2013) and mRS score (Choi-Kwon et al., 2012; Ko et al., 2017). These factors could highlight general severity of stroke and the extent of damage to brain areas, which is consistent with the neuropsychological findings of an association between emotionalism and poorer intellectual functioning. However, this highlights a threshold effect whereby the greater the degree of cognitive deterioration the more likely it will be that areas specific to emotionalism will be implicated so research needs to control for this. Additionally, these findings could support the neurochemical

serotonin hypothesis whereby more brain areas/greater damage leads to greater serotonin network disruptions which leads to dysregulation of affective responding.

Neurophysiological predictors and correlates

The findings summarised in this review support previous theories and hypotheses about the mechanisms of emotionalism that have been proposed. An early theory of the pathophysiology of emotionalism proposed emotionalism may be due to disruptions to the cortical inhibition to the upper brainstem centre and release of the lower bulbar nuclei (Wilson, 1924). This review offers support for this theory with an association between lesions located at the pons and PSPLC identified with pontine lesion independently related to PSPLC in participants following a stroke (Wang et al., 2016).

Furthermore, there was a considerable amount of research included in this review which supported the gate control theory highlighting the role of the cerebellum in the modulation of emotion and cerebellar pathways or lesions from the motor, frontal and temporal lobes to the brainstem (Parvizi et al., 2009). Research found individuals with emotionalism had increased mean diffusivity of white matter tracts underlying the frontotemporal cortex, the transverse pontine fibers and the middle cerebellar peduncle following a stroke (Floeter et al., 2014). Also, the left cerebellum posterior lobe was significantly lower for ALS individuals with emotionalism (Liu et al., 2017). Additionally, there was a greater frequency of frontal lobe injury and a difference in the frequency of frontal lobe lesions in individuals with emotionalism following a TBI (Tateno et al., 2004).

These neurophysiological findings highlight the possible involvement of the frontostriatal network, which consists of both bulbar and frontal inhibition (Wiecki & Frank, 2013). Furthermore, frontal-subcortical circuits which mediate motor activity and behaviour in humans could be implicated (Tekin & Cummings, 2002). The frontal-subcortical circuits link specific areas of the frontal cortex to the basal ganglia and

thalamus. In this review lesions to brain areas involved in these networks/circuits have been highlighted across neurological disorders. This emphasises important possible mechanisms of emotionalism which could help enhance theoretical understanding of emotionalism and extend clinicians' understanding. However, further research is required to validate these findings.

Disruptions of neurotransmitters such as serotonin or dopamine have been hypothesised to lead to changes in emotional expression (Rabins & Arciniegas, 2007). Selective serotonin reuptake inhibitors (SSRI's), which increase the synaptic availability of serotonin were found to show improvements in emotionalism (Andersen et al., 1993; Brown et al., 1998; Burns et al., 1999). Additionally, midbrain/pons [I]β-CIT binding ratios of serotonin transporter densities were significantly lower in stroke participants with emotionalism (Murai et al., 2003). These studies included in this review suggested the role of serotonergic pathways in the pathophysiology of emotionalism and supports the neuroanatomical evidence discussed in this review of different brain areas which are intimately involved in the production of serotonin or have functions strongly modulated by serotonin as indicated by more dense occurrence of serotonin receptors. The serotonergic circuits in the brain have a large set of 5-HT receptors in the substantia nigra, the hippocampal formation, the hypothalamus, the amygdala, the striatum, and the frontal cortex (Charnay & Leger, 2010).

These different theories of the pathophysiology of emotionalism have been expanded upon over time. The earliest release hypothesis theory (Wilson, 1924) was amended and extended with the gate control theory which used neuroimaging studies to allow examination of more precise localisation of brain lesions rather than relying on post-mortem examinations. The gate-control theory suggested disruptions to the corticobulbar/cerebellar pathways or lesions in the frontal lobes may contribute to the

development of emotionalism (Parvizi et al., 2009). Furthermore, changes to neurotransmitters have been hypothesised to play a role in the development of emotionalism which support previous theories based on neurotransmitters involvement in the circuits believed to be involved in the pathophysiology of emotionalism (Rabins & Arciniegas, 2007). These theories seem to provide further insights into how possible neurophysiological elements such as brain circuits, anatomy and neurochemistry interact in the development of emotionalism. However, these theories have not attempted to explore neuropsychological or psychological mechanisms of emotionalism which could amend, extend or reject previous theories.

Neuropsychological predictors and correlates

Neuropsychological predictors and correlates were identified across three neurological disorders: stroke, multiple sclerosis and ALS. Overall, evidence of general intellectual impairments was revealed by several studies whereby emotionalism was correlated with a lower performance and full-scale IQ scores on the WAIS-R (Feinstein et al., 1997). Mild cognitive impairment following a stroke was associated with emotionalism (Wang et al., 2016) and there is an increased risk of moderate cognitive impairment of emotionalism in a sample of participants with multiple sclerosis (Fitzgerald et al., 2018). However, an association between emotionalism and intellectual impairments at six and 12-months post-stroke was identified but with no association at one-month (Andersen et al., 1995). This could highlight further questions of whether global impairments reflect the extent of damage in the brain and/or the likely impact on network functioning rather than specific lesion locations for emotionalism.

Investigations of executive functioning were assessed and revealed a negative correlation on DKEFS card sort and card sort description (Hannah et al., 2016). Additionally, individuals with emotionalism generated significantly less words on the

COWAT, revealed deficits in verbal fluency, visual memory, slower processing speed (Hannah et al., 2016), took longer to perform the Stroop test and a trend was revealed for patients with emotionalism making more total errors on the WSCT (Feinstein et al., 1999; McCullagh et al., 1999). These studies highlight individuals with emotionalism had impairments in inhibition and strategy generation.

This evidence could highlight the disruptions of the anterior cingulate cortex and prefrontal regions of the brain, which are involved in the executive attention network, which has a key component of executive functioning (Posner et al., 2007). Overall the findings of the neuropsychological studies are consistent with deficits in working memory, inhibitory control and regulating emotions. Although the evidence in this review is correlational, meaning it is difficult to draw definite conclusions due to issues with causality, these studies do suggest a relationship between certain neuropsychological factors and emotionalism, which should be further investigated.

Psychological predictors and correlates

There were only three studies exploring psychological predictors and correlates of emotionalism in participants following a stroke. Evidence of an association between avoidance and emotionalism was reported in two studies (Eccles et al., 1999; Wei et al., 2016). Previous research has found that emotionalism causes distress, embarrassment and avoidance of social interactions (Wortzel et al., 2012). In this review two studies also found an association between low social support and emotionalism. Further research would be beneficial to validate these findings and could be important to explore if associations exist in other neurological disorders. To date there is no psychological theory explaining emotionalism, however these findings indicate the potential to explore social support, social self-consciousness and related avoidance as possible modifiable psychological treatment targets with individuals with emotionalism.

Future research

Further research is required exploring psychological predictors and correlates in individuals following a stroke as only a limited number of studies were identified and no studies explored psychological predictors in other neurological disorders. Future research could help to identify potential reversible psychological/behavioural maintaining factors. This is clinically important as research has indicated how prevalent emotionalism is in neurological disorders whereby further research could help inform clinical practice and potential psychological treatments. It is important more longitudinal and RCT studies are carried out to explore potential predictors and correlates, which could help to overcome the limitation of causality raised with cross-sectional studies and increase the methodological quality. Further research is also required exploring predictors and correlates in neurological disorders such as vascular dementia as this review highlighted a disproportionate number of studies across neurological disorders.

Specifically, from the findings in this review, future research could investigate the hypothesis relating genetic vulnerability, serotonergic pathways, executive inhibitory control, avoidance and social support in the development and longer-term maintenance of emotionalism. Furthermore, there is a need for a better measurement of emotionalism as the majority of studies included in this review used either the CNS-LS (Moore et al., 1997) or PLACS (Robinson et al., 1993). Both these measures have limitations and undetermined psychometric characteristics in stroke populations and were not derived from consensus diagnostic criteria.

Conclusions and clinical implications

This was the first systematic review which investigated the predictors and correlates of emotionalism across neurological disorders. In summary, this review has highlighted key brain areas or networks which extends the previous published models with

both aspects of bulbar involvement and frontal inhibition networks potentially involved. Consistent with the involvement of these areas/networks, this review highlights disruptions to the serotoninergic pathways with potential for SSRI treatment and psychological variables such as avoidant coping style and social self-consciousness, which could be key possible factors for future psychological treatment models. This highlights important factors that could be considered by clinicians and health care policy whereby support is offered to individuals to assist with earlier identification of emotionalism following a diagnosis of a neurological disorder as well as offering treatment or management.

Declaration of interest

There is no conflict of interest.

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Chapter Two: Bridging Chapter

Bridging Chapter

This chapter outlines the connection between the systematic review and empirical paper and presents a rationale for how the research questions for the empirical paper were developed.

The systematic review investigated the neurophysiological, neuropsychological and psychological predictors and correlates of emotionalism across neurological disorders. The review highlighted research varied in methodological quality and it was difficult to draw definitive answers due to the disproportionate number of studies identified across neurological disorders. The majority of studies investigated neurophysiological predictors and correlates in participants following a stroke. A range of common predictors and correlates were found across different neurological disorders such as such as bulbar networks, serotonergic pathways and genetics. Furthermore, only psychological predictors and correlates were investigated in participants following a stroke with psychological variables of avoidant coping style and social self-consciousness associated with emotionalism. This review emphasised the need for further research to be conducted to gain further knowledge of emotionalism.

The empirical paper presented in the next chapter was developed to explore participants' lived experience of emotionalism over time. The systematic review identified psychological predictors and correlates of emotionalism in participants following a stroke, however research exploring psychological predictors and correlates was limited and further research is needed to inform clinical practice. To date to the authors' knowledge no research has investigated participants' lived experience of emotionalism over time. Therefore, a key focus for the empirical paper was to gain an understanding of participants' lived experience of emotionalism over time using a longitudinal qualitative approach. The empirical paper presents the major themes and sub-themes from the

qualitative content analysis and framework analysis. Each major theme is outlined with examples and descriptions of each sub-theme. Further analyses were completed to explore the differences between time-points in how participants described their experiences of emotionalism over time for each major theme. Finally, a framework/model to account for the experiences of emotionalism based on participants included in this research project is presented. This research is clinically important to gain further understanding of emotionalism and to help shape future psychological interventions.

An extended results chapter is presented following the empirical paper (Chapter five). Within this chapter further analyses were completed following clusters of participants over time to investigate changes within sub-themes. Participants were clustered into categories such as level of distress or embarrassment, feeling sad/not feeling sad in a deductive way based on a priori understanding of clinical characteristics from clinical practice and limited research to date. Finally, trajectory analyses were completed following individual participants across time-points which enabled an investigation of changes in frequency of codes within sub-themes rather than the whole sample of participants at one time-point.

Chapter Three: Empirical Paper

Prepared for Submission to Disability and Rehabilitation

Author Guidelines available in Appendix H.

Post-stroke emotionalism (PSE): A qualitative longitudinal study exploring individuals' experience with PSE

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Post-stroke emotionalism (PSE): A qualitative longitudinal study exploring individuals' experience with PSE

Background: Post-stroke emotionalism (PSE) is a common emotional consequence of stroke characterised by episodes of crying or laughing. There is only one published qualitative study exploring the experience of emotionalism to date.

Objectives: To explore individuals' experience of PSE, describe how experience of living with PSE may change over time, and develop a theoretical client-derived framework to shape future psychological interventions.

Method: A qualitative secondary analysis of pseudonymised pre-collected semi-structured interview data was completed. Participants were recruited from nine acute stroke units in Scotland. Interviews were completed at two-weeks, six-months and 12-months post-stroke.

Results: Data was analysed from 52 participants at two-weeks, 25 participants at six-months and 23 participants at 12-months. Three major themes were identified: '*In the moment*', describing characteristics and triggers, '*Ways of coping*', highlighted a range of coping strategies including avoidance or acceptance and '*Impact*', outlining the longer-term effects of PSE such as individuals' beliefs. Analysis of changes over time highlighted increases in participants' reporting of barriers to control, aspects of avoidance and wishing to hide emotional responses.

Conclusion: The results indicate specific psychological aspects of PSE which could be viable targets in psychological interventions such as increasing adaptive coping strategies and challenging negatively held beliefs.

Implications for rehabilitation

- Emotionalism can cause emotional consequences such as distress, embarrassment and fear.
- Increases in avoidance, withdrawing from social interactions, negative perceptions of emotionalism and episodes of emotionalism making no sense were highlighted over time.
- Helping individuals to develop insight into triggers for emotionalism could be beneficial to help gain understanding and awareness whereby PSE has the potential to be anticipated and adapted to psychologically.
- Potential adaptive responses such as acceptance or control and better anticipation of episodes of emotionalism could help to reduce the emotional consequences of PSE.

Keywords: stroke; emotionalism; experience; longitudinal qualitative analysis

Introduction

Stroke is a major public health problem and the single largest cause of adult disability in the United Kingdom (UK; Department of Health, 2007). There are over 1.2 million stroke survivors in the UK with one individual suffering a stroke every five minutes (Stroke Association, 2018) and approximately 30% of individuals who have had a stroke will experience another stroke (Public Health England, 2018). Worldwide approximately 15 million individuals suffer a stroke each year (World Health Organisation, 2002). Stroke can have a lasting impact on individual lives and on the lives of carers and families. The effects of stroke can vary considerably, but can include loss of mobility, language, vision, cognition and sensory function.

Stroke can also have wide ranging mood and emotional consequences. For many individuals, adjusting psychologically to the sudden onset functional changes of a stroke may lead to a phase of heightened distress, low mood and/or anxiety (Taylor et al., 2011). For some individuals, there may be more long-lasting mood difficulties. Research shows for example that one-third of stroke survivors experience post-stroke depression (Hackett & Pickles, 2014) and one-quarter experience post-stroke anxiety (Knapp et al., 2020).

A common but much less researched emotional consequence of stroke is emotionalism. The condition is also known as emotional incontinence, pseudobulbar affect, emotional lability, pathological laughing and crying or involuntary emotional expression disorder, and it is not stroke specific, arising following a range of neurological conditions including traumatic brain injury (TBI) and multiple sclerosis (Schiffer & Pope, 2005).

Emotionalism produces a lessening of the ability to control emotional expression (House et al., 1989). Individuals with post-stroke emotionalism (PSE) experience episodes of crying or laughing that occur with little or no warning and which are not under usual

social control. Typically, crying outbursts in PSE occur in situations in which the person would not have cried before the stroke. Emotionalism can cause significant distress for the individual, and lead to associated social avoidance and reduced quality of contact with others (Allman et al., 1992), as sufferers fear uncontrollable crying outbursts and the embarrassment associated with these. Research has also suggested PSE can impact rehabilitation efforts with sufferers less able to engage with services due to embarrassment or shame due to uncontrolled emotional outbursts (Allman, 1991).

The prevalence of PSE has been found to be high. Gillespie et al. (2016) conducted a systematic review and meta-analysis of 15 PSE prevalence studies. They found 17% of stroke survivors suffer from PSE acutely, 20% at six-months and 12% beyond six-months. It is thought that approximately 82% of individuals with PSE will experience crying episodes only, 15% experience both crying and laughing episodes and 2% experience laughing episodes only (Calvert et al., 1998). Due to the high prevalence of PSE further research is required to understand the frequency of episodes longitudinally, to explore how individuals experience these episodes and to provide reliable recommendations about the best way to treat or manage emotionalism. This could help to identify individuals who describe difficulties with their experience of emotionalism whereby future psychological interventions could be developed to help with rehabilitation.

Emotionalism has been associated with depression and these two conditions have been found to be comorbid. Calvert et al. (1998) studied 448 stroke survivors and found 38 had emotionalism *and* a clinically significant mood disorder. However, there are distinct differences (Robinson et al., 1993). PSE is a disorder of emotional expression rather than emotional experience, so very often sufferers will report crying episodes but not feel sad on the inside. Approximately one-third of stroke survivors have been found to have depression one-month following their stroke and this remained at this level at nine-

months (Townend et al., 2010). Research has found that PSE is a risk factor for developing depression following a stroke within the first year (Carota et al., 2005).

Evidence of the effectiveness of pharmacological interventions has found that antidepressants have only a small positive effect, which is not specific to one drug or class of drugs (Hackett et al., 2010). Medication can produce unwanted side effects and some people living with stroke may not wish to take medication or may simply prefer non-pharmacological treatments. Currently there are no randomised trials of non-pharmacological treatments and no psychological or behavioural model to understand and formulate PSE. Gillespie et al. (2019) investigated the types of non-pharmacological interventions provided by stroke professionals in inpatient stroke settings. They revealed stroke clinicians reported regular use of non-pharmacological interventions with the most common interventions being offering reassurance and talking to patients about their goals. There is therefore an important gap in the literature whereby further research is required to improve our understanding of the psychological and behavioural aspects of PSE, to inform a psychological model of PSE and to help develop a future psychological intervention.

Research investigating how stroke survivors adjust psychologically to emotionalism after stroke is limited. Taylor et al. (2011) adapted the Social Cognitive Transition Model to cancer (Brennan, 2001) by adding stroke-specific components developed from the evidence base and clinical practice. The model proposes that an individual's assumptive world is important in shaping their adjustment following a stroke. If an individual's assumptive world is rigid and the experience of stroke disconfirms certain previously held assumptions this may impact on the duration and severity of adjustment distress. Furthermore, an attribution model of illness proposes those who believe that illness chronicity and severity is due to psychological causes express higher levels of distress and poorer psychosocial outcomes (King, 1983).

To date, however, there is one small scale qualitative study ever progressed to investigate stroke survivors' experiences of emotionalism and how they manage their symptoms. McAleese et al. (2019) conducted semi-structured interviews with N=18 individuals with PSE recruited from inpatient and outpatient stroke settings. They found individuals with negative experiences of PSE described associated social avoidance and embarrassment which impacted their quality of life. Additionally, social support, increased sense of control and optimism were found to increase positive experiences. The researchers used framework analysis to help develop the Post-Stroke Emotionalism Cognitions Questionnaire (PEC-Q) to measure cognitions that can be used as early detection of emotionalism. The sample size was small with participants interviewed at 4.3 months' post-stroke, so there was no consideration of the lived experience of PSE at the acute stage, and longitudinally, over time.

Therefore, it is important that larger scale qualitative research is conducted to more fully investigate stroke survivors' lived experiences of emotionalism, and to consider this not only across the acute phase, but also longitudinally, over time. This should in turn allow the development of a framework/model to help shape future psychological interventions, which have never yet been established.

Aims

The current study aimed to explore individuals' experience of PSE, ways of coping and to describe how experience of living with PSE may change over time. In line with a critical realist approach (Maxwell, 2012) a further aim was to explain these experiences and common mechanisms of emotionalism by developing a theoretical client-derived framework, which could help to shape future psychological interventions.

Research Questions

1. How do individuals with PSE describe their experience living with PSE?
2. Are there differences in how individuals with PSE describe their experience of emotionalism over time?
3. How do themes and changes in themes identified from questions one and two relate in contributing to a psychological account of the common mechanisms and experience of PSE over time?

Methodology

Design

A qualitative secondary analysis of pseudo-anonymised pre-collected interview data was completed. Interview data from individuals diagnosed with PSE was explored longitudinally using qualitative content analysis to provide a rich detailed account of the experience of individuals diagnosed with PSE over time. The researcher used a critical realist approach to understand participants' experience of emotionalism, which proposes individuals' views and experiences are considered to be 'real' entities and this allows an investigation of causal mechanisms with the potential to develop explanatory mechanisms of a phenomenon or event (Maxwell, 2012). Thereby, a theoretical client-derived framework which captures participants' experience and common mechanisms of emotionalism could be developed to improve understanding of stroke survivors' experience of PSE, which it is hoped may help shape future psychological interventions.

Ethical Approval

Ethical approval for the 'Testing for Emotionalism After Recent Stroke' (TEARS) study was granted from Scotland A Research Ethics Committee (reference 14/SS/1103; Integrated Research Application System reference 157483). As the current study is using the TEARS pre-collected data, a non-substantial amendment form was completed and

agreed by the study sponsor of the TEARS study indicating approval for the researcher's involvement for the current study. Following this, approval was sought and confirmed from University of East Anglia's Faculty of Medicine and Health Sciences (FMH) Research Ethics Committee. General Data Protection Regulation (GDPR) approval was granted from University of East Anglia's Data Protection Information Compliance Specialist.

Anonymised TEARS pre-collected data

This research project used pseudo-anonymised pre-collected data from the TEARS cohort study (Testing Emotionalism After Recent Stroke: NHS Research Scotland Stroke Research Network Identification 18980; https://www.stroke.org.uk/research/understanding-difficulty-controlling-emotions-after-stroke; full protocol from first author). The TEARS study aimed to determine the prevalence, impact, neurological and neuropsychological phenotypes of PSE. This was an observational, longitudinal study of PSE following individuals who experienced a stroke at two-weeks, six-months and 12-months.

Participants

Participants were recruited across nine acute stroke units in Scotland from October 1st 2015 to September 30th 2018. The same sample of participants were followed longitudinally at two-weeks, six-months and 12-months. Participants commenced the study at baseline (two-weeks) and either continued or dropped out.

Inclusion criteria for the TEARS study were: (a) clinical diagnosis of ischemic or hemorrhagic stroke (first ever or repeat); (b) written informed consent or informed consent from nearest relative/welfare guardian; (c) male or non-pregnant female over the age of 18. Individuals with a lack of spoken English, severe concurrent medical conditions or with life expectancy ≤ 3 months were excluded, as were individuals with distressing

behaviours secondary to stroke/dementia, subarachnoid haemorrhage, other extra axial bleeds or suspected transient ischaemic attack (TIA).

Participants were excluded if they did not consent to their interview being audio recorded.

Measures

Semi-structured diagnostic interview

The TEARS study interviews were conducted at three time-points; two-weeks, six-months and 12-months following stroke. Interviews were completed face-to-face at two-weeks and six-months and over the telephone at 12-months by pre-trained Scottish Stroke Research Network (SRRN) stroke research nurses. Participants completed a Testing for Emotionalism After Recent Stroke Interview (TEARS-IV) at each time point and the clinical interviews were recorded on an audio recorder. The semi-structured interview was developed based on House diagnostic criteria for PSE including increased tearfulness, crying suddenly with no warning, crying with a lack of control and crying occurring at least once a week (House et al., 1989; Calvert et al., 1998; Eccles et al., 1999). The interview included two screening questions whereby if participants answered "no" to both the interview was discontinued by the research nurses as they did not meet the criteria for emotionalism at that time-point. If participants answered "yes" to one or both screening questions the interview continued based on the participant meeting criteria for emotionalism. All participants answered the screening questions as part of the interview at each time-point to determine if emotionalism was absent or present.

The semi-structured interview included both open and closed questions and the interview covered; post-stroke crying screen questions, case characteristics, frequency and impact, post-stroke laughter screen questions, case characteristics and case summary (see Appendix M).

Data Analysis Plans

Qualitative analysis of the interview data was completed to explore participants' experience of emotionalism longitudinally. The analysis involved several steps: qualitative content analysis, framework analysis and recurrent cross-sectional/trajectory analysis. The interviews were transcribed and the transcriptions were imported to NVivo software (QSR International Pty Ltd, 2020). The study followed four main stages of qualitative content analysis as outlined in Table 1 (Bengtsson, 2016).

Table 1. Stages of qualitative content analysis.

Stage	Description
Decontextualisation	Identifying codes from text and creating a list of codes/meaning units.
Recontextualisation	Comparing codes with the original text to ensure the content is captured in the code.
Categorisation	Compiling codes into themes which summarise the collective codes.
Compilation	Draw conclusions from themes and codes to provide a deeper understanding.

The next stage of analysis involved framework analysis whereby the codes ('meaning units') and themes from the previous stage of analysis were organised into matrices (Ritchie et al., 2003). The interview questions were also used to shape the framework. The framework was applied to several transcripts that had certain framework themes. Finally, the themes and codes were explored longitudinally using recurrent cross-sectional analysis (Grossoehme & Lipstein, 2016). Matrices were created with themes

along the y-axis and time points across the x-axis. See Chapter Five for further information.

In line with a critical realist approach 25% of the codes were reviewed by a second independent coder (trainee clinical psychologist) to increase the 'trustworthiness' of the findings (Lincoln & Guba, 1985). Initial agreement between coders was 92%. Any discrepancies between coders were resolved through discussions and the revision of code definitions to reduce any overlap between codes and to ensure clarity. The newly revised code framework was reapplied to the transcripts and following this agreement rose to 100%.

Results

At two-weeks 52 of 62 participants (84%), at six-months 25 of 34 participants (73%) and at 12-months 23 of 27 participants (85%) consented to their interview being recorded thus providing the final included sample. Participants were followed across time-points with a total of 24 participants with PSE across more than one time-point, Figure 1. Across time-points 'new-onset' cases of PSE were identified such as participants who did not have PSE at baseline but did at six-months. At time-point two 14 participants continued to have PSE from time-point one with 11 participants with 'new-onset' PSE. At time-point three 10 participants continued to have PSE from time-point two with 13 participants with 'new-onset' PSE.

The length of interviews ranged from eight minutes to 29 minutes.

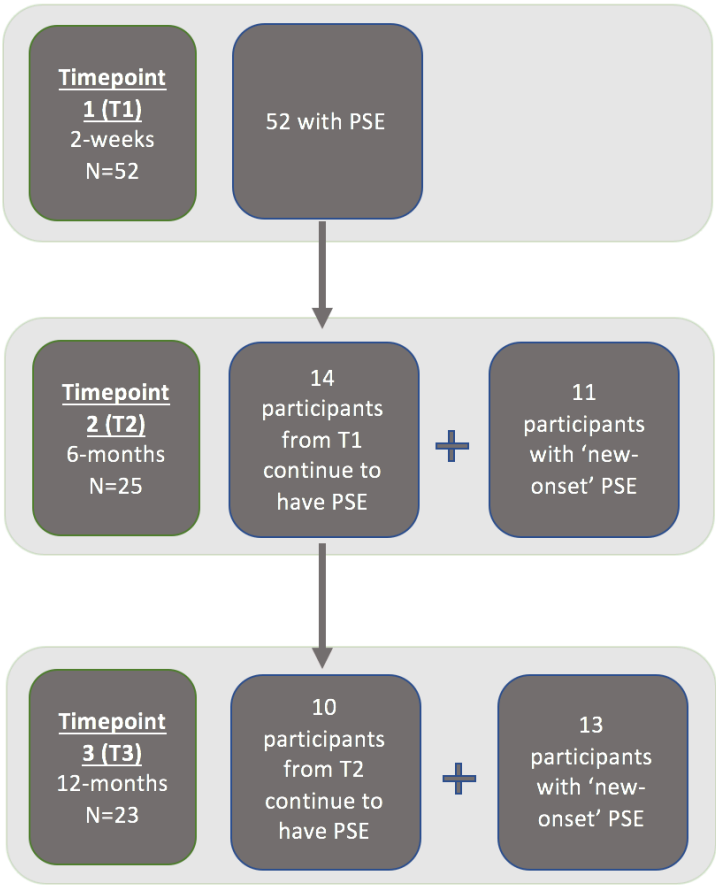

Figure 1. Flow of participants with emotionalism across time-points which shows 'new-onset' cases of emotionalism.

Descriptive statistics

The mean age of the sample at two-weeks was 59 and at both six-months and 12-months was 57 years. Similar numbers of men and women completed interviews across the time-points apart from at 12-months. The majority of participants left education after secondary school, had an infarct stroke, did not have a prior stroke and had a cortical or sub-cortical lesion location across the time-points. The mean Barthel Index score ranged from 15.7 to 17.25 (Table 2).

Table 2. Participant characteristics.

	Emotionalism at 2-weeks	Emotionalism at 6-months	Emotionalism at 12-months
Number	52	25	23
Age, years (X ±SD)	59±14.57	57±13.74	57±12.67
Sex			
Female	25 (48%)	10 (40%)	7 (30%)
Male	27 (52%)	15 (60%)	16 (70%)
Education level			
Primary	2 (4%)	0 (0%)	0 (0%)
Secondary	37 (71%)	17 (68%)	16 (69%)
College	5 (10%)	5 (20%)	4 (17%)
University	6 (11%)	2 (8%)	2 (8%)
Unknown	2 (4%)	1 (4%)	1 (4%)
Prior stroke			
Yes	10 (19%)	6 (24%)	6 (26%)
No	42 (81%)	19 (76%)	17 (73%)
Stroke type			
Infarct	49 (94%)	24 (96%)	19 (82%)
Haemorrhage	3 (6%)	1 (4%)	4 (17%)
Hemisphere affected			
Right	27 (52%)	16 (64%)	12 (52%)
Left	21 (40%)	8 (32%)	11 (47%)
Brainstem	1 (2%)	0 (0%)	0 (0%)
Left and right	3 (6%)	0 (0%)	0 (0%)
No value	0 (0%)	1 (4%)	0 (0%)
Lesion location			
Cortical	24 (46%)	10 (40%)	7 (30%)
Sub-cortical	24 (46%)	12 (48%)	14 (60%)
Brainstem	2 (4%)	1 (4%)	0 (0%)
Cortical and sub-cortical	1 (2%)	0 (0%)	0 (0%)
No value	1 (2%)	2 (8%)	2 (8%)
Barthel Index (X ±SD)	15.7±5.12	15.8±4.78	17.25±4.32

Research question one: How do individuals with PSE describe their experience of living with PSE?

Three master themes and 17 sub-themes emerged as providing an account of participants experience of living with PSE (Table 3). The first master theme '*In the moment*' highlights the experience that participants reported following the initial stages of an episode of emotionalism. The second master theme '*Ways of coping*' summarises participants' responses to episodes of emotionalism and ways in which they attempt to regain control. The final master theme '*Impact'* emphasises the emotional consequences in terms of identity and attempts to make sense of their experiences of PSE.

Table 3. Overview of master themes and sub-themes.

Master theme	Sub-theme
1. In the moment	a. Characteristics
	b. Control
	c. Barriers to control
	d. External triggers
	e. Internal triggers
	f. Predictability
	g. Unpredictability
2. Ways of coping	a. Acceptance
	b. Avoidance of triggers
	c. Concealing emotionalism from others
	d. Behavioural attempts at symptom management
	e. Cognitive attempts at symptom management
	f. Support from others
3. Impact	a. Changes to self
	b. Emotional impact
	c. Trying to make sense
	d. Uncertainty

Each major theme is presented with descriptions of each sub-theme whereby codes ('meaning units') and quotes are outlined. A visual overview of each major theme has also been provided for the reader.

Master theme one: 'In the moment'

This theme consists of seven sub-themes: *characteristics, control, barriers to control, external triggers, internal triggers, predictability* and *unpredictability* (Figure 2). These capture participants key elements of episodes of emotionalism based on their experience and factors that trigger the onset.

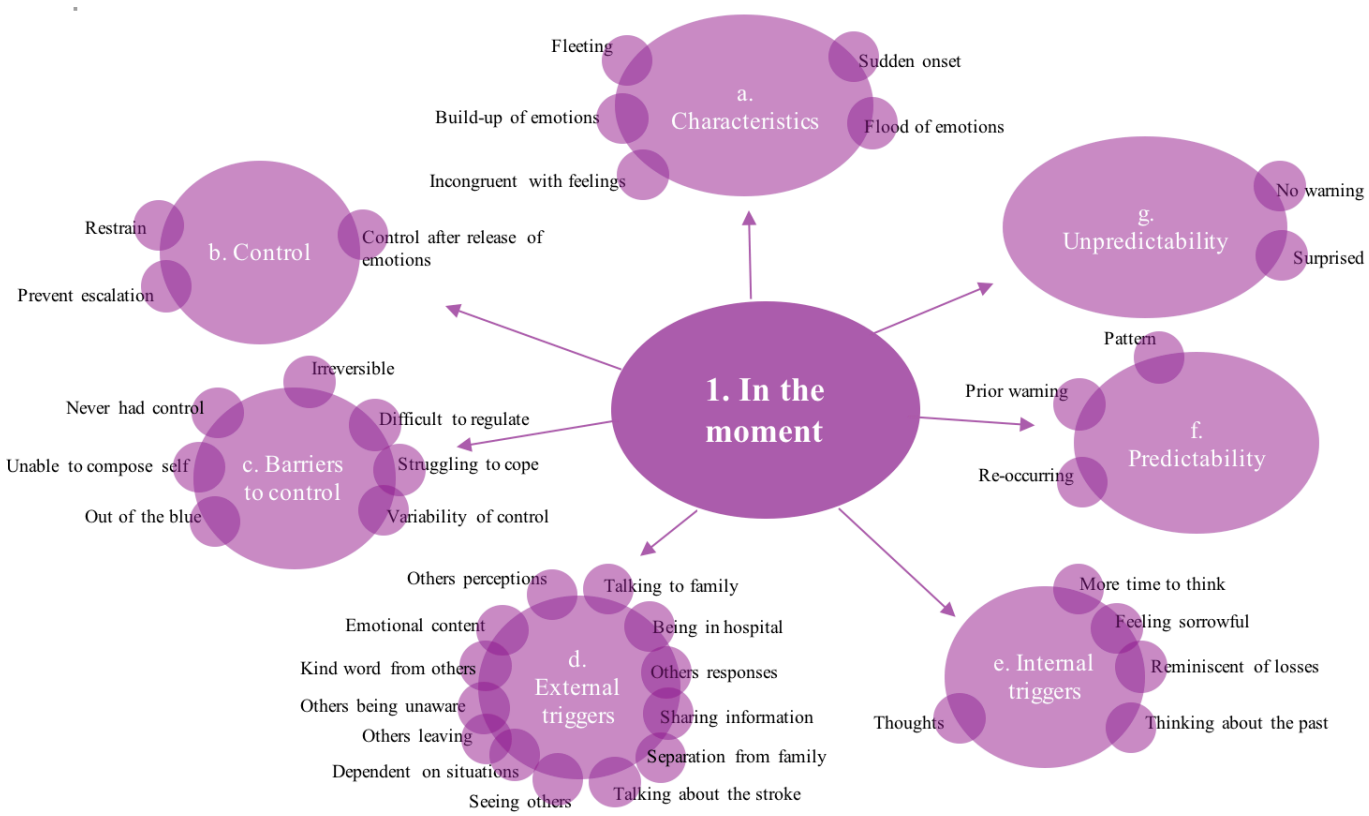

Figure 2. Codes and sub-themes within the 'In the moment' theme.

a. Characteristics

Participants described a range of key characteristics of emotional episodes with a build-up of emotions over time, which led to an emotional display.

> *"I get I suppose just a build-up of emotion I have never really thought about that one I suppose there is a bomb that is going to happen and you have no control at that point", 036, two-weeks.*

"it's just an emotional burst", 037, two-weeks.

"I can feel it bubbling up" 041, 12-months.

Additionally, episodes being incongruent with their feelings at the time led to a discord with participants, which was another frequently reported characteristic.

"I am not feeling sad and I just start crying", 016, six-months.

"when they come on I don't feel sad before they come on it hits me", 04, 12-months.

"it's when I have news whether it is good news or bad news I tend to be tearful", 010, 12-months.

The episodes were described by many participants as happening suddenly and lasting a few seconds to minutes.

"it feels as if like someone has a sledgehammer to you and all of a sudden", 066, two-weeks

"You just have tears for a minute or so and then it stops so its short bursts", 052, two-weeks.

"it is just really for a couple of seconds", 048, six-months

b. Control

This sub-theme captured participants' ability to regulate their episodes of emotionalism once they had released some of their emotion.

"I would say I can after I have got it out somewhat…I can kind of reign it in a little bit", 054, two-weeks.

"I would say probably about a minute…I can manage to do something about it", 075, two-weeks.

Other participants highlighted being able to restrain episodes of emotionalism whereby they could prevent the episode from developing or described a battle to stop the emotions from being displayed.

"I can feel it coming but I stop it so I stop that emotion coming out", 01, two-weeks.

"I could handle it…cos you can I think you can get a grip of yourself", 022, two-weeks.

"what happens is when you feel something like that coming on you try and fight it now", 052, six-months

c. Barriers to control

Some participants described a battle to control episodes of emotionalism once they had begun.

"I couldn't control it I tried my best, but I couldn't" 05, two-weeks.

"I find it very difficult at times to control it… it's terrible what gets you is you can't control" 010, 12-months.

"it's like you trying to control diarrhoea", 01, 12-months

Episodes occurring out of the blue were frequently reported by most participants as a barrier, which made controlling the episodes difficult or harder to compose themselves.

"I couldn't hold myself together so for the first couple of days I felt myself very prone to crying than previously which I felt unusual", 05, two-weeks.

"I am just sitting on a bus and before I know it I am starting to cry for no reason at all", 016, six-months.

"it's just completely out of the blue", 017, 12-months.

A difference in control was highlighted by participants dependent on the intensity of their episodes of emotionalism or circumstances where they described a variability in the ability to control.

> *"it depends how big the cry is…I have tried but it doesn't always help", 020, two-weeks.*

> *"I would say maybe it depends on the circumstances", 043, 12-months.*

d. External triggers

Participants described a range of triggers which occurred due to environmental stimuli including; seeing emotional content on the television, others leaving and talking about the stroke.

> *"comes on more of when people are going away or leaving me that's it mostly friends or family it just seems to be when people are going away", 018, two-weeks*

> *"if I hear anything sad that is definitely a trigger for me I become emotional and I feel for other people", 06, two-weeks.*

> *"if I watch something on the telly that is sad I can feel myself welling up", 012, six-months.*

> *"it is always someone that triggers it", 060, six-months.*

> *"talking about the stroke because it is hard to talk about…I know it seems so stupid to some people", 049, 12-months.*

e. Internal triggers

A variety of internal triggers were highlighted by participants including; thoughts, being reminded of losses and remembering past memories.

> *"start thinking about things that have happened to you in the past", 052, two-weeks*

"I can be sitting contemplating and thinking", 063, six-months

"when I am sitting thinking about things and it comes on", 012, six-months.

"I was just thinking of my husband at the time which normally I can think and I am ok", 010, 12-months

f. Predictability

Within this sub-theme participants most frequently reported the occurrence of a prior warning to the onset of episodes of emotionalism, the episodes being re-occurring or noticing a pattern.

"I have warning", 044, two-weeks.

"I got myself a bit better and then it goes back again it comes back again as quick as it goes away", 068, two-weeks.

"I can wake up and I can know I am going to have a bad day", 041, 12-months.

"I would say there is a pattern…because I notice a pattern, 053, 12-months.

g. Unpredictability

The nature of emotionalism was described by participants as unpredictable and captured the element of surprise and no prior warning before the emergence of episodes of emotionalism.

"just like the stroke no warning", 039, two-weeks.

"you are never expecting it", 052, two-weeks.

"it just comes on when I am not expecting it", 057, two-weeks.

"I just find myself surprised that I have tears rolling down my face", 011, 12-months.

Master theme two: 'Ways of coping'

This theme consists of six sub-themes: *acceptance, avoidance of triggers, concealing emotionalism from others, behavioural attempts at symptom management, cognitive attempts at symptom management and support from others* (Figure 3). '*Ways of coping*' incorporates the diverse ways participants described managing episodes of emotionalism as well as help from others.

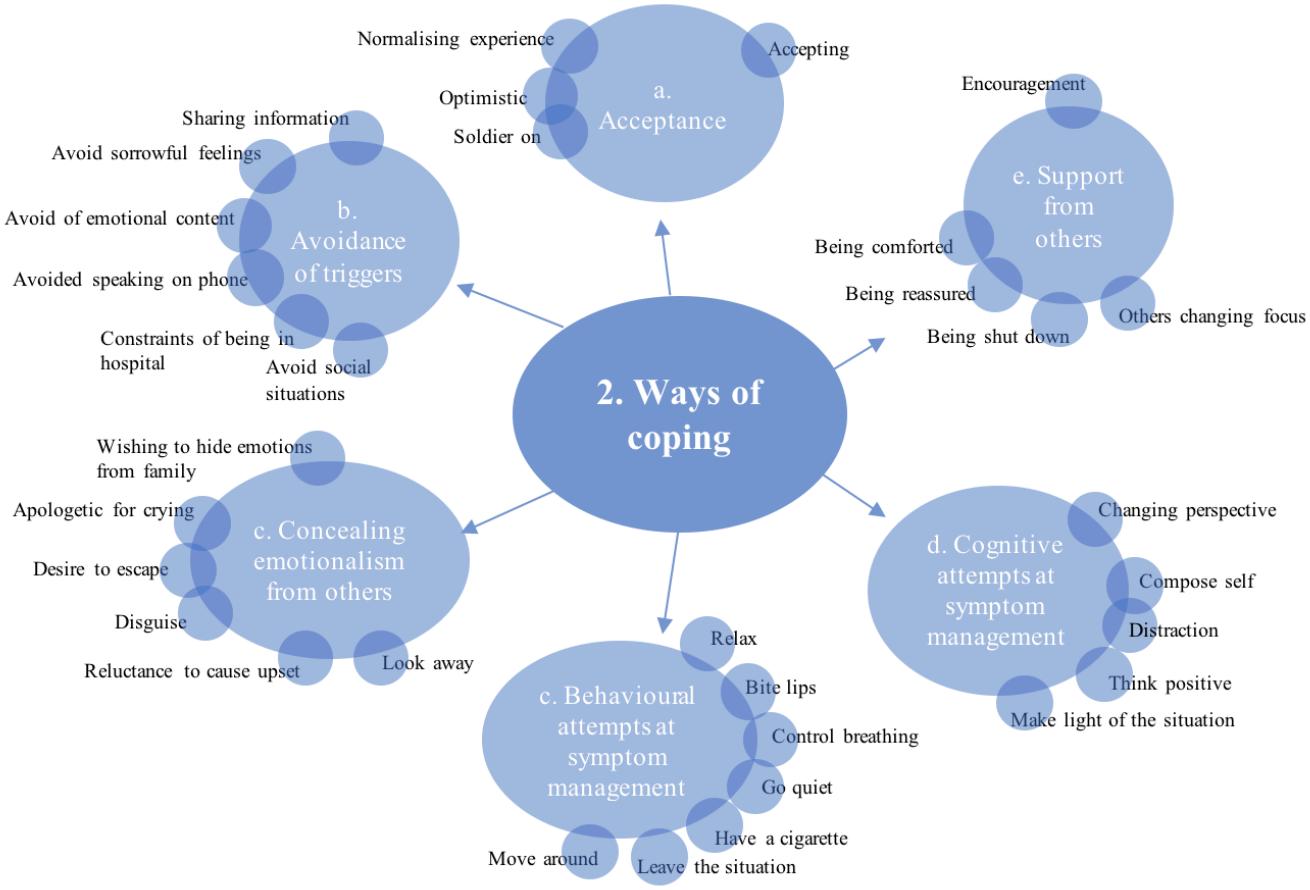

Figure 3. Codes and sub-themes within the 'Ways of coping' theme.

a. Acceptance

This sub-theme highlighted participants' recognition of episodes of emotionalism without attempting to change these.

> *"I just assume it is all part and parcel of what is happening I just assume that is pretty normal", 054, two-weeks.*

"I just let them happen, if they are going to happen they are going to happen", 018, two-weeks.

"I just accept it now", 030, 12-months.

Additionally, participants frequently reported attempts to normalise their experience of emotionalism and at times highlighted the benefits of not challenging them.

"just like my limbs aren't back to normal my emotions aren't back to normal", 05, two-weeks.

"I think bringing it out in the open if anything is beneficial to me", 06, two-weeks.

"I just let it out it's better out", 07, 12-months.

"I kind of expected that see just after the stroke…everyone is different", 025, 12-months.

Participants described optimism regarding their emotionalism in the future and remaining hopeful that these will improve over time.

"I know it is going to be alright", 034, two-weeks.

"I think in time I hope that will change back…as time goes on everything is getting better slowly", 052, six-months.

b. Avoidance of triggers

Within this sub-theme participants frequently reported avoiding specific triggers as an attempt to reduce the onset of episodes of emotionalism such as by avoiding sharing information with others regarding their feelings or certain topics.

"when I speak to people I try and stay away from certain subjects", 05, two-weeks.

"I wasn't telling them how I felt", 040, two-weeks.

Additionally, participants described avoiding emotional content that they were aware could trigger their episodes of emotionalism such as films, news, TV programmes or speaking on the phone.

"I would get rid of it if it was something on the telly I would get rid of it", 035, two-weeks.

"She was just telling me something sad and I just had to put my hand up", 029, six-months.

"I have stopped reading papers I don't watch the news something that comes on there that is sad I just burst into tears it's awful", 04, 12-months.

"I don't want to talk to anyone on the phone…sometimes when my family want to speak to me I say I will call them back I put it off as long as possible", 016, six-months.

Avoidance of social situations was also highlighted by participants which was a change from before they had the stroke.

"I used to always talk to people on the buses but now I don't I just sit ignore everything", 04, 12-months.

"it's avoiding everything that includes family and everything because I don't want them to see", 053, 12-months.

c. Concealing emotionalism from others

Participants described an attempt to disguise their emotions or hide their emotions from their family when they experience episodes of emotionalism.

"with a hankie pretending to blow my nose or cough", 071, two-weeks.

"I would just walk away", 076, two-weeks.

"if I have something in my hands I will be using that", 029, six-months.

"I try hide it from the wee one I don't like it my boy seeing me…I don't like my Mrs see me", 027, six-months.

"I lock myself in the toilet until I am finished", 047, 12-months.

"I go in the kitchen and take yourself away from it for a wee bit", 053, 12-months.

"if the kids are in and that is happening they will say are you ok I will say aye I have just got a runny nose and a cold", 03, 12-months.

d. Behavioural attempts at symptom management

Participants reported a variety of behavioural strategies that were described as helpful to manage episodes of emotionalism that included breathing and looking away to allow time to pass before returning.

"I breathe in and breathe out…give myself some bursts of breathing in and out", 016, six-months.

"I just have to do some deep breathing and just clear my mind", 011, 12-months.

"looking away helps when people are asking me things", 047, 12-months.

"I just try and do something completely different", 051, six-months.

e. Cognitive attempts at symptom management

Cognitive strategies incorporated helpful techniques reported by participants that changed their perspective and could be used discreetly without the awareness of others.

"think of something else you take your mind of it", 041, two-weeks.

"put things into perspective", 054, two-weeks.

"try and use my brain and think why am I doing this, I don't need to do it", 040, two-weeks.

"I tell myself to stop keep saying stop crying stop crying", 012, six-months.

"I will go oh pull yourself together", 031, six-months.

"I try and distract myself", 047, 12-months.

f. Support from others

Participants frequently reported others' support as helpful when they experienced episodes of emotionalism such as being comforted, reassured, others showing encouragement and emotional assistance. These appeared to assist with reducing participants' level of distress.

"he just cuddled me because that is what I felt I needed", 063, six-months.

"give me a hug and it is fine", 011, 12-months.

"she comes and wipes the tears off my face and cuddles into me", 027, two-weeks.

"they tend to reassure me all the time…they keep saying it is going to be alright", 01, two-weeks.

"put my mind to ease when they tell me not to worry about these things", 054, two-weeks.

"they come down all the time and they come and see if I am ok", 031, two-weeks.

"I am lucky that I have got friends I just need to phone them and they come up", 042, six-months.

Master theme three: 'Impact'

This theme consists of four sub-themes: *changes to self, emotional impact, trying to make sense and uncertainty* (Figure 4). '*Impact*' highlights participants' experience of adjusting to a new experience in terms of their emotional response and how emotionalism affected their sense of identity and their views of the future.

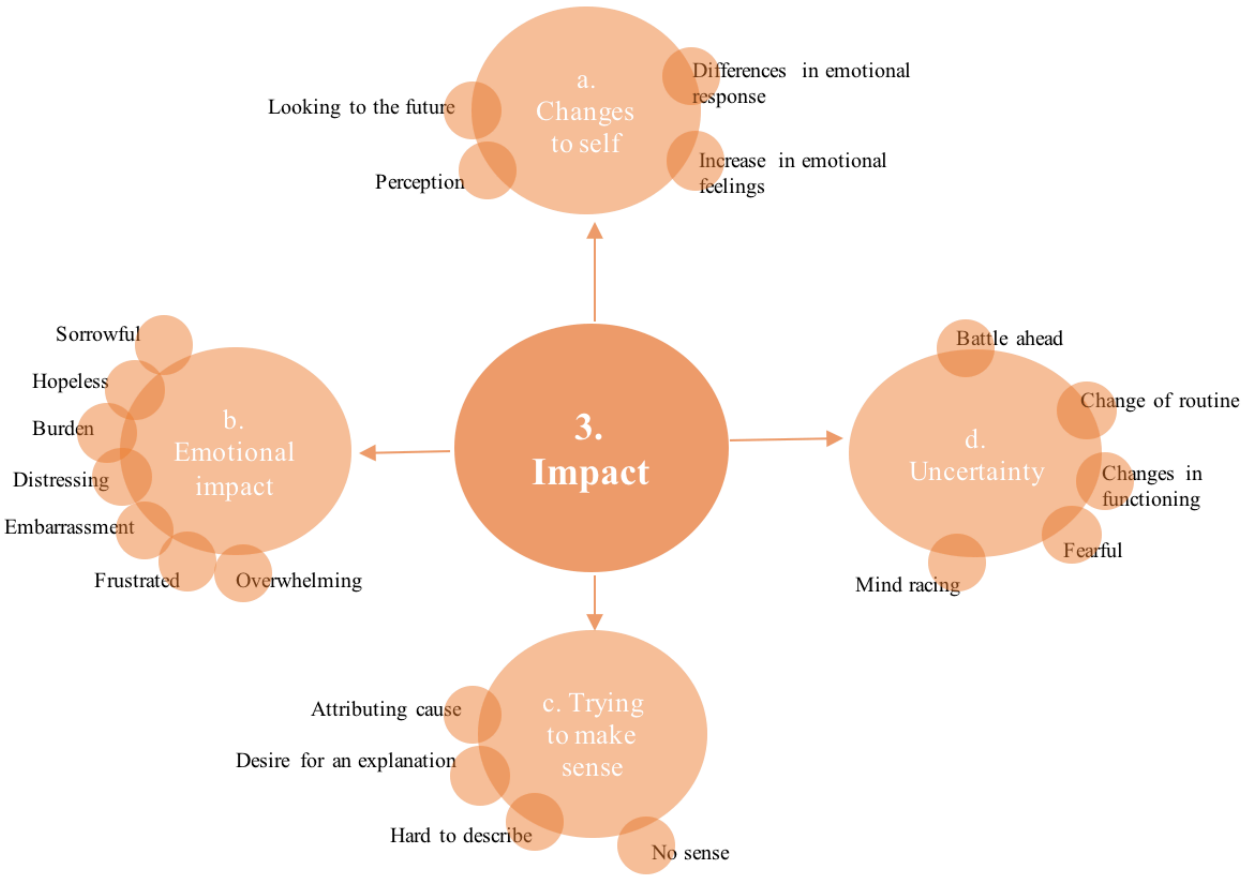

Figure 4. Codes and sub-themes within the 'Impact' theme.

a. Changes to self

Participants highlighted episodes of emotionalism as having been a new experience since the stroke with an emphasis on responding differently in emotional terms compared to the way they were before the stroke.

> *"I was battling something that wasn't very important and I would be very upset and crying, where before it would not bother me and I would answer back and stand up for myself more now, I am not", 020, two-weeks.*
>
> *"everything has changed", 031, six-months.*
>
> *"it's not how I would have dealt with it before I would have probably shouted and got it off my chest but now I just find myself…just tears running down my face quietly crying and that definitely isn't me", 011, 12-months.*

"before the stroke I was quite emotionless", 047, 12-months.

Participants emphasised certain perceptions/beliefs about how they viewed themselves and stereotypical views of displays of emotions. Participants described a process of mean making where some seemed to struggle to make sense of their emotional experience which is at odds with social attitudes they subscribe to or at odds with their own pre-injury relationship to emotion.

"to me I am broken", 076, two-weeks.

"I was a tough cookie I don't mean that dead hard or cold but I can feel the difference", 014, two-weeks.

"I usually am a tough sort of person", 04, six-months.

"I'm just totally different I was always happy easy go lucky person", 042, six-months.

"I am quite an old school kind of guy… men don't cry", 027, 12-months.

b. Emotional impact

Participants highlighted a range of feelings that were caused by experiencing episodes of emotionalism. The most frequently reported increase in emotions were distress and embarrassment which was emphasised by feeling foolish.

"I find it distressing I shouldn't be crying", 021, 12-months.

"I find it embarrassing very because I don't like people seeing me like that you know", 049, 12-months.

"you feel like a complete total idiot to be honest", 048, two-weeks.

"because I go down the shops and I start crying and everyone is looking at you as if you are daft and I begin to agree with them", 03, 12-months.

c. Trying to make sense

As a result of the episodes of emotionalism following their stroke, participants stressed the importance of '*trying to make sense*' of their experience by searching for a cause.

> *"I need explanations…I need a reason for everything", 076, two-weeks.*
>
> *"I'm just been worried about it and hoping to get some answers", 038, two-weeks.*

Participants emphasised the difficulty in explaining their episodes of emotionalism to others, which seemed to be complicated by the issue that participants felt they have not got the answers.

> *"very upsetting… you can't explain it to somebody why you are crying", 020, two-weeks.*
>
> *"I don't know what to call what it is…as I don't know I can't explain why", 0425, two-weeks.*

Additionally, participants frequently reported that their episodes of emotionalism did not make sense.

> *"well I have wondered what this has been about all this cos I don't get it", 035, two-weeks.*
>
> *"they don't make sense it doesn't make sense I was feeling ok I was just watching the film with the kids and the next minute the tears were just flowing and I was like huh?", 03, 12-months.*

d. Uncertainty

The sub-theme '*uncertainty*' captured participants' concerns about the future in terms of feeling fearful and their descriptions of a battle ahead.

> *"I think the fear of the unknown", 07, two-weeks.*
>
> *"because I don't want it happening outside or anything", 025, two-weeks.*

"it's just an uphill battle that I have got in front of me", 039, two-weeks.

Participants frequently reported racing thoughts related to the past, current and future.

"I maybe just thinking what is going to happen, what can happen, what has happened", 028, two-weeks.

"your imagination is running away and it does but when you are lying in here on your own", 07, six-months.

Research question two: Are there differences in how individuals with PSE describe their experience of emotionalism over time?

A recurrent cross-sectional analysis was completed to explore differences between time-points in how participants described their experiences of emotionalism over time. An overview of changes in the frequencies of codes overall for each sub-theme was produced (Appendix P). Matrices were completed to explore the frequencies of codes within each sub-theme and these will be discussed in relation to each major theme.

Master Theme: 'In the moment'

Within '*barriers to control*' the code '*difficult to regulate*' increased to 60% of the sample at 12-months from 21% at two-weeks. However, other codes such as '*irreversible*' decreased to 8% of the sample at 12-months. An increase was observed over time-points for the code '*restrain*' within '*control*' from 23% at two-weeks to 43% at 12-months. At the two-week time-point codes related to being in hospital for '*external triggers*' such as '*others leaving*' were only reported at this time-point and interestingly within the '*predictability*' sub-theme the frequency of '*prior warning*' increased to 43% by 12-months (Table 4). This potentially indicates that participants learn to anticipate the onset of emotionalism over time.

Table 4. Changes in the frequency of codes within '*In the moment*' theme.

Sub-themes	Codes	2-weeks N=52 n, % of sample	6-months N=25 n, % of sample	12-months N=23 n, % of sample
a. Characteristics	*Build-up of emotions*	7, 13%	6, 24%	3, 13%
	Fleeting	24, 46%	7, 28%	12, 52%
	Flood of emotions	8, 15%		6, 26%
	Incongruent with feelings	12, 23%	5, 20%	10, 43%
	Sudden onset	37, 71%	15, 60%	12, 52%
b. Control	*Control after release of emotions*	6, 11%	3, 12%	2, 8%
	Prevent escalation	2, 3%		
	Restrain	12, 23%	10, 40%	10, 43%
c. Barriers to control	*Difficult to regulate*	11, 21%	6, 24%	14, 60%
	Irreversible	20, 38%	1, 4%	2, 8%
	Never had control	20, 38%		
	Out of the blue	17, 32%	10, 40%	10, 43%
	Struggling to cope	6, 11%	6, 24%	
	Unable to compose self	5, 9%		
	Variability of control	4, 7%	3, 12%	1, 4%
d. External triggers	*Being in hospital*	6, 11%		
	Dependent on situations	7, 13%	5, 20%	2, 8%
	Emotional content	12, 23%	11, 44%	6, 26%
	Kind word from others	6, 11%	5, 20%	5, 21%
	Others being unaware	1, 1%		
	Others leaving	6, 11%		
	Others perceptions	3, 5%	2, 8%	1, 4%
	Others responses	8, 15%	3, 12%	1, 4%
	Seeing others	23, 44%	2, 8%	1, 4%
	Separation from family	17, 32%	1, 4%	
	Sharing information	2, 3%		
	Talking about the stroke	7, 13%	2, 8%	5, 21%
	Talking to family	15, 28%	1, 4%	2, 8%
e. Internal triggers	*Feeling sorrowful*	14, 26%	5, 20%	5, 21%
	More time to think	7, 13%	2, 8%	
	Reminiscent of losses	8, 15%	3, 12%	5, 21%
	Thoughts	7, 13%	3, 12%	2, 8%
	Thinking about the past	6, 30%	3, 12%	5, 21%
f. Predictability	*Pattern*	1, 1%	2, 8%	3, 13%
	Prior warning	8, 15%	1, 4%	10, 43%
	Re-occurring	4, 7%	4, 16%	1, 4%
g. unpredictability	*No warning*	20, 38%	7, 28%	7, 30%
	Surprised	5, 9%	3, 12%	2, 8%

Master Theme: 'Ways of coping'

Within the sub-theme of '*Acceptance*' the code of '*soldier on*' was only reported at two-weeks with a decrease in the frequencies of 'optimistic' as well as a slight increase in '*normalising experience*' and '*let it go*' over the time-points. The largest increase within '*avoidance*' was noted for '*avoid social situations'*, which increased to 69% of the sample at 12-months. This potentially suggests increases in displays of avoidance over time. Furthermore, '*wishing to hide emotions from family*' showed an increase to 52% by 12-

months compared to 11% at two-weeks and the frequency of '*leave the situation*' increased to 52% at 12-months. Within '*cognitive attempts at symptom management*', codes such as '*make light of the situation*' and '*being shut down*' were only reported at two-weeks (Table 5).

Table 5. Changes in the frequency of codes within 'Ways of coping' theme.

Sub-themes	Codes	2-weeks N=52 n, % of sample	6-months N=25 n, % of sample	12-months N=23 n, % of sample
a. Acceptance	*Accepting*	21, 40%	8, 32%	12, 52%
	Normalising experience	3, 5%	1, 4%	2, 8%
	Optimistic	4, 7%	1, 4%	
	Solider on	2, 3%		
b. Avoidance of triggers	*Avoid sorrowful feelings*	4, 7%	1, 4%	1, 4%
	Avoid emotional content	7, 13%	3, 12%	5, 21%
	Avoided speaking on phone	4, 7%	3, 12%	2, 8%
	Constraints of being in hospital	7, 13%	1, 4%	
	Going out alone			2, 8%
	Sharing information	8, 15%		3, 13%
	Avoid social situations	8, 15%	8,32%	16, 69%
c. Concealing emotionalism from others	*Apologetic for crying*	2, 3%		1, 4%
	Desire to escape	3, 5%		
	Disguise	11, 21%	5, 20%	6, 26%
	Reluctance to cause upset	7, 13%		2, 8%
	Wishing to hide emotions from family	6, 11%	5, 20%	12, 52%
	Leave the situation	5, 9%	9, 36%	12, 52%
d. Behavioural attempts at symptom management	*Bite lip*	1, 1%		
	Control breathing	11, 21%	6, 24%	8, 34%
	Go quiet	1, 1%		2, 8%
	Have a cigarette	1, 1%		
	Look away	12, 23%	4, 16%	8, 34%
	Move around	1, 1%		
	Relax	4, 7%	4, 16%	
e. Cognitive attempts at symptom management	*Changing perspective*	12, 23%		6, 26%
	Compose self	16, 30%	9, 36%	7, 30%
	Distraction	5, 9%	9, 36%	4, 17%
	Make light of the situation	1, 1%		
	Think positive	7, 13%	3, 12%	3, 13%
f. Support from others	*Being comforted*	18, 34%	2, 8%	3, 13%
	Being reassured	8, 15%		2, 8%
	Being shut down	1, 1%		
	Others changing focus	2, 3%		3, 13%
	Encouragement	11, 21%	5, 20%	5, 21%

Master Theme: 'Impact'

The sub-theme '*changes to self*' showed increased frequencies of codes such as '*increase in emotional feelings*' and '*differences in emotional response*' by 12-months. Within '*emotional impact*' an increase to 65% at 12-months was noted for '*embarrassment*' compared with 40% of the sample at two-weeks. Additionally, increases were reported for

'*distressing*' and '*embarrassment*' codes. Only '*hoping for an explanation*' was recorded at two-weeks and making '*no sense*' showed an increase to 47% of the sample at 12-months. The sub-theme of '*uncertainty*' highlighted two unique codes of '*battle ahead*' and '*changes of routine*' that were only reported at the two-week time-point (Table 6). This could indicate the emotional consequences of emotionalism such as distress, embarrassment and sense of difference in self increase over time.

Table 6. Changes in the frequency of codes within 'Impact' theme.

Sub-themes	Codes	2-weeks N=52 n, % of sample	6-months N=25 n, % of sample	12-months N=23 n, % of sample
a. Changes to self	*Differences in emotional response*	33, 63%	8, 32%	19, 82%
	Increase in emotional feelings	13, 25%	13, 25%	16, 69%
	Looking to the future	3, 5%	9, 36%	2, 8%
	Perception	21, 40%		7, 30%
b. Emotional impact	*Burden*	1, 2%	1, 4%	
	Distressing	15, 28%	9, 36%	12, 52%
	Embarrassment	21, 40%	15, 60%	15, 65%
	Frustrated	13, 25%	7, 28%	5, 21%
	Hopeless	7, 13%	3, 12%	3, 13%
	Overwhelming	7, 13%	6, 24%	6, 26%
	Sorrowful	3, 5%	5, 20%	
c. Trying to make sense	*Attributing cause*	1, 1%		1, 4%
	Desire for an explanation	12, 23%	6, 24%	1, 4%
	Hard to describe	1, 1%		3, 13%
	No sense	5, 9%	9, 36%	11, 47%
d. Uncertainty	*Battle ahead*	18, 34%	2, 8%	
	Change of routine	8, 15%		
	Changes in functioning	1, 1%		3, 13%
	Fearful	2, 3%		3, 13%
	Mind racing	11, 21%	5, 20%	2, 8%

Research question three: How do themes and changes in themes identified from questions one and two relate in contributing to a psychological account of the common mechanisms and experience of PSE over time?

A diagram has been produced to visually propose a model/framework to account for the experiences and common mechanisms of PSE described by participants included in this research project (Figure 5). This model is solely based on participants responses to the open questions in the interview and the qualitative content analysis and recurrent cross-sectional analysis.

This model/framework indicates the onset of episodes of emotionalism as described by participants being triggered by internal or external triggers, which appeared to be considered as 'emotionally moving' and differed from how they might have reacted pre-stroke. This then led to key elements such as the predictability or unpredictability of the episodes and their perception of self-control.

Following the onset of episodes of emotionalism participants moved into ways of coping. If participants described an ability to control episodes, they engaged in '*in-the-moment*' coping behavioural or cognitive strategies. However, participants who felt they had no control or experienced barriers to control described engaging in longer-term coping strategies such as avoidance and concealment. These could be hypothesised as mechanisms which keep PSE going over time and could be reversible maintaining factors targeted through psychological interventions.

Longer-term consequences of emotionalism such as distress and embarrassment could be proposed as contributing to episodes re-occurring and reinforcing further avoidance and/or concealment of emotionalism from others. Acceptance was highlighted by participants as a helpful coping strategy such as normalising their experience and could be hypothesised as a mechanism which helps with assisting recovery from PSE.

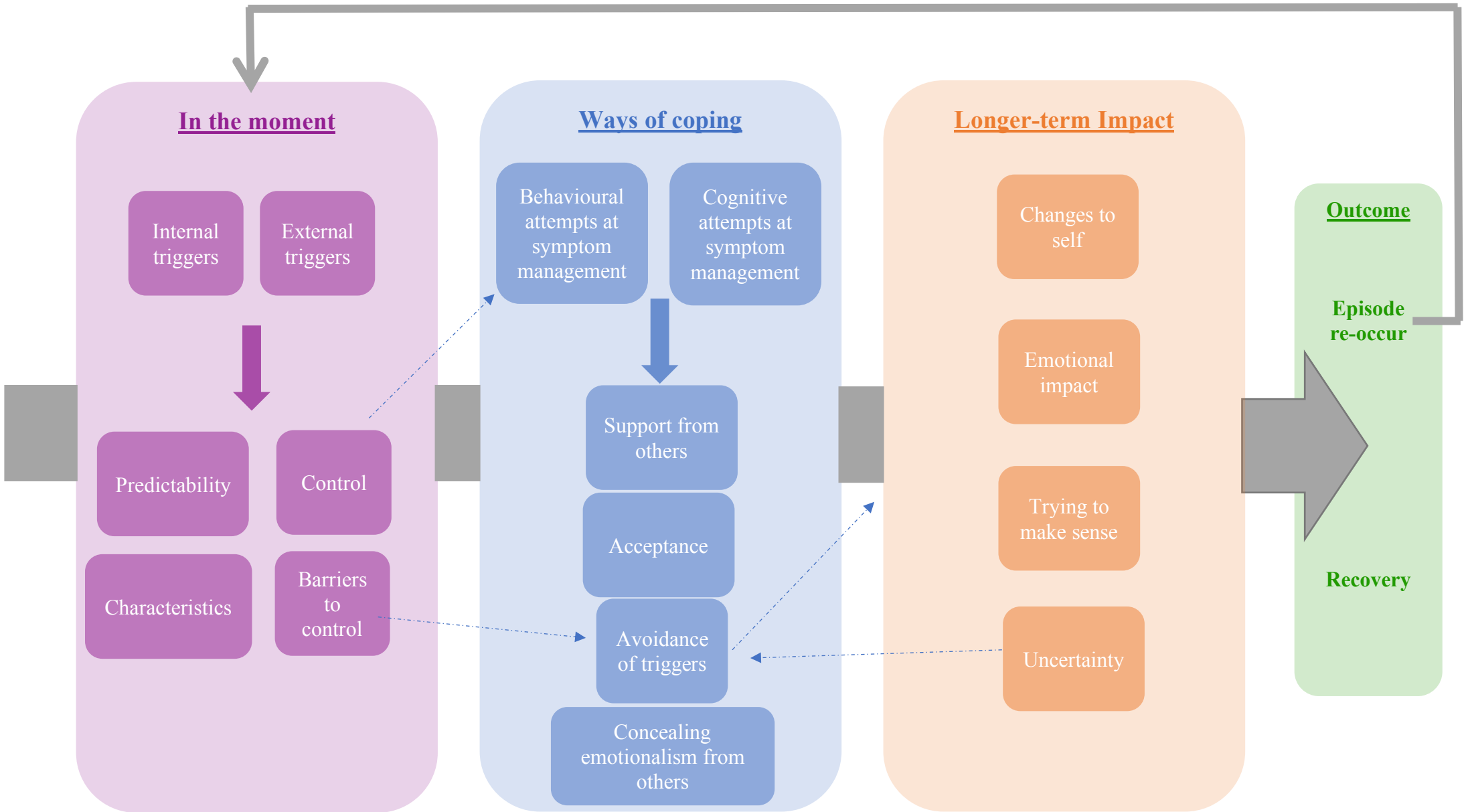

Figure 5. Proposed model of experiences of post-stroke emotionalism (PSE) based on qualitative content analysis and recurrent cross-sectional analysis of open-ended questions.

**Blue dashed lines indicate tentative links between experiences of PSE*

Discussion

To date this is the only study that has explored participants' experience of emotionalism, ways of coping and how their experience of emotionalism may change over time using a longitudinal qualitative approach. Overall three major themes were identified: '*In the moment*' included characteristics, triggers and aspects of control; '*Ways of coping*' highlighted a variation in participants' strategies such as avoidance or acceptance; and '*Impac*t' emphasised longer-term effects of PSE. Analysis of the frequencies of themes across time-points revealed increases in normalising of the PSE experience, withdrawing from social interactions, negative perceptions of emotionalism, episodes of emotionalism making no sense and heightened emotional consequences such as distress or embarrassment. Over time some participants described an increased awareness of triggers for episodes of emotionalism which highlighted an element of predictability which may indicate that psychologically there are aspects that enable people to develop better anticipation and adaptive coping strategies.

Strengths and Limitations of the study

A key strength of this study was the use of a longitudinal qualitative approach. This has allowed for a rich in-depth exploration of participants' experiences as well as exploring differences over time. The analysis was summarised into a framework/model which could help inform clinical practice and shape future psychological formulations of PSE. Furthermore, this study had a large sample size of N = 100 participants with PSE, which allowed for multiple experiences of PSE to be explored in the analysis of the interview data.

A limitation of this study was that the interview schedule was structured so that no interview data could be explored for those who had recovered from emotionalism. To increase the richness of the analysis and add to a proposed model/framework of PSE, it

would have been helpful to ask questions to participants who had recovered from emotionalism at a subsequent time-point. Further questions for example 'what do you feel helped contribute to your recovery of PSE?' could have been added to the interview schedule. Additionally, participants who did not have emotionalism were not asked any further questions across the time-points which meant during analysis the experience of participants with emotionalism were not able to be compared to those without.

The participants in this study were recruited from the West of Scotland and the sample had generally low NIHSS/Barthel scores which highlights potential limitations regarding the generalisability of the findings to individuals for example with aphasia and emotionalism. Furthermore, previous research has diagnosed participants with PSE using the PLACS (Robinson et al., 1993) or CNS-LS (Moore et al., 1997) which has limitations. However, in this study a new measure of the TEARS-IV was used based on House's criteria (House et al., 1989) and is now validated, showing high internal consistency and diagnostic accuracy of tearful episodes (Broomfield et al., 2020).

A further limitation was the researchers attempt to draw conclusions from the recurrent cross-sectional analysis in exploring research question two of changes in participants experience of emotionalism across time. Confounding variables, chance differences due to information coming from open-ended interview questions, 'new-onset' cases of PSE identified at subsequent time-points and relatively small samples at each time-point could impact and influence the changes identified over time. One approach to address this could have been to complete statistical analyses such as Chi-Square or Fishers exact test to explore changes in the frequencies of codes over time.

Some caution should be taken when interpreting these findings based on the researcher's epistemological perspective. The experiences of participants were understood within a critical realist perspective and through the lens of social processes which

acknowledges the findings and interpretations of meanings whilst relate to an underlying 'reality' are clouded by social processes and are a specific context/version of this reality.

When completing qualitative analysis, ensuring codes are reviewed by a second-coder is seen as good practice to increase transparency, promote reflexivity and ensure a systematic approach to coding (O'Connor & Joffe, 2020). In line with a critical realist approach 25% of codes were reviewed by a second independent coder (trainee clinical psychologist) to increase the 'trustworthiness' of the findings (Lincoln & Guba, 1985). Furthermore, triangulation in qualitative research helps to develop further understanding of a phenomenon (Patton, 2002). Investigator triangulation was completed through the use of a second coder, however this study could have triangulated data sources further by interviewing partners or carers to explore their views to support participants' experiences. This could have provided further information regarding changes and ways of coping if participants felt embarrassed or lacked insight into their experience of PSE.

Previous Research

Previous research exploring individuals' experience of emotionalism following a stroke highlighted those with negative experiences reported greater disability and avoidance as well as embarrassment and that social withdrawal had a negative effect on individuals' quality of life (McAleese et al., 2019). The current study found participants described avoidance, heightened emotional consequences and negatively held beliefs about episodes of emotionalism which appeared to maintain PSE. These findings support the limited qualitative research to date and provides further evidence of ways of coping, beliefs held by individuals and changes over time which is important to shape future psychological interventions.

Interpretation of the study findings

Participants reported a range of internal triggers: thinking about the past, memories, thoughts, feeling sorrowful, reminiscent of losses and external triggers: others leaving, emotional content on the television, seeing family, talking about the stroke, others' perceptions, others being unaware and separation from family which led to episodes of emotionalism. Many of the triggers could be objectively considered potentially 'moving' and participant themselves highlighted how their responses differed from how they might have reacted pre-stroke. A kind word from others was frequently reported by participants as a trigger and this increased in frequency over time. Compassion from others was highlighted as both intensifying episodes of emotionalism whilst also described as helpful to feel reassured and comforted. Insight into triggers for emotionalism is valuable to gain further understanding and awareness, which is helpful for clinicians in clinical practice when offering support to individuals with emotionalism. This could potentially help individuals to anticipate episodes of emotionalism and adapt to these psychologically.

The ability to control episodes of emotionalism varied for participants as well as changing over time for some participants. Locus of control refers to an individual's belief about the ability to change a situation (Rotter, 1954). Some participants described the ability to control an episode of emotionalism at the onset, which then allowed them to engage in coping strategies. However, participants who highlighted barriers to control emphasised the difficulty in finding helpful ways to reduce the emotional intensity. This highlights key information regarding how the ability to control can influence an individual's perception and action, which would be important to incorporate in future psychological interventions.

Within the theme '*Ways of coping*', avoidance was common across all time-points. Participants frequently reported withdrawing from social interactions, sharing limited

information with others and avoidance of emotional content that could trigger episodes of emotionalism. This supports previous research which has found emotionalism can cause distress, embarrassment and social avoidance (Allman et al., 1992). Participants highlighted the distress caused by episodes of emotionalism at the onset as well as over time. Calvert et al. (1998) found ideas of reference and emotionalism were associated with embarrassment potentially interacting in this relationship. In the current study participants highlighted high levels of embarrassment caused by episodes of emotionalism and described how these shaped beliefs individuals held about their experience. This emphasises the interaction of levels of distress, embarrassment, avoidance and beliefs about emotionalism whereby earlier detection and intervention could support to break this cycle.

Some participants emphasised the process of acceptance through normalising their experience, remaining optimistic and refraining from attempting to change their emotional response. Acceptance of oneself as intrinsically worthy is central to achieving psychological adjustment following a disability according to the theory of acceptance of disability (Dembo et al., 1956). Additionally, the final stage within a model of adjustment following a stroke highlights the process of acceptance and adjustment of a new reality (Wade et al., 1985). This current study supports these models whereby some participants reflected on adjustment and normalising their experience. Adjustment to PSE could to some degree be considered alongside adjustment to other consequences of stroke as highlighted in models of adjustment following a disability and Brennan's (2001) adapted Social Cognitive Transition Model (Taylor et al., 2011).

Additionally, participants frequently reported the longer-term impact of emotionalism in relation to changes to self and beliefs they held about their experience. A comparison of the self pre-stroke and post-stroke was emphasised by many participants

accompanied with feelings of frustration, distress and hopelessness. Dependency and lack of control following a stroke has been found to contribute to a significant decline in individuals' self-concept (Ellis-Hill & Horn, 2000). Overall, this current study highlighted participants with a negative experience of emotionalism described avoidance, unhelpful beliefs and a lack of acceptance. These components are important and could be targeted by offering psychological interventions such as CBT or third wave Acceptance and Commitment Therapy (ACT) with individuals with emotionalism (Kangas & McDonald, 2011). Both of these therapeutic approaches require an engagement with the current potentially distressing reality as a first step and could potentially help to increase insight into triggers of episodes of emotionalism, acceptance of one's emotional experience, reduce avoidance and help to develop a more compassionate relationship with their experience in the long-term (Hayes et al., 2006). Further research to build on the present findings will be needed before any potential candidate treatment model can be tested to determine feasibility, efficacy and acceptability to patients.

Longer-term consequences were frequently reported by participants trying to make sense of their experience. In the acute phase participants described a search for an explanation for their episodes of emotionalism and finding it hard to describe to others. Worry and fears for the future were frequently reported with this seen to exacerbate feelings of distress. Uncertainty plays an important role in the rehabilitation of individuals following a stroke (Ni et al., 2018). Worry and uncertainty could be seen to feedback into avoidance in participants, which then limits an individual's opportunity to challenge held beliefs and the consequences of emotionalism. This highlights the requirement for psycho-education regarding emotionalism to be offered in clinical practice to individuals which could help to reduce uncertainty and provide understanding of this condition.

Clinical Implications

Previous research has revealed emotionalism is under-recognised and under-diagnosed. The current study highlights the clear importance of earlier detection and identification of emotionalism, which could help to support individuals in terms of adaptive coping strategies and challenging negative beliefs. The latest development of a new measure of emotionalism following stroke Testing Emotionalism After Recent Stroke – Questionnaire (TEARS-Q; Broomfield et al., 2020) has shown high internal consistency and diagnostic accuracy of tearful episodes. This means that clinicians can accurately screen all patients following a stroke as part of a standard battery of assessments with the two-item screen and/or the full eight item TEARS-Q to assist with earlier detection.

Secondly, this study was the first to explore experiences of emotionalism over time using longitudinal qualitative methods, which helps to shape psychological formulation and understanding of PSE. This research has highlighted that emotionalism is not simply random and out of the blue which means that PSE is potentially psychologically understandable. Participants with negative experiences of emotionalism described high levels of avoidance, negative perceptions, lack of understanding of the condition, distress and embarrassment. This was more apparent in participants at 12-months and increased over time. Participants who described acceptance of emotionalism described less distress and normalised their experience.

Finally, the findings from this study help to improve our knowledge of potential mechanisms of emotionalism and inform clinical practice whereby earlier detection of distress, embarrassment, avoidance and beliefs about emotionalism as well as interventions targeting these aspects could potentially help to break this cycle. The findings further suggest the relevance of behavioural/psychological interventions which we know clinicians use (Gillespie et al., 2019) but for which there is currently no evidence base. The findings

could shape future psychological interventions highlighting the importance of providing psychoeducation to help provide further understanding to patients and their families to normalise emotionalism.

Recommendations for Future Research

Further research is needed to explore experiences of individuals who have recovered from emotionalism focusing on aspects of what helped, what changed and barriers to recovery. This could help further our understanding of individuals' experience of emotionalism and further highlight individuals' beliefs regarding emotionalism and how this impacts their coping and/or experience. Additionally, further research could test the efficacy of psychological interventions using single-case experimental designs (SCEDs) or randomised controlled trials (RCTs) focused on testing the causal mechanism which tentatively emerged from the analysis such as reducing aspects of avoidance, enhancing acceptance of their experience, learning relaxation/mindfulness strategies such as controlled breathing and providing psycho-education to create greater awareness of triggers for episodes of emotionalism. Potentially, normalisation and immediate coping strategies could lessen the likelihood of individuals experiencing longer-term consequences of emotionalism characterised by avoidance, embarrassment and distress.

Disclosure of interest

There is no conflict of interest. This research was conducted as part of the first authors Doctorate in Clinical Psychology Training at the University of East Anglia (UEA).

Additional information

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Chapter Four: Extended Methodology

Word Count: 3013

Extended methodology

This chapter provides further details in relation to the methodology of the empirical paper. The ontology and epistemology, the rationale regarding the chosen qualitative methods, a comparison of alternative qualitative methods and the qualitative analysis process are presented.

Ontology and Epistemology

Ontology can be defined as the study of the nature of being or reality and epistemology refers to the views on truth and legitimate knowledge (Slevitch, 2011). Broadly, there are three distinct philosophical research paradigms which exist to guide research methods and design: positivism, interpretivism and critical realism (Rehman & Alharthi, 2016). This research took a critical realist approach with this perspective originating as an alternative to positivism and constructivism by distinguishing between what is real and what we know (Denzin & Lincoln, 2011).

Ontology in critical realism stratifies reality into three levels: the 'empirical level' at the top, is our experiences or observations of events that are observable; the 'actual level' refers to events which occur but may or may not be observed; and the 'real level' is regarded as the deepest level of reality comprising mechanisms that have causal power which creates the conditions for higher levels (Jeppesen, 2005). Within the 'empirical level' individuals are able to explain events, however this is always mediated through the filter of human experience and interpretation.

Critical realism proposes that an objective 'real world' exists which has powers and properties that can be uncovered and more accurately known through research (Maxwell, 2012[a]). However, critical realists embrace epistemological relativism whereby they recognise that knowledge is subjective, transitive and constantly changing (Vincent & O'Mahoney, 2018). Researchers can obtain knowledge about reality through different

methods such as interviews with researchers acknowledging that deeper levels of understanding are awaiting discovery. However, this proposed knowledge or theory can be fallible (Sayer, 1999). A central element of critical realism is the concept of transitive knowledge whereby knowledge and theories derived from qualitative analysis may over time be extended, changed or rejected (Haigh et al., 2019). Within this approach the focus is placed on the researchers' attempts to ensure trustworthiness and practical usefulness of the theory generated. A critical realist seeks to discover the deeper causal mechanisms of an experience or event, whereby some degree of interpretation is needed to provide access to the underlying mechanisms of the data, as although the data is informative of reality this does not simply mirror it (Maxwell, 2012[b]).

Researcher perspective

Within qualitative research a researchers' epistemological perspective can shape the research process such as the choice of methodology, research questions and interpretation of results (Jackson, 2013). My ontological and epistemological perspective aligns with a critical realist approach. Within this approach research is concerned with exploring underlying mechanisms which can be seen or unseen to seek explanations of phenomena (Vincent & O'Mahoney, 2018). My philosophical position of a critical realist shaped the research questions for the empirical paper with the main research aim to understand emotionalism and explore underlying mechanisms over time. As a trainee clinical psychologist working in different clinical placements being curious and listening to others' experiences helps to gain an understanding about a phenomenon. In line with critical realism the researcher explored participants' experience of emotionalism to understand and provide possible explanations of the mechanisms of emotionalism over time whilst acknowledging that the explanations drawn can be fallible and open to different explanations.

Rationale for qualitative methodology conducted

Emotionalism is under-researched with only one previous qualitative study exploring individuals' experience of emotionalism. To date there is no proposed psychological model of emotionalism, which highlights the importance of further qualitative studies to increase our understanding and support the development of a psychological model. A qualitative methodology was adopted for the empirical paper which focuses on 'making sense' or understanding a phenomenon (Maxwell, 2012[a]). Critical realism prioritises qualitative research to enable a deeper exploration of individuals' experience and to capture causal mechanisms (Bhaskar, 1975). The empirical paper involved several stages of analysis: qualitative content analysis, framework analysis, recurrent cross-sectional and trajectory longitudinal analysis which align with a critical realist approach.

Content analysis focuses on the characteristics of communication whereby attention is focused on the meaning of what participants have expressed (Tesch, 1990). The aim of content analysis is to provide an understanding of the phenomenon being investigated (Downe-Wamboldt, 1992). Content analysis can be conducted from different epistemological perspectives but this approach was ideal for the empirical paper as conventional qualitative analysis allows the researcher to investigate differences in experiences among participants and to discover relationships between participants (Hsieh & Shannon, 2005). Furthermore, the researcher can explore the frequency of codes as well as interpret the codes and themes, which helps to provide further understanding and comparison of participants' experience and mechanisms (Morgan, 1993). This aligns with a critical realist approach whereby common mechanisms could be identified from participants' experience across the sample which enables a framework/model to be proposed.

Framework analysis and qualitative content analysis can be seen to be grouped in the same broad family of analysis methods. Framework analysis was initially developed to assist with large scale policy research, however this method has been found to be helpful in other areas such as health research (Ritchie & Spencer, 1994). Matrices are created, which allow the researcher to explore similarities and differences across as well as within participants. This approach is not rooted in a particular epistemological perspective, which enables the researcher to adopt this approach for either inductive or deductive analysis (Gale et al., 2013). Large data sets are suited to this method as the matrices enable comparisons of themes and acknowledges the complexity of data and allows a structured systematic approach to be conducted by the researcher (Ritchie et al., 2013).

A recurrent cross-sectional and trajectory qualitative approach can be used in combination with other qualitative approaches to allow researchers to explore participants' experiences longitudinally rather than adopting a cross-sectional approach. A recurrent cross-sectional approach explores the differences between time-points and focuses at group-level data. A trajectory approach investigates how experience changes over time and focuses on an individual participant level (Grossoehme & Lipstein, 2016). Within this empirical paper this allowed a study of individual change (e.g. recovery from or maintenance of PSE) over time as well as looking at relative proportions of different coping styles at each time-point. These approaches are compatible with different philosophical research paradigms and allow researchers to use a combination of different qualitative methods. Adopting a longitudinal approach to analysing qualitative data is not used frequently but can be very useful to explore participants' experience of a phenomenon especially with studies concerned with changes over time and with a clinical issue. This could provide information about possible recovery or deterioration which is highly relevant to clinical model development. This approach was viewed as essential for the empirical

paper to explore the experience of emotionalism over time, which has not been investigated to date.

Critical realist approach

These qualitative methods described above are compatible with critical realism, which enables a researcher to explore how participants understand and give meaning to their experience with the acknowledgment that knowledge is fallible (Sayer, 1999). In line with a critical realist approach to qualitative data analysis, this process is not linear but can involve several steps: induction, deduction, abduction and retroduction to enable exploration of causal mechanisms (Vincent & O'Mahoney, 2018). There is an acknowledgement that research questions and design may be guided by theory or previous research and whilst this can be recognised, critical realists highlight these may not accurately reflect reality and therefore the researcher should treat these as initial theories that can be disproved, amended or changed (Bhaskar, 1979).

As this study was a secondary analysis of pre-collected data, certain aspects were already pre-determined such as the interview time-points and in the TEARS study participants were asked factual-type questions about PSE and how it affects them, but these questions were designed with a diagnostic focus of emotionalism based on psychological constructs and clinical practice. Therefore, this created a slight tension with certain aspects using a deductive approach prior to the qualitative analysis of the interviews whereby existing research, psychological constructs and theory informed data collection and creation of the interview schedule. However, during analysis the researcher adopted a largely inductive approach working exclusively from the participants' experience when deriving the codes and themes, which is in line with critical realism. In inductive analysis, there is acknowledgement that although the findings can be influenced by the research questions, the findings arise directly from the analysis with the researcher

holding back as much as possible from any preconceived ideas or holding theses lightly/flexibly and being transparent throughout the process (Thomas, 2006). However, it cannot be claimed that this was purely an inductive approach as themes can be shaped in part by the researchers' experiences and beliefs, which means that some knowledge of psychological constructs was brought into the research process (Boyatzis, 1998).

Within critical realism qualitative analysis abduction allows the researcher to review and be aware of concepts related to the phenomenon being studied. The researcher produces a re-description of the findings from observations such as interviews into a theory to describe the mechanisms or relationships which helps to explain a phenomenon. The empirical paper adopted a latent approach exploring the underlying meaning of the text (Elo et al., 2014). For this study emotionalism was explored to understand participants' experiences and common mechanisms whereby the researcher completed a process of 're-description' with some acknowledgement of existing psychological constructs to produce a new interpretation/framework of reality which supports a critical realist approach (Danermark et al., 2019).

Alternative qualitative methods

A brief summary of alternative qualitative methods and the strengths and limitations of these approaches are presented. Research questions/aims, epistemology and methods should all be aligned and a rationale is provided for why these alternative methods were not chosen for the empirical paper as either they were not applicable due to the limitations of the nature of the data available or the assumptions about the nature of reality or aims of the analysis.

Grounded theory was briefly considered due to the empirical paper's aim to develop a clinically applicable model. Traditional grounded theory is rooted in positivism whereby a conceptual theory is generated, which is grounded in the data (Glaser & Strauss,

1967). The theory emerges whereby the researcher is objective, detached and neutral. In contrast, constructivist grounded theory highlights the role of the researcher in generating data and theory (Charmaz, 2006). A grounded theory approach is theory-building rather than theory-testing and inductively derived (Strauss & Corbin, 1990). A constant comparative technique is employed whereby each element of the data such as codes and categories are constantly compared with all other parts of the data (Glaser, 2002). This increases the conceptual level of data analysis and begins from the moment analysis has begun.

Theoretical sampling is viewed as an important characteristic of grounded theory. This involves the researcher analysing data and then collecting further data based on the analysis of previous data (Corbin & Strauss, 2014). Theoretical sampling can help to address questions or potential gaps in the data during the analysis process whereby the researcher decides what data to collect next to assist in developing a theory as it emerges. Within this approach rich detailed data can be collected, which allows the researcher to explore the meaning, beliefs and intentions of participants' experience (Charmaz, 2006). However, this approach has been criticised for being time consuming and increases the likelihood of novice researchers making methodological errors (Hussein et al., 2014).

Grounded theory could have been suitable to explore the empirical paper research questions as this approach is suitable for studies exploring individual processes such as personal experience or emotions. It could have helped to reveal detailed accounts of participants experience and help generate a theory of emotionalism. However, as the data was pre-collected the researcher was unable to conduct theoretical sampling or collect data from multiple sources, which would decrease the richness of the findings and limit the generalisability of the results/theory produced.

Interpretative phenomenological analysis (IPA) could have been considered given the researchers' interest in developing a deeper understanding of the experience of PSE. IPA is rooted in phenomenology which aims to generate an understanding of participants' experience in its own terms at a 'deeper level' rather than one that is shaped by pre-existing theories (Smith & Osborn, 2015). This approach emphasises the requirement for researchers to engage in double hermeneutic whereby the participant is attempting to make sense of their experience and the researcher is attempting to make sense of the participants' account of their experience (Smith, 2004). This means that the analysis is dependent on the participants' ability to describe their experience and the researcher's skill in interviewing and then interpreting the data (Noon, 2018). A relatively homogenous purposive sample is sought allowing participants to describe a rich detailed account of their experience (Smith et al., 2009). Given people's own experiences are so unique, the more homogenous the sample the more likely it will be that commonalities in the themes as well as meaningful nuanced differences in experiences can be made.

A strength of IPA is the focus IPA places on how participants perceive and make sense of a phenomena where an open interview is ideal to obtain a very in-depth account of their experience (Smith & Osborn, 2008). This approach is flexible to allow a range of data collection methods such as interviews, diary entries and notes. However, this approach has been criticised for assuming that participants and researchers are effective in communicating with one another the nuances of their experience (Willig, 2008). This could result in a biased sample, which includes only participants that are able to describe and reflect on their experience and limits the involvement of participants with cognitive difficulties.

An IPA approach could have been selected to explore participants' experience of emotionalism as IPA would enable an investigation of how participants make sense of

emotionalism. However, IPA was not chosen for the empirical paper as one of the main research questions was to develop a model/framework that accounted for the differences in experience of emotionalism, which would be incompatible with this approach. The focus of IPA is to look at unique subjective not necessarily generalisable experience whereby the empirical paper aimed to explore the common and generalisable mechanisms to inform broader clinical practice which means that IPA is incompatible with the aims and questions of the study. To have used IPA very rich data and interviews that were heavy with detailed introspective language of participants plus the researcher's own experience of the interview and notes about the participants' behaviour would be needed to drive the deep interpretations.

Qualitative analysis process

A detailed explanation of the step-by-step qualitative analysis process: qualitative content analysis, framework analysis, recurrent cross-sectional and trajectory will be presented. Finally, an appraisal using Lincoln and Guba (1985) 'trustworthiness' of data criterion of the qualitative methods is outlined.

Qualitative content analysis

Each transcript was analysed using qualitative content analysis with NVivo software (QSR International Pty Ltd, 2020) starting with time-point one. The study followed four main stages of qualitative content analysis as outlined by Bengtsson (2016). The interviews were first coded line-by-line to capture the meaning of the text (Appendix Q). Following this the codes were compared with the text to ensure the code captures the meaning. Once the interviews were coded the codes were compiled into themes, which allowed conclusions to be drawn to gain a deeper understanding of participants' experience (Appendix R). This process was repeated across time-points for each interview transcript.

Framework Analysis

Framework analysis allows for a comparison of themes within and between participants through the use of matrices (Ritchie et al., 2013). The next stage of data analysis organised the compiled codes, themes and categories generated from the qualitative content analysis into matrices using framework analysis. The categories and themes which were clearly defined in the previous stage of analysis became a working analytical framework. Each code was assigned a number for identification and annotated onto the transcripts. Matrices were created using NVivo whereby the codes were charted into the matrices. During the indexing stage the developing analytical framework was applied to several transcripts that had certain framework themes (Appendix S). The semi-structured interview questions were also used to shape this framework.

In the final stage of mapping and interpretation, the researcher searched for similar and different experiences among participants where a framework was developed based on emerging themes, which accounted for a variety of experiences.

Recurrent cross-sectional analysis

The next stage of analysis used a recurrent cross-sectional approach, which was described by Grossoehme and Lipstein (2016) where themes are explored across time-points to investigate differences in individuals' experience longitudinally. This approach allowed for an exploration of factors that influence participants' experiences over time and how participants cope with emotionalism from the initial acute phase and into the longer term. Sequential time-ordered matrices were created in a chronological pattern, which allowed the researcher to explore changes in experiences over time. Using the codes and themes identified from the qualitative content analysis, matrices were created with themes along the y-axis and time points across the x-axis (Appendix T).

Trustworthiness of data

The term trustworthiness is the most commonly used criterion to evaluate qualitative analysis developed by Lincoln and Guba (1985). Trustworthiness confirms the findings are 'worthy of attention'. This was extended to include credibility, dependability, conformability and transferability. Clear descriptions of participants included in this study were presented in the empirical paper to increase credibility. Transferability measures if the findings can be generalised and dependability refers to the stability of data over time. This raises challenges with qualitative research whereby the researcher presented 'thick descriptions' and definitions of codes and themes which could be used by other researchers who seek to explore the generalisability and stability of the findings. Finally, to increase conformability an independent second-coder, a trainee clinical psychologist reviewed 20% of the quotes and 25% of the overall codes to ensure congruence between multiple independent reviewers regarding the captured meaning of the codes. Any discrepancies were resolved through discussions whereby codes or descriptions were re-examined.

Chapter Five: Additional Results

Word count: 1363

Overview of chapter

This chapter provides further analysis of the qualitative interview data from the main empirical paper. Firstly, this chapter will present the results from the analysis of clusters of participants followed by an analysis of individual participants over time.

Research question two: Are there differences in how individuals with PSE describe their experience of emotionalism over time?

Further exploration of the qualitative interview data was completed based on certain categories. Participants were clustered into categories in a deductive way based on a priori understanding of clinical characteristics from clinical practice and limited research to date. Participants were clustered in terms of PSE and level of distress or embarrassment, feeling sad/not feeling sad and type of emotional episodes either felt like crying or actual crying. The interview consisted of both open and closed questions where the closed questions were used to cluster participants based on clear categories, which minimised the potential bias and subjectivity of the researcher. Following the completion of coding and qualitative analysis, participants were classified based on these clusters to allow for an exploration of changes over time within sub-themes such as feelings of control, avoidance of triggers and acceptance.

Part a) Clusters

Cluster one: Distress

Participants' level of distress was measured with four options: minimal/not, moderate, very or very severe using their responses from the closed questions of the interview. '*Acceptance*' highlighted higher frequencies of participants who were classed as minimal/not distressed across all time-points and this decreased as

the levels of distress increased. This suggests minimal/not distressed participants were more accepting than those who were very or very severely distressed. Within '*avoidance of triggers*' the frequencies fluctuated according to the levels of distress whereby at two-weeks there were greater numbers of participants both minimally/not and very distressed. However, this could be influenced by the context of being in hospital at the time and this changing the definition of avoidance. At six-months and 12-months there was a greater number of participants who were moderately distressed compared to minimally/not within '*avoidance of triggers'*. The largest increase in frequencies of participants within '*concealing emotionalism from others*' and '*unpredictability*' was highlighted at the 12-month time-point with a higher number of participants categorised as moderately distressed than minimally/not. The sub-theme '*control*' showed variation with higher number of participants with lower levels of distress at two-weeks and the lowest number of participants with very severe levels of distress across all time-points, see Table 1.

Table 1: Exploration of number of participants according to levels of distress over time within sub-themes

Sub-themes	Distress - 2-weeks N=52 (n=, % of sample)				Distress - 6-months N=25 (n=, % of sample)				Distress - 12-months N=23 (n=, % of sample)			
	Minimal / not	Moderate	Very	Very severe	Minimal / not	Moderate	Very	Very severe	Minimal / not	Moderate	Very	Very severe
Acceptance	12 (23%)	4 (7%)	2 (3%)	2 (3%)	4 (16%)	4 (16%)	0 (0%)	0 (0%)	4 (17%)	1 (4%)	3 (13%)	1 (4%)
Avoidance of triggers	9 (17%)	5 (9%)	7 (13%)	1 (1%)	1 (4%)	6 (24%)	3 (12%)	0 (0%)	4 (17%)	7 (30%)	4 (17%)	2 (8%)
Concealing emotionalism from others	9 (17%)	5 (9%)	5 (9%)	1 (1%)	0 (0%)	3 (12%)	1 (4%)	0 (0%)	2 (8%)	9 (39%)	4 (17%)	1 (4%)
Unpredictability	10 (19%)	5 (9%)	6 (11%)	1 (1%)	3 (12%)	4 (16%)	2 (8%)	0 (0%)	3 (13%)	7 (30%)	2 (8%)	0 (0%)
Control	5 (9%)	1 (1%)	2 (3%)	1 (1%)	2 (8%)	3 (12%)	3 (12%)	0 (0%)	5 (21%)	3 (13%)	2 (8%)	1 (4%)

Cluster two: Embarrassment

Participants' level of embarrassment was measured with four options: minimal/not, moderate, very or very severe using their responses from the closed questions of the interview. For '*acceptance*' across two-weeks and six-months a greater number of participants were classed as minimally/not embarrassed and this decreased as the levels of embarrassment increased. However, at 12-months there was a greater number of participants classed as very embarrassed compared to those that were minimally/not embarrassed. Those that were minimally/not and very embarrassed showed similar numbers of participants within '*avoidance of triggers*' sub-theme across time-points, which highlights avoidance across different levels of embarrassment. '*Concealing emotionalism from others*' and '*unpredictability*' showed an increase in the number of participants as the levels of embarrassment increased for the 12-month time-point. Interestingly at two-weeks within the sub-theme '*control*' there was a higher number of participants with lower levels of embarrassment, however at six-months and 12-months there was variation according to the levels of embarrassment, see Table 2.

Table 2: Exploration of number of participants according to levels of embarrassment over time within sub-themes

Sub-themes	Embarrassment - 2-weeks N=52 (n=, % of sample)				Embarrassment - 6-months N=25 (n=, % of sample)				Embarrassment - 12-months N=23 (n=, % of sample)			
	Minimal / not	Moderate	Very	Very severe	Minimal / not	Moderate	Very	Very severe	Minimal / not	Moderate	Very	Very severe
Acceptance	11 (21%)	7 (13%)	4 (7%)	1 (1%)	4 (16%)	3 (12%)	1 (4%)	0 (0%)	4 (17%)	0 (0%)	5 (21%)	0 (0%)
Avoidance of triggers	10 (19%)	5 (9%)	7 (13%)	0 (0%)	2 (8%)	5 (20%)	3 (12%)	0 (0%)	7 (30%)	4 (17%)	6 (26%)	0 (0%)
Concealing emotionalism from others	10 (19%)	5 (9%)	7 (13%)	0 (0%)	0 (0%)	2 (8%)	3 (12%)	0 (0%)	4 (17%)	7 (30%)	5 (21%)	0 (0%)
Unpredictability	9 (17%)	5 (9%)	7 (13%)	2 (3%)	4 (16%)	2 (8%)	4 (16%)	0 (0%)	3 (13%)	5 (21%)	3 (13%)	0 (0%)
Control	6 (11%)	8 (15%)	5 (9%)	0 (0%)	3 (12%)	2 (8%)	4 (16%)	0 (0%)	6 (26%)	2 (8%)	3 (13%)	0 (0%)

Cluster three: Feeling sad at the time

Participants were divided into two groups based on whether they felt sad at the time of their crying episodes. A greater number of participants did feel sad at the time of their crying episodes across all time-points within '*acceptance*', '*avoidance of triggers*', '*unpredictability*' and '*control*' sub-themes. Of note within '*avoidance of triggers*' at 12-months 65% of participants did not feel sad compared with three participants who felt sad. This could indicate the phenomenon of immediate relief in the short-term when participants engage in avoidance but this could reinforce further avoidance. A larger number of participants did not feel sad at two-weeks within the '*concealing emotionalism from others*' sub-theme, see Table 3.

Table 3: Exploration of number of participants according to feelings of sadness over time within sub-themes

	Feelings of sadness - 2-weeks N=52 (n=, % of sample)		Feelings of sadness - 6-months N=25 (n=, % of sample)		Feelings of sadness - 12-months N=23 (n=, % of sample)	
Sub-themes	Did not feel sad	Felt sad	Did not feel sad	Felt sad	Did not feel sad	Felt sad
Acceptance	13 (25%)	10 (19%)	5 (20%)	3 (12%)	7 (30%)	2 (8%)
Avoidance of triggers	14 (26%)	9 (17%)	8 (32%)	3 (12%)	15 (65%)	3 (13%)
Concealing emotionalism from others	8 (15%)	14 (26%)	5 (20%)	0 (0%)	13 (56%)	3 (13%)
Unpredictability	14 (26%)	9 (17%)	9 (36%)	1 (4%)	8 (34%)	3 (13%)
Control	10 (19%)	9 (17%)	10 (40%)	2 (8%)	9 (39%)	2 (8%)

Cluster four: Types of emotional episodes

Participants were classified according to type of crying episode: felt like crying or actually cried. Within all sub-themes there was a greater number of participants who had crying episodes compared with those who only felt like crying across time-points. Interestingly, for both '*acceptance*' and '*avoidance of triggers*' sub-themes there were a similar number of participants who experienced episodes of feeling like crying and actually crying which could suggest that although they were avoidant they also showed some degree of acceptance for their emotional episodes. Additionally, those who only had only felt like crying only showed very low levels of avoidance or acceptance, see Table 4. Overall, it suggests that there was variation in participants experience of emotionalism dependent on the type of emotional episodes they experienced.

Table 4: Exploration of number of participants according to types of episodes over time within sub-themes

	Type of episodes - 2-weeks N=52 (n=, % of sample)		Type of episodes - 6-months N=25 (n=, % of sample)		Type of episodes - 12-months N=23 (n=, % of sample)	
Sub-themes	Felt like crying only	Crying	Felt like crying only	Crying	Felt like crying only	Crying
Acceptance	2 (3%)	21 (40%)	2 (8%)	6 (24%)	0 (0%)	9 (39%)
Avoidance of triggers	2 (3%)	21 (40%)	1 (4%)	10 (40%)	0 (0%)	18 (78%)
Concealing emotionalism from others	1 (1%)	18 (34%)	1 (4%)	4 (16%)	0 (0%)	10 (43%)
Unpredictability	3 (5%)	20 (38%)	1 (4%)	9 (36%)	1 (4%)	10 (43%)
Control	3 (5%)	16 (30%)	1 (4%)	11 (44%)	0 (0%)	11 (47%)

Part b) Trajectory analysis

A trajectory analysis of participants with emotionalism over more than one time-point was completed which focused on changes over time for individual participants (Grossoehme & Lipstein, 2016). This allowed for an exploration of how experiences of emotionalism changed over the recovery period of one year. Individual participants were followed across time-points exploring changes in frequency of codes within sub-themes rather than the whole sample of participants at one time-point. A total of 24 participants who had emotionalism across more than one time-point consented to their interview being recorded. Matrices are presented for each major theme with the sub-themes on the y-axis and time points on the x-axis. Individual participant numbers are plotted across time allowing the reader to explore changes over time for each participant.

Major theme: '*In the moment*'

The largest number of participants reported codes over time within the sub-theme '*characteristics*' with only five participants reporting this at one time-point. This could suggest consistency in the way participants experience the key elements of their emotional episodes. The sub-theme '*control*' highlighted variation over time with six participants reporting codes related to ability to control over time, however twelve participants reported this at only one time-point. Within '*barriers to control*' twelve participants highlighted codes across more than one time-point with seven participants reporting this at one time-point. One participant (031) reported '*barriers to control*' at two-weeks but at six-months emphasised codes related to '*control*' revealing a change in ability to control emotionalism over time. Both '*internal triggers*' and '*external triggers*' revealed participants reported triggers over more than one time-point. Only two participants reported codes

related to the sub-theme '*predictability*' across time with nine participants reporting this at one time-point. Several participants (025, 053, 072) reported codes related to '*unpredictability*' at one-time point but at subsequent time-points reported codes related to '*predictability*', see Table 5.

Table 5: Exploration of frequency sub-themes for each participant within 'In the moment' theme

	Emotionalism across more than one time point (N=24)											
'In the moment' sub-themes	2-weeks				6-months				12-months			
a. Barriers to control	06	09	012	020	01	04	06	**07**	01	04	016	020
	022	025	**031**	035	09	012	016	020	025	027	035	053
	062	**073**	075		027	**042**	053	075	**072**			
b. Characteristics	09	012	020	024	**01**	04	07	09	04	07	**016**	025
	025	031	**035**	044	012	020	024	027	027	042	044	053
	052	**062**	**070**	075	031	042	052	053				
					072	075						
c. Control	**01**	09	020	**035**	07	09	**012**	020	07	020	**025**	042
	042	075			**022**	**024**	**027**	**031**	053	**072**		
					052	053	**062**	**070**				
					075							
d. External triggers	**06**	**09**	012	**020**	04	07	012	**016**	04	07	**025**	035
	022	024	031	035	022	024	031	042	053	075		
	042	**044**	**062**	**073**	**052**	053						
	075											

e. Internal triggers	**01**	**06**	09	**020**	**04**	09	**012**	031	**016**	**025**	035	042
	024	031	035	042	042	052	062					
	052	062	**070**	**073**								
f. Predictability	**020**	044			**012**	**031**	042		**01**	**07**	**025**	**035**
									042	044	**053**	**072**
g. Unpredictability	09	**022**	**025**	031	**01**	**06**	09	016	016	025	**027**	**035**
	052	**044**	**062**	070	**024**	031	052	**053**				
	075				070	**072**	075					

** Bold indicates codes only reported at one time-point by participants*

Major theme: '*Ways of coping*'

All participants apart from two within the sub-theme '*acceptance*' reported this across time points, which could suggest that this is a consistent experience over time and does not change. Variation within the sub-theme '*avoidance of triggers*' is highlighted whereby some participants (e.g. 06, 025, 052) reported codes related to avoidance at one point and no longer reported this at another time-point, however other participants (e.g. 01, 09, 012) reported this across time-points. This variation was also noted within '*behavioural attempts at symptom management* and '*cognitive attempts at symptom management'* whereby differences were noted over time as well as between these sub-themes. For example, one participant (012) reported behavioural strategies only at one time-point but reported cognitive strategies across two time-points. Within '*support from others*' it was noted that 50% of participants highlighted this sub-theme across all time points, see Table 6.

Table 6: Exploration of frequency sub-themes for each participant within 'Ways of coping' theme

	Emotionalism across more than one time point (N=24)											
'Ways of coping' sub-themes	2-weeks				6-months				12-months			
a. Acceptance	09	024	025	052	04	07	09	024	04	07	025	**027**
	073				052	**072**	073					
b. Avoidance of triggers	**06**	09	012	020	01	04	**07**	09	01	04	016	020
	022	**025**	031	**052**	012	016	**024**	027	027	031	042	**044**
	070	**075**			042				**053**	**072**		
c. Concealing emotionalism from others	**01**	**06**	09	024	**04**	09	016	**022**	016	025	027	035
	025	**031**	035	042	024	027	042	**052**	**044**	053		
	062	**070**			053							
d. Behavioural attempts at symptom	09	**012**	020	**022**	01	04	**07**	09	01	04	**016**	020
management	024	042	**044**	052	024	**027**	**031**	042	**025**	**035**	042	053
	062	**070**			052	053			**072**			
e. Cognitive attempts at symptom	01	**06**	09	012	04	07	09	012	01	04	07	**025**
management	020	024	042	**052**	**016**	020	024	**027**	**035**	042	053	
					031	053	**062**	**073**				

					075							
f. Support from others	**06**	09	012	020	09	012	**022**	027	**07**	020	027	042
	031	**035**	**044**	**052**	031	042						

** Bold indicates codes only reported at one time-point by participants*

Major theme: '*Impact*'

The majority of participants who reported comments related to the sub-theme '*changes to self*' and '*emotional impact*' reported this across time-points. Within the sub-theme '*trying to make sense*' only two participants (016, 042) reported this across time points, which could suggest this was a unique experience for participants at one time-point and was no longer representative of their experience at the following time-point. Within '*uncertainty*' participants reported codes within this sub-theme across time-points but overall, this sub-theme was highlighted less frequently than other sub-themes within this major theme, see Table 7.

Table 7: Exploration of frequency sub-themes for each participant within 'Impact' theme

	Emotionalism across more than one time point (N=24)											
'Impact" sub-themes	2-weeks				6-months				12-months			
a. Changes to self	**01**	09	012	**020**	04	07	09	012	04	07	025	027
	022	**024**	025	031	**016**	027	042	**052**	031	035	042	044
	035	042	044	**062**	053	**073**	075		053	**072**		
	075											
b. Emotional impact	01	09	**06**	020	01	04	07	09	01	04	07	09
	022	**024**	**025**	031	**012**	**016**	022	027	020	027	031	**035**
	042	**044**	052	062	042	052	053	062	042	053	072	
	070	073	075		072	073	075					
c. Trying to make sense	**06**	**012**	**020**	**025**	**09**	016	**024**		016	**027**	042	**044**
	031	**035**	042	**062**					**053**			
	073											
d. Uncertainty	**09**	022	**025**	031	01	**06**	07	022	01	07	027	
	035	042			027	031	042	**052**				
					062	**073**						

** Bold indicates codes only reported at one time-point by participants*

Chapter Six: Thesis portfolio discussion and critical review

Word Count: 2653

Thesis portfolio discussion and critical review

This chapter summarises findings from the systematic review and empirical paper where these will be briefly discussed in relation to previous research and literature in the field. An overview of the strengths and limitations of the thesis portfolio, theoretical and clinical implications and ideas for future research are presented.

Overview of results

The thesis portfolio aimed to explore the neurological, psychological and experiential factors of emotionalism in neurological disorders. A systematic review was conducted to investigate the neurophysiological, neuropsychological and psychological predictors and correlates of emotionalism across neurological disorders. Secondly, an empirical paper explored individuals' experience of post-stroke emotionalism (PSE) over time using a qualitative longitudinal approach.

Systematic review

The review identified predictors and correlates for emotionalism across several neurological disorders such as bulbar networks, serotonergic pathways, frontal areas, white matter, genetics, executive functioning, psychological impact, coping style and female gender. The majority of research investigated neurophysiological predictors and correlates of emotionalism across neurological disorders with a large number of these explored in only stroke populations. The methodological quality of the studies varied with the overall quality ratings ranging from 'good' to 'fair'. A comparison of predictors and correlates was completed tentatively but definitive conclusions could not be drawn due to the disproportionate number of studies across neurological disorders. This suggests a possible complex interplay of various neurophysiological, neuropsychological and psychological predictors and correlates of emotionalism which confirms and extends the current research to date. Overall, the evidence in the review emphasises the importance of serotonin which

highlights any brain area that is relatively more involved in serotonin (production, modulation of function) might show up as more likely damaged across neurological populations thereby increasing the likelihood of emotionalism. This highlights there may not be a specific anatomical or neuropsychological 'signature' due to the widespread presence of serotonin-related mechanisms in the brain and beyond, however findings from the review implicated bulbar and frontal areas as well as white matter tracts involved in connecting frontal, posterior/brain stem/midbrain regions. In addition, neuropsychological findings seem to suggest a role for executive inhibitory control processes.

Empirical paper

The qualitative research explored individuals' experiences of PSE and described how experience of living with PSE may change over time. Three major themes were identified: '*In the moment*', described characteristics and triggers, '*Ways of coping*', highlighted a variation of coping strategies including avoidance or acceptance and '*Impact*', outlined the longer-term effects of PSE such as individuals' beliefs. Over time changes to themes were identified which included increases in avoidance, barriers to control and elements of concealment. This study indicated specific psychological aspects of PSE as highlighted by participants which could potentially be targeted through psychological interventions.

Summary of additional findings

Further analyses were completed of the qualitative interview data from the empirical paper to explore clusters of participants with emotionalism as well as investigating changes in codes and themes of individuals with emotionalism over more than one time-point. The sub-theme 'a*cceptance*' highlighted higher frequencies of participants who were classed as minimally/not distressed across all time-points and this decreased as the levels of distress increased. The frequencies fluctuated according to the

levels of distress within the sub-theme '*avoidance of triggers*' whereby at two-weeks there were greater numbers of participants who were minimally/not distressed compared to those who were moderately distressed. Participants definition of distress could be different at the acute stage post-stroke where the context of being in hospital could potentially impact levels of distress. Furthermore, participants who were minimally/not and very embarrassed showed similar numbers of participants within the '*avoidance of triggers*' sub-theme across time-points, which highlights avoidance of triggers across different levels of embarrassment. However, '*concealing emotionalism from others*' revealed greater frequencies of participants at six-months and 12-months as levels of embarrassment increased, which could suggest embarrassment is a potential mechanism for further concealment of episodes of emotionalism and could reinforce further avoidance.

Interpretation of thesis portfolio findings in relation to previous research and theories

The systematic review supported several proposed neurobiological theories or hypotheses of emotionalism. The theory that it arises because of disruptions to cortical inhibition in the upper brainstem centre and release of the lower bulbar nuclei (Wilson, 1924) was supported by associations with lesions in the pons and emotionalism in stroke participants (Wang et al., 2016) and lesions to the pons were identified as a significant risk factor associated with emotionalism, three months following a stroke (Wei et al., 2016). Furthermore, research also supported the gate control theory highlighting the role of the cerebellum in the modulation of emotion (Parvizi et al., 2009). Reduced mean diffusivity of white matter tracts (Floeter et al., 2014), greater frequency of frontal lobe injury (Tateno et al., 2004) and lower amplitude of frequency fluctuations in the left cerebellum posterior lobe (Liu et al., 2017) were revealed. The role of neurotransmitters was also implicated in the pathophysiology of emotionalism. Selective serotonin reuptake inhibitors (SSRI's) in

the context of RCTs were found to lead to improvements in emotionalism (Andersen et al., 1993; Brown et al., 1998; Burns et al., 1999).

Overall, the findings in the review supported the bulbar account of emotionalism but also extended this to an emerging hypothesis on serotonergic pathways/networks involving raphe nuclei, frontal white matter projections including thalamus, and frontal areas. Frontal lobes are denser with serotonin receptors and the raphe nuclei located in the brainstem are heavily implicated in serotonin production which further supports a proposed serotonin-related network of emotionalism. Any brain area that is relatively more involved in serotonin production or modulation might be highlighted as more likely damaged across neurological populations. Due to the widespread presence of serotonin-related mechanisms in the brain, if this pathway is disrupted in any specific location and potentially disruptions to top-down regulation due to frontal damage this might increase vulnerability to emotional expression in response to a trigger/stressor which could suggest a diathesis-stress model of emotionalism. To conclude, the systematic review expands on existing theories by proposing a serotoninergic model that extends the 1924 'bulbar' model and incorporates a role for brainstem/raphe nuclei and associated structures due to role in serotonin production, white matter/thalamic pathways and frontal pathways.

Only a few studies were identified from the systematic review that explored psychological predictors or correlates of emotionalism. The empirical paper in contrast aimed to explore and provide further knowledge about the psychological and experiential experiences of emotionalism. To date there are no proposed psychological models or theories of PSE, however the empirical paper supported previous research which has revealed emotionalism as associated with avoidance and acceptance-resignation (Eccles et al., 1999; Wei et al., 2016). The study extends this research by highlighting the wide range of situations participants avoided due to episodes of emotionalism such as socialising,

talking on the phone, emotional content on the television and sharing limited information with others.

Furthermore, this current study found participants described avoidance, heightened emotional consequences and negatively held beliefs about episodes of emotionalism which appeared to maintain PSE. For example, some participants described '*feeling a fool*', '*a burden', 'broken'* and '*a complete total idiot'* which also appeared to reinforce further avoidance. This supports the previous smaller-scale qualitative study which revealed participants with negative experiences of emotionalism reported greater disability and avoidance as well as embarrassment and that social withdrawal had a negative effect on individuals' quality of life (McAleese et al., 2019).

Participants frequently described their episodes of emotionalism occurring '*out of the blue*', however over time some participants highlighted a change in the predictability of PSE. An increase in '*prior warning*' and a '*pattern*' in episodes of emotional outburst was revealed at 12-months. When considering the findings from both the systematic review and empirical paper these highlight a potential diathesis-stress model of PSE with a possible convergence about the role of serotonin in responding to (social) stressors (i.e. objectively affective triggers) and the report by many participants of episodes of emotionalism not always being simply totally '*out of the blue*' reactions but rather, overly strong reactions to mild emotional triggers. In addition, some individuals might also experience the *'out of the blue*' type symptoms which fits with the finding of increased emotional reactivity to objectively 'neutral' stimuli demonstrated by studies using electrophysiological methods. This all indicates a multi-process model implicating vulnerability to emotional reaction to any stimuli, but perhaps especially emotive stimuli as well as failed top-down regulation and psychological and behavioural coping that might serve to prevent reappraisal or learning top-down control.

Strengths and limitations

The thesis portfolio presents two novel main papers, which provide a comprehensive synthesis of predictors and correlates of emotionalism across neurological disorders and a longitudinal qualitative investigation of the lived experience of emotionalism over time. The systematic review included a range of neurological disorders enabling a comparison of similarities and differences in terms of predictors and correlates, which to the authors' knowledge to date has not been completed before. There is limited research to date exploring emotionalism and this thesis portfolio helps to increase knowledge of this condition, inform clinical practice and provide recommendations for future research.

Additionally, the use of a longitudinal qualitative approach for the empirical paper allowed for detailed descriptions of participants' experiences of emotionalism to be investigated and this enabled a framework/model to be proposed to account for the participants' experiences and common mechanisms of PSE. The sample size was large with N = 100 participants with emotionalism which enabled an investigation of experiences of emotionalism in the acute stage and over time. This research took a critical realist approach which allowed the researcher to discover the deeper causal mechanisms and experience of emotionalism, however further research is required to extend, change or reject the proposed psychological framework/model of PSE due to its possible fallibility.

A limitation of the empirical paper which needs to be considered when interpreting the results was that the majority of participants were from the West of Scotland. This sample were largely less disabled (lower NIHSS/Barthel Index scores) which could highlight questions of how representative the participants were of the stroke population and of PSE. This means that caution is required when generalising the results to other populations and cultures, which highlights the need for further research to explore

emotionalism in diverse samples. Furthermore, 25% of the codes were reviewed by a second independent rater where ideally a higher percentage would have been second reviewed. This could have further reduced researcher bias, increased transparency and promoted reflexivity. However, ensuring a percentage of codes were second reviewed improved the definitions of the codes and rigour of coding of the quotes.

The systematic review adopted broad inclusion and exclusion criteria, which allowed for a detailed synthesis to be completed on the published literature to date. However, this highlighted the differences in methods used to diagnose emotionalism which increases bias and potential error of the previous research findings with some researchers using clinical judgement and others using standardised criteria such as Kim's criteria (Kim & Choi-Kwon, 2000). There is a lack of consensus in the literature regarding how to measure emotionalism using validated psychometric approaches which highlights the need for development of such measures. The Testing Emotionalism After Recent Stroke – Questionnaire (TEARS-Q; Broomfield et al., 2020) has been recently developed for stroke populations but similar emotionalism measures need to be developed for other neurologic disorders.

Theoretical implications

The empirical paper supports previous research and understanding of emotionalism where participants described their episodes of emotionalism as brief, lasting a few seconds to several minutes (House et al., 1989; Calvert et al., 1998; Eccles et al., 1999). Furthermore, participants highlighted episodes of emotionalism were incongruent with their emotions at the time whereby they were crying but did not experience feelings of sadness. This supports previous research which has revealed that although emotionalism can be co-morbid with depression these are two distinct entities (Robinson et al., 1993).

Until the work conducted in this portfolio no psychological model of emotionalism had been proposed. This thesis portfolio highlights a biopsychosocial model that comprises neurobiology/anatomy, neuropsychological regulator processes, psychological aspects such as meaning making and behaviour in social context including avoidance, concealment and embarrassment which will vary depending on social attitudes towards emotional expression and the responses of others. A visual model/framework was produced summarising the qualitative interview data from participants with lived experience of PSE. Although the proposed framework/model is based on experiences of PSE there is no a priori research to consider emotionalism is different in other neurological disorders. This hopes to increase knowledge and understanding of PSE but also highlights the importance of further qualitative research across neurological disorders.

Clinical implications

The empirical paper highlighted the possible benefits of providing psycho-education to individuals with PSE. The qualitative analysis identified a comprehensive list of internal and external triggers which appeared to set off episodes of emotionalism. Participants described attempts to understand emotionalism or a 'search for facts' where clinicians could consider providing information regarding emotionalism and possible triggers to create further awareness and insight. In the systematic review correlations between younger age and female gender with emotionalism were revealed, which could provide a targeted approach in delivering psycho-education.

The narrative synthesis of the systematic review identified a number of key brain areas or networks, disruptions to the serotoninergic pathways and psychological variables such as avoidance coping style. These emphasise important factors that could be targeted through possible SSRI treatment or psychological treatment models. The empirical paper also helps to improve our knowledge and understanding of potential mechanisms of

emotionalism. Both papers can inform clinical practice with the empirical paper suggesting possible targets for psychological interventions such as normalising their experience and providing immediate coping strategies to potentially reduce the longer-term consequences of emotionalism characterised by avoidance, embarrassment and distress.

Future work and ideas

The methodological quality of the studies included in the systematic review varied which emphasises the need for further high-quality research to be conducted. The majority of the studies were cross-sectional where future randomised controlled trials (RCT's) would enable more definitive conclusions to be drawn and extend the findings from the systematic review. Additionally, further research is required to explore predictors and correlates in neurological disorders such as vascular dementia, traumatic brain injury (TBI) and Alzheimer's disease as this review highlighted very limited research of emotionalism in these disorders.

The empirical paper could be extended by exploring individuals' lived experience who have recovered from emotionalism. This could help to confirm findings from the empirical paper but also to add further information such as what they found helpful and to reflect on their experience since they have recovered. The emotional consequences of emotionalism were highlighted by participants and future research could explore the emotional impact of emotionalism on carers and relatives. This could explore how carers or relatives support individuals with emotionalism and also the possible impact of burden or distress on carers or relatives.

Finally, single case experimental designs (SCEDs) or RCTs could be conducted to evaluate the efficacy of psychological interventions in supporting individuals with emotionalism. The empirical paper highlighted possible mechanisms which could be targeted through psychological interventions such as reducing avoidance, increasing

acceptance of their experience and providing psycho-education to increase knowledge about emotionalism. This future research would extend theoretical and clinical knowledge of emotionalism and support future clinical practice and definitive clinical trial research.

Overall conclusion

This thesis portfolio presents an overview of neurological, psychological and experiential factors of emotionalism in neurological disorders. As highlighted throughout the discussion, a range of neurophysiological and several psychological predictors and correlates of emotionalism were identified across neurological disorders. This provides insight into the possible complex interplay of multiple variables associated with emotionalism where future research is required to shape further understanding. Participants' experience and emotional consequences of emotionalism were revealed from the qualitative analysis. This research indicates possible psychological aspects of emotionalism which could be targeted through psychological interventions. Overall, this thesis portfolio provides further understanding and knowledge of emotionalism which hopes to inform clinicians and clinical practice to provide treatment and management to individuals with emotionalism.

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Appendices

Appendix A: Submission guidelines for Neuropsychological Rehabilitation

About the Journal

Neuropsychological Rehabilitation is an international, peer-reviewed journal publishing high-quality, original research. Please see the journal's Aims & Scope for information about its focus and peer-review policy.

Please note that this journal only publishes manuscripts in English.

Neuropsychological Rehabilitation accepts the following types of article: original articles, scholarly reviews, book reviews.

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All authors submitting to medicine, biomedicine, health sciences, allied and public health journals should conform to the Uniform Requirements for Manuscripts Submitted to

Biomedical Journals, prepared by the International Committee of Medical Journal Editors (ICMJE).

Clinical trials: must conform to the Consort guidelines http://www.consort-statement.org. Submitted papers should include a checklist confirming that all of the Consort requirements have been met, together with the corresponding page number of the manuscript where the information is located. In addition, trials must be pre-registered on a site such as clinicaltrials.gov or equivalent, and the manuscript should include the reference number to the relevant pre-registration.

Systematic reviews: submitted papers should follow PRISMA http://www.prisma-statement.org/ guidelines and submission should also be accompanied by a completed PRISMA checklist, together with the corresponding page number of the manuscript where the information is located.

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Observational studies: submitted papers should follow the STROBE guidelines (https://www.strobe-statement.org/index.php?id=strobe-home) and also include a completed checklist of compliance, together with the corresponding page number of the manuscript where the information is located.

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The EQUATOR Network (Enhancing the Quality and Transparency of Health Research) website provides further information on available guidelines.

Structure

Your paper should be compiled in the following order: title page; abstract; keywords; main text introduction, materials and methods, results, discussion; acknowledgments; declaration of interest statement; references; appendices (as appropriate); table(s) with caption(s) (on individual pages); figures; figure captions (as a list).

Word Limits

Please include a word count for your paper. There are no word limits for papers in this journal.

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Authors may submit their paper in any scholarly format or layout. Manuscripts may be supplied as single or multiple files. These can be Word, rich text format (rtf), open

document format (odt), or PDF files. Figures and tables can be placed within the text or submitted as separate documents. Figures should be of sufficient resolution to enable refereeing.

- There are no strict formatting requirements, but all manuscripts must contain the essential elements needed to evaluate a manuscript: abstract, author affiliation, figures, tables, funder information, and references. Further details may be requested upon acceptance.
- References can be in any style or format, so long as a consistent scholarly citation format is applied. Author name(s), journal or book title, article or chapter title, year of publication, volume and issue (where appropriate) and page numbers are essential. All bibliographic entries must contain a corresponding in-text citation. The addition of DOI (Digital Object Identifier) numbers is recommended but not essential.
- The journal reference style will be applied to the paper post-acceptance by Taylor & Francis.
- Spelling can be US or UK English so long as usage is consistent.

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Checklist: What to Include

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2. Should contain an unstructured abstract of 200 words.
3. You can opt to include a **video abstract** with your article. Find out how these can help your work reach a wider audience, and what to think about when filming.
4. Between 5 and 5 **keywords**. Read making your article more discoverable, including information on choosing a title and search engine optimization.
5. **Funding details.** Please supply all details required by your funding and grant-awarding bodies as follows:
   For single agency grants
   This work was supported by the [Funding Agency] under Grant [number xxxx].
   For multiple agency grants

This work was supported by the [Funding Agency #1] under Grant [number xxxx]; [Funding Agency #2] under Grant [number xxxx]; and [Funding Agency #3] under Grant [number xxxx].

6. **Disclosure statement.** This is to acknowledge any financial interest or benefit that has arisen from the direct applications of your research. Further guidance on what is a conflict of interest and how to disclose it.
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8. **Data deposition.** If you choose to share or make the data underlying the study open, please deposit your data in a recognized data repository prior to or at the time of submission. You will be asked to provide the DOI, pre-reserved DOI, or other persistent identifier for the data set.
9. **Geolocation information.** Submitting a geolocation information section, as a separate paragraph before your acknowledgements, means we can index your paper's study area accurately in JournalMap's geographic literature database and make your article more discoverable to others. More information.
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11. **Figures.** Figures should be high quality (1200 dpi for line art, 600 dpi for grayscale and 300 dpi for colour, at the correct size). Figures should be supplied in one of our preferred file formats: EPS, PS, JPEG, TIFF, or Microsoft Word (DOC or DOCX) files are acceptable for figures that have been drawn in Word. For information relating to other file types, please consult our Submission of electronic artwork document.
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14. **Units.** Please use SI units (non-italicized).

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Please ensure that all research reported in submitted papers has been conducted in an ethical and responsible manner, and is in full compliance with all relevant codes of experimentation and legislation. All papers which report in vivo experiments or clinical trials on humans or animals must include a written statement in the Methods section. This should explain that all work was conducted with the formal approval of the local human subject or animal care committees (institutional and national), and that clinical trials have been registered as legislation requires. Authors who do not have formal ethics review committees should include a statement that their study follows the principles of the Declaration of Helsinki.

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Appendix B: Prospero registration

PROSPERO
International prospective register of systematic reviews

NHS
National Institute for Health Research

UNIVERSITY of York
Centre for Reviews and Dissemination

Systematic review

*Fields that have an **asterisk (*)** next to them means that they **must be answered. Word limits** are provided for each section. You will be unable to submit the form if the word limits are exceeded for any section. Registrant means the person filling out the form.*

1. * Review title.

Give the title of the review in English

Predictors and correlates of emotionalism across acquired and progressive neurological conditions: a systematic review

2. Original language title.

For reviews in languages other than English, give the title in the original language. This will be displayed with the English language title.

3. * Anticipated or actual start date.

Give the date the systematic review started or is expected to start.

28/02/2020

4. * Anticipated completion date.

Give the date by which the review is expected to be completed.

28/08/2020

5. * Stage of review at time of this submission.

Tick the boxes to show which review tasks have been started and which have been completed. Update this field each time any amendments are made to a published record.

Reviews that have started data extraction (at the time of initial submission) are not eligible for inclusion in PROSPERO. If there is later evidence that incorrect status and/or completion date has been supplied, the published PROSPERO record will be marked as retracted.

This field uses answers to initial screening questions. It cannot be edited until after registration.

The review has not yet started: No

Review stage	Started	Completed
Preliminary searches	Yes	Yes
Piloting of the study selection process	Yes	Yes
Formal screening of search results against eligibility criteria	Yes	No
Data extraction	No	No
Risk of bias (quality) assessment	No	No
Data analysis	No	No

Provide any other relevant information about the stage of the review here.

6. * Named contact.

The named contact is the guarantor for the accuracy of the information in the register record. This may be any member of the review team.

Sophie Fitzgerald

Email salutation (e.g. "Dr Smith" or "Joanne") for correspondence:

Miss Fitzgerald

7. * Named contact email.

Give the electronic email address of the named contact.

sophie.fitzgerald@uea.ac.uk

8. Named contact address

Give the full institutional/organisational postal address for the named contact.

Department of Clinical Psychology, Norwich Medical School, University of East Anglia, Norwich Research Park, Norwich, NR4 7TJ

9. Named contact phone number.

Give the telephone number for the named contact, including international dialling code.

01603 591258

10. * Organisational affiliation of the review.

Full title of the organisational affiliations for this review and website address if available. This field may be completed as 'None' if the review is not affiliated to any organisation.

University of East Anglia (UEA)

Organisation web address:

https://www.uea.ac.uk/

11. * Review team members and their organisational affiliations.

Give the personal details and the organisational affiliations of each member of the review team. Affiliation refers to groups or organisations to which review team members belong. **NOTE: email and country now MUST be entered for each person, unless you are amending a published record.**

Miss Sophie Fitzgerald. UEA
Professor Niall Broomfield. University of East Anglia
Dr Fergus Gracey. University of East Anglia

12. * Funding sources/sponsors.

Details of the individuals, organizations, groups, companies or other legal entities who have funded or sponsored the review.

None.

Grant number(s)

State the funder, grant or award number and the date of award

13. * Conflicts of interest.

List actual or perceived conflicts of interest (financial or academic).

None

14. Collaborators.

Give the name and affiliation of any individuals or organisations who are working on the review but who are not listed as review team members. **NOTE: email and country must be completed for each person, unless you are amending a published record.**

Dr David Gillespie.

15. * Review question.

State the review question(s) clearly and precisely. It may be appropriate to break very broad questions down into a series of related more specific questions. Questions may be framed or refined using PI(E)COS or similar where relevant.

This systematic review aims to:

1. To identify the neurophysiological/neuropsychological predictors and correlates of emotionalism across neurological disorders: Stroke, Parkinson's Disease, Multiple Sclerosis, Traumatic Brain Injury (TBI), Alzheimer's Disease, Vascular Dementia and Amyotrophic Lateral Sclerosis (ALS).

2. To identify the psychological predictors of emotionalism across neurological disorders.

P: Neurological disorders.

I: Neurophysiological / neuropsychological / psychological predictors and correlates

C: Not applicable.

O: Emotionalism (how it presents/defined across disorders).

S: Correlational studies, prospective studies and cross-sectional studies; observational, cohort and case control.

16. * Searches.

State the sources that will be searched (e.g. Medline). Give the search dates, and any restrictions (e.g. language or publication date). Do NOT enter the full search strategy (it may be provided as a link or attachment below.)

A comprehensive, systematic search of MEDLINE, Cumulative Index to Nursing and Allied Health Literature (CINAHL complete), PsycINFO and EMBASE will be completed.

Boolean operators (OR, AND) will be used to search each neurological disorder with the search terms for emotionalism individually. Results generated will then be searched individually to determine if they meet the inclusion criteria for this systematic review.

Keywords and Medical Subject Headings (MeSH) will be used when completing the search strategy. The participant/population search terms will be combined with the outcome search term.

17. URL to search strategy.

Upload a file with your search strategy, or an example of a search strategy for a specific database, (including the keywords) in pdf or word format. In doing so you are consenting to the file being made publicly accessible. Or provide a URL or link to the strategy. Do NOT provide links to your search **results**.

https://www.crd.york.ac.uk/PROSPEROFILES/159413_STRATEGY_20200228.pdf

Alternatively, upload your search strategy to CRD in pdf format. Please note that by doing so you are consenting to the file being made publicly accessible.

Do not make this file publicly available until the review is complete

18. * Condition or domain being studied.

Give a short description of the disease, condition or healthcare domain being studied in your systematic review.

Emotionalism, also known as pseudobulbar affect (PBA) is characterised by uncontrolled episodes of sudden crying or laughing which can be disproportionate or inappropriate to the social context. Emotionalism can occur in neurological conditions such as Stroke, Parkinson's Disease, Multiple Sclerosis (MS), Traumatic Brain Injury (TBI) Alzheimer's Disease, Vascular Dementia and Amyotrophic Lateral Sclerosis (ALS). Emotionalism can have a significant impact on an individual's life and can result in embarrassment and avoidance which can limit social interactions and lower quality of life.

19. * Participants/population.

Specify the participants or populations being studied in the review. The preferred format includes details of both inclusion and exclusion criteria.

Inclusion criteria – studies of emotionalism with adults (over the age of 18) with a neurological condition will be included in this review. No restrictions on time since onset of emotionalism.

No defined exclusion criteria.

20. * Intervention(s), exposure(s).

Give full and clear descriptions or definitions of the interventions or the exposures to be reviewed. The preferred format includes details of both inclusion and exclusion criteria.

Investigation of predictors and correlates: 1. Biological variables (neurophysiological, anatomical, neuropsychological) and 2. Psychological variables.

21. * Comparator(s)/control.

Where relevant, give details of the alternatives against which the intervention/exposure will be compared (e.g. another intervention or a non-exposed control group). The preferred format includes details of both inclusion and exclusion criteria.

Not applicable.

22. * Types of study to be included.

Give details of the study designs (e.g. RCT) that are eligible for inclusion in the review. The preferred format includes both inclusion and exclusion criteria. If there are no restrictions on the types of study, this should be stated.

Inclusion criteria – quantitative studies, cross sectional studies, observational, cohort study and case control.

Exclusion criteria – qualitative studies, reviews, dissertations, unpublished 'grey' literature and non-English studies.

23. Context.

Give summary details of the setting or other relevant characteristics, which help define the inclusion or exclusion criteria.

No limits in terms of context. Studies across different settings such as hospital, residential nursing home, supported living and independent living in the community will be included.

24. * Main outcome(s).

Give the pre-specified main (most important) outcomes of the review, including details of how the outcome is defined and measured and when these measurement are made, if these are part of the review inclusion criteria.

How emotionalism is defined across neurological disorders and the presence/absence of emotionalism or degree of emotionalism across neurological disorders diagnosed/assessed using methods such as standardised Kim's criteria or House, Dennis, Molyneux, Warlow & Hawton (1989), interviews or self-report.

* Measures of effect

Please specify the effect measure(s) for you main outcome(s) e.g. relative risks, odds ratios, risk difference, and/or 'number needed to treat.

Not applicable.

25. * Additional outcome(s).

List the pre-specified additional outcomes of the review, with a similar level of detail to that required for main outcomes. Where there are no additional outcomes please state 'None' or 'Not applicable' as appropriate to the review

None.

* Measures of effect

Please specify the effect measure(s) for you additional outcome(s) e.g. relative risks, odds ratios, risk difference, and/or 'number needed to treat.

Not applicable.

26. * Data extraction (selection and coding).

Describe how studies will be selected for inclusion. State what data will be extracted or obtained. State how this will be done and recorded.

The search strategy will be completed for each neurological condition and all search results will be gathered and managed using an excel spreadsheet (SF). Any duplications will be removed.

The reviewer (SF) will review the titles and abstracts of the studies from the search results for eligibility and exclude studies that do not meet criteria.

Full texts of the studies that meet the eligibility criteria will be reviewed and reference lists of studies will be hand-searched to check if any potential studies were not captured by the search strategy. Reasons for excluding studies will be recorded and a diagram to show flow of information will be created.

After this has been completed, the reviewer (SF) will extract data on (a) study characteristics; authors, year, country/setting, diagnosis, sample size and makeup, independent variables/predictors/correlates, measures of emotionalism used, context or setting, study design, age range and other relevant characteristics, (b) neurophysiological/neuropsychological predictors and correlates of emotionalism across neurological disorders: Stroke, Parkinson's Disease, Multiple Sclerosis, Traumatic Brain Injury (TBI), Alzheimer's Disease, Vascular Dementia, Amyotrophic Lateral Sclerosis (ALS) and (c) the psychological predictors of emotionalism across neurological disorders. This data will be summarised in a table with methodology quality ratings and summary of study conclusions.

A second reviewer will independently review a proportion of papers at the initial title and abstract stage and full text stage to ensure agreement between reviewers and to ensure that they meet the inclusion criteria. If there are any disagreements a third reviewer will be consulted.

27. * Risk of bias (quality) assessment.

State which characteristics of the studies will be assessed and/or any formal risk of bias/quality assessment

tools that will be used.

Quality Assessment Tool for Observational Cohort and Cross-sectional Studies (QATOCCS) will be used to rate the methodological quality of studies. This tool will examine researcher bias, sample bias, sample size, time effects, accuracy and reliability of outcome measures, drop-out rates and if confounding variables were accounted for. This consists of 14 questions whereby two independent reviewers will rate each element from good to poor for each study.

Studies will be graded using Scottish Intercollegiate Guidelines Network (2002) and summarised according to the level of evidence. This ranges from 1++ for high quality meta-analyses to 4 for expert opinion, formal consensus.

28. * Strategy for data synthesis.

Describe the methods you plan to use to synthesise data. This **must not be generic text** but should be **specific to your review** and describe how the proposed approach will be applied to your data. If meta-analysis is planned, describe the models to be used, methods to explore statistical heterogeneity, and software package to be used.

Due to the significant heterogeneity in how emotionalism is measured and small sample sizes a narrative synthesis will be completed rather than a meta-analysis.

This systematic review will follow the narrative synthesis framework by Popay et al. (2006) to describe the neurophysiological/neuropsychological predictors and correlates of emotionalism across each neurological disorder: Stroke, Parkinson's Disease, Multiple Sclerosis, Traumatic Brain Injury (TBI), Alzheimer's Disease, Vascular Dementia, Amyotrophic Lateral Sclerosis (ALS) and (b) the psychological predictors of emotionalism across neurological disorders. The narrative synthesis will adopt a textual approach to summarise and explain the findings of the synthesis, explore relationships in the data and assess the robustness of the synthesis.

29. * Analysis of subgroups or subsets.

State any planned investigation of 'subgroups'. Be clear and specific about which type of study or participant will be included in each group or covariate investigated. State the planned analytic approach.

Neurophysiological/neuropsychological and psychological predictors and correlates of emotionalism will be explored independently for each neurological disorder initially. Following this, neurophysiological/neuropsychological and psychological predictors and correlates could be compared to explore differences between neurological disorders.

30. * Type and method of review.

Select the type of review, review method and health area from the lists below.

Type of review

Cost effectiveness
No

Diagnostic

No

Epidemiologic
No

Individual patient data (IPD) meta-analysis
No

Intervention
No

Meta-analysis
No

Methodology
No

Narrative synthesis
Yes

Network meta-analysis
No

Pre-clinical
No

Prevention
No

Prognostic
No

Prospective meta-analysis (PMA)
No

Review of reviews
No

Service delivery
No

Synthesis of qualitative studies
No

Systematic review
Yes

Other
No

Health area of the review

Alcohol/substance misuse/abuse
No

Blood and immune system
No

Cancer
No

Cardiovascular
Yes

Care of the elderly
No

Child health
No

Complementary therapies
No

COVID-19
No

Crime and justice
No

Dental
No

Digestive system
No

Ear, nose and throat
No

Education
No

Endocrine and metabolic disorders
No

Eye disorders
No

General interest
No

Genetics
No

Health inequalities/health equity
No

Infections and infestations
No

International development
No

Mental health and behavioural conditions
Yes

Musculoskeletal
No

Neurological
Yes

Nursing
No

Obstetrics and gynaecology
No

Oral health

No

Palliative care
No

Perioperative care
No

Physiotherapy
No

Pregnancy and childbirth
No

Public health (including social determinants of health)
Yes

Rehabilitation
No

Respiratory disorders
No

Service delivery
No

Skin disorders
No

Social care
No

Surgery
No

Tropical Medicine
No

Urological
No

Wounds, injuries and accidents
No

Violence and abuse
No

31. Language.

Select each language individually to add it to the list below, use the bin icon to remove any added in error.

There is an English language summary.

32. * Country.

Select the country in which the review is being carried out. For multi-national collaborations select all the countries involved.

England

33. Other registration details.

Name any other organisation where the systematic review title or protocol is registered (e.g. Campbell, or The Joanna Briggs Institute) together with any unique identification number assigned by them. If extracted data will be stored and made available through a repository such as the Systematic Review Data Repository (SRDR), details and a link should be included here. If none, leave blank.

34. Reference and/or URL for published protocol.

If the protocol for this review is published provide details (authors, title and journal details, preferably in Vancouver format)

Add web link to the published protocol.

Or, upload your published protocol here in pdf format. Note that the upload will be publicly accessible.

No I do not make this file publicly available until the review is complete

Please note that the information required in the PROSPERO registration form must be completed in full even if access to a protocol is given.

35. Dissemination plans.

Do you intend to publish the review on completion?

Yes

Give brief details of plans for communicating review findings.?

Once the systematic review has been completed a paper will be submitted to a leading journal in this field for publication.

36. Keywords.

Give words or phrases that best describe the review. Separate keywords with a semicolon or new line. Keywords help PROSPERO users find your review (keywords do not appear in the public record but are included in searches). Be as specific and precise as possible. Avoid acronyms and abbreviations unless these are in wide use.

Emotionalism; Pseudobulbar Affect; Stroke; Parkinson's Disease; Multiple Sclerosis; Traumatic Brain Injury; Alzheimer's Disease; Vascular Dementia; Amyotrophic Lateral Sclerosis; predictor; correlate.

37. Details of any existing review of the same topic by the same authors.

If you are registering an update of an existing review give details of the earlier versions and include a full bibliographic reference, if available.

38. * Current review status.

Update review status when the review is completed and when it is published.New registrations must be ongoing so this field is not editable for initial submission.
Please provide anticipated publication date

Review_Ongoing

39. Any additional information.

Provide any other information relevant to the registration of this review.

40. Details of final report/publication(s) or preprints if available.

Leave empty until publication details are available OR you have a link to a preprint (NOTE: this field is not editable for initial submission). List authors, title and journal details preferably in Vancouver format.

Give the link to the published review or preprint.

Appendix C: Systematic Review search strategy

Table 1: Search terms for PsycINFO, PubMed and CINAHL Complete databases

Participant / Population	**AND**	**Outcome**
Stroke - "stroke" OR "cerebr* accident" OR "cva" OR "apoplexy" OR (MM "Stroke")	AND	"emotionalism" OR "emotional lability" OR "emotional dysregulation" OR "involuntary emotional expression disorder" OR "involuntary crying" OR "involuntary laughing" OR "lability of mood" OR "pathological laughing" OR "pathological crying" OR "pseudobulbar affect" OR "emotional incontinence" OR "pathological display of affect" OR "inappropriate laughing" OR "inappropriate crying"
Multiple Sclerosis - "MS" OR "multiple sclerosis" OR "Sclerosis Disseminated" OR (MM "Multiple Sclerosis")	AND	"emotionalism" OR "emotional lability" OR "emotional dysregulation" OR "involuntary emotional expression disorder" OR "involuntary crying" OR "involuntary laughing" OR "lability of mood" OR "pathological laughing" OR "pathological crying" OR "pseudobulbar affect" OR "emotional incontinence" OR "pathological display of affect" OR "inappropriate laughing" OR "inappropriate crying"
Parkinson's Disease - "idiopathic parkinson* disease" OR "paralysis agitans" OR "parkinson* disease idiopathic" OR	AND	"emotionalism" OR "emotional lability" OR "emotional dysregulation" OR "involuntary emotional expression disorder" OR "involuntary crying" OR "involuntary laughing" OR "lability of mood" OR "pathological laughing" OR "pathological

"parkinson* disease" OR "primary parkinsonism" OR (MM "Parkinsonian Disorders")		crying" OR "pseudobulbar affect" OR "emotional incontinence" OR "pathological display of affect" OR "inappropriate laughing" OR "inappropriate crying"
Traumatic Brain Injury - "traumatic brain injury" OR "TBI" OR "brain injury" OR "head injury" OR "head trauma" OR "traumatic encephalopathy" OR "acquired brain injur*" OR (MM "Brain Injuries")	AND	"emotionalism" OR "emotional lability" OR "emotional dysregulation" OR "involuntary emotional expression disorder" OR "involuntary crying" OR "involuntary laughing" OR "lability of mood" OR "pathological laughing" OR "pathological crying" OR "pseudobulbar affect" OR "emotional incontinence" OR "pathological display of affect" OR "inappropriate laughing" OR "inappropriate crying"
Alzheimer's disease - "alzheimer* disease" OR "dementia" OR (MM "Alzheimer Disease")	AND	"emotionalism" OR "emotional lability" OR "emotional dysregulation" OR "involuntary emotional expression disorder" OR "involuntary crying" OR "involuntary laughing" OR "lability of mood" OR "pathological laughing" OR "pathological crying" OR "pseudobulbar affect" OR "emotional incontinence" OR "pathological display of affect" OR "inappropriate laughing" OR "inappropriate crying"
Vascular dementia - "vascular dementia" OR	AND	"emotionalism" OR "emotional lability" OR "emotional dysregulation" OR "involuntary emotional expression disorder" OR "involuntary

"VaD" OR (MM "Dementia, Vascular")		crying" OR "involuntary laughing" OR "lability of mood" OR "pathological laughing" OR "pathological crying" OR "pseudobulbar affect" OR "emotional incontinence" OR "pathological display of affect" OR "inappropriate laughing" OR "inappropriate crying"
Amyotrophic lateral sclerosis - "ALS" OR "amyotrophic lateral sclerosis" OR "Lou Gehrig* disease" OR "motor neurone disease" OR "MND" OR (MM "Amyotrophic Lateral Sclerosis")	AND	"emotionalism" OR "emotional lability" OR "emotional dysregulation" OR "involuntary emotional expression disorder" OR "involuntary crying" OR "involuntary laughing" OR "lability of mood" OR "pathological laughing" OR "pathological crying" OR "pseudobulbar affect" OR "emotional incontinence" OR "pathological display of affect" OR "inappropriate laughing" OR "inappropriate crying"

Table 2: Search terms for Embase database

1. stroke.mp.
2. (cerebr* adj3 accident).mp.
3. cva.mp.
4. apoplexy.mp.
5. cerebrovascular accident/
6. MS.mp.
7. (multiple adj3 sclerosis).mp.
8. (sclerosis adj3 disseminated).mp.

9. multiple sclerosis/
10. (idiopathic adj3 parkinson* adj3 disease).mp.
11. (paralysis adj3 agitans).mp.
12. (parkinson* adj3 disease adj3 idiopathic).mp.
13. (parkinson* adj3 disease).mp.
14. (primary adj3 parkinsonism).mp.
15. Parkinson disease/
16. (traumatic adj3 brain adj3 injury).mp.
17. TBI.mp.
18. (brain adj3 injury).mp.
19. (head adj3 injury).mp.
20. (head adj3 trauma).mp.
21. (traumatic adj3 encephalopathy).mp.
22. (acquired adj3 brain adj3 injur*).mp.
23. traumatic brain injury/
24. (alzheimer* adj3 disease).mp.
25. dementia.mp.
26. Alzheimer disease/
27. (vascular adj3 dementia).mp.
28. VaD.mp.
29. multiinfarct dementia/
30. ALS.mp.’
31. (amyotrophic adj3 lateral adj3 sclerosis).mp.
32. (Lou adj3 Gehrig* adj3 disease).mp.

33. (motor adj3 neurone adj3 disease).mp.
34. MND.mp.
35. amyotrophic lateral sclerosis/
36. emotionalism.mp.
37. (emotional adj3 lability).mp.
38. (emotional adj3 dysregulation).mp.
39. (involuntary adj3 emotional adj3 expression adj3 disorder).mp.
40. (involuntary adj3 crying).mp.
41. (involuntary adj3 laughing).mp.
42. (lability adj3 of adj3 mood).mp.
43. (pathological adj3 laughing).mp.
44. (pathological adj3 crying).mp.
45. (pseudobulbar adj3 affect).mp.
46. (emotional adj3 incontinence).mp.
47. (pathological adj3 display adj3 of adj3 affect).mp.
48. (inappropriate adj3 laughing).mp.
49. (inappropriate adj3 crying).mp.

*[mp=title, abstract, heading word, drug trade name, original title, device manufacturer, drug manufacturer, device trade name, keyword, floating subheading word, candidate term word]

Stroke: 1 or 2 or 3 or 4 or 5 and 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49

Multiple sclerosis: 6 or 7 or 8 or 9 and 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49

Parkinson's disease: 10 or 11 or 12 or 13 or 14 or 15 and 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49

Traumatic Brain Injury: 16 or 17 or 18 or 19 or 20 or 21 or 22 or 23 and 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49

Alzheimer's disease: 24 or 25 or 26 and 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49

Vascular Dementia: 27 or 28 or 29 and 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49

Amyotrophic lateral sclerosis: 30 or 31 or 32 or 33 or 34 or 35 and 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49

Appendix D: Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies (QATOCCS)

Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies

Criteria	Yes	No	Other (CD/NR/NA)
1. Was the research question or objective in this paper clearly stated?			
2. Was the study population clearly specified and defined?			
3. Was the participation rate of eligible persons at least 50%?			
4. Were all the subjects selected or recruited from the same or similar populations (including the same time period)? Were inclusion and exclusion criteria for being in the study prespecified and applied uniformly to all participants?			
5. Was a sample size justification, power description, or variance and effect estimates provided?			
6. For the analyses in this paper, were the exposure(s) of interest measured prior to the outcome(s) being measured?			
7. Was the timeframe sufficient so that one could reasonably expect to see an association between exposure and outcome if it existed?			
8. For exposures that can vary in amount or level, did the study examine different levels of the exposure as related to the outcome (e.g., categories of exposure, or exposure measured as continuous variable)?			
9. Were the exposure measures (independent variables) clearly defined, valid, reliable, and implemented consistently across all study participants?			
10. Was the exposure(s) assessed more than once over time?			
11. Were the outcome measures (dependent variables) clearly defined, valid, reliable, and implemented consistently across all study participants?			
12. Were the outcome assessors blinded to the exposure status of participants?			
13. Was loss to follow-up after baseline 20% or less?			
14. Were key potential confounding variables measured and adjusted statistically for their impact on the relationship between exposure(s) and outcome(s)?			

*CD, cannot determine; NA, not applicable; NR, not reported

Quality Rating (Good, Fair or Poor)	
Rater 1 initials	
Rater 2 initials	
Additional Comments	

Appendix E: Kim and Choi-Kwon (2000) Criteria

Emotionalism is present if both patients and relatives agreed that ≥ 2 occasions of excessive or inappropriate laughing or crying or both has occurred as compared with their premorbid state. Additionally, laughter or crying episodes must be either excessive and/or inappropriate.

Appendix F: House et al. (1989) Criteria

1. Have you been more tearful since the stroke than you were beforehand? Have you actually cried more in the past month (not just felt like it)?
2. Does the weepiness come suddenly, at times when you aren't expecting it? (Suddenly means with only a few moments or no warning, not after several minutes trying to control yourself?
3. If you feel the tears coming on, or it they have started, can you control yourself to stop them? Have you been unable to stop yourself crying in front of other people? Is that a new experience for you?

Appendix G: Prisma Checklist

Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	15
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.	16
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	20-21
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	21
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.	21
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.	21-24
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.	24
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	24
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).	41-42
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.	25
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	22-24
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.	26-27

Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	NA
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.	NA

Section/topic	#	Checklist item	Reported on page #
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	NA
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	25
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	42
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	43-45
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	45-50
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	51-60
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	NA
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	NA
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	61
DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	61-62
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	64-66
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	66-71
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	72

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(7): e1000097. doi:10.1371/journal.pmed1000097

For more information, visit: **www.prisma-statement.org**.

Appendix H: Submission guidelines for Disability and Rehabilitation

About the journal

Disability and Rehabilitation is an international, peer reviewed journal, publishing high-quality, original research. Please see the journal's Aims & Scope for information about its focus and peer-review policy.

From 2018, this journal will be online only, and will no longer provide print copies.

Please note that this journal only publishes manuscripts in English.

Disability and Rehabilitation accepts the following types of article: Reviews, Research Papers, Case Studies, Perspectives on Rehabilitation, Reports on Rehabilitation in Practice, Education and Training, and Correspondence. Systematic Reviews should be submitted as "Review" and Narrative Reviews should be submitted as "Perspectives in Rehabilitation".

Special Issues and specific sections on contemporary themes of interest to the Journal's readership are published. Please contact the Editor for more information.

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Please visit our Author Services website or contact openaccess@tandf.co.uk if you would like more information about our Open Select Program.

*Citations received up to Jan 31st 2020 for articles published in 2015-2019 in journals listed in Web of Science®.
**Usage in 2017-2019 for articles published in 2015-2019.

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Taylor & Francis is committed to peer-review integrity and upholding the highest standards of review. For submissions to *Disability and Rehabilitation* authors are given the option to remain anonymous during the peer-review process. Authors will be able to

indicate whether their paper is ‘Anonymous’ or ‘Not Anonymous’ during submission, and should pay particular attention to the below:

- Authors who wish to remain **anonymous** should prepare a complete text with information identifying the author(s) removed. This should be uploaded as the “Main Document” and will be sent to the referees. A separate title page should be included providing the full affiliations of all authors. Any acknowledgements and the Declaration of Interest statement must be included but should be worded mindful that these sections will be made available to referees.
- Authors who wish to be **identified** should include the name(s) and affiliation(s) of author(s) on the first page of the manuscript. The complete text should be uploaded as the “Main Document”.

Once your paper has been assessed for suitability by the editor, it will be peer-reviewed by independent, anonymous expert referees. Find out more about what to expect during peer review and read our guidance on publishing ethics.

Preparing your paper

All authors submitting to medicine, biomedicine, health sciences, allied and public health journals should conform to the Uniform Requirements for Manuscripts Submitted to Biomedical Journals, prepared by the International Committee of Medical Journal Editors (ICMJE).

We also refer authors to the community standards explicit in the American Psychological Association's (APA) Ethical Principles of Psychologists and Code of Conduct.

We encourage authors to be aware of standardised reporting guidelines below when preparing their manuscripts:

- Case reports - CARE
- Diagnostic accuracy - STARD
- Observational studies - STROBE
- Randomized controlled trial - CONSORT
- Systematic reviews, meta-analyses - PRISMA

Whilst the use of such guidelines is supported, due to the multi-disciplinary nature of the Journal, it is not compulsory.

Structure

Your paper should be compiled in the following order: title page; abstract; keywords; main text, introduction, materials and methods, results, discussion; acknowledgments; declaration of interest statement; references; appendices (as appropriate); table(s) with caption(s); figures; figure captions (as a list).

In the main text, an introductory section should state the purpose of the paper and give a brief account of previous work. New techniques and modifications should be described concisely but in sufficient detail to permit their evaluation. Standard methods should

simply be referenced. Experimental results should be presented in the most appropriate form, with sufficient explanation to assist their interpretation; their discussion should form a distinct section.

Tables and figures should be referred to in text as follows: figure 1, table 1, i.e. lower case. The place at which a table or figure is to be inserted in the printed text should be indicated clearly on a manuscript. Each table and/or figure must have a title that explains its purpose without reference to the text.

The title page should include the full names and affiliations of all authors involved in the preparation of the manuscript. The corresponding author should be clearly designated, with full contact information provided for this person.

Word count

Please include a word count for your paper. There is no word limit for papers submitted to this journal, but succinct and well-constructed papers are preferred.

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Please refer to these style guidelines when preparing your paper, rather than any published articles or a sample copy.

Please use any spelling consistently throughout your manuscript.

Please use double quotation marks, except where "a quotation is 'within' a quotation". Please note that long quotations should be indented without quotation marks.

For tables and figures, the usual statistical conventions should be used.

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Papers may be submitted in any standard format, including Word and LaTeX. Figures should be saved separately from the text. To assist you in preparing your paper, we provide formatting template(s).

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References

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Checklist: what to include

1. **Author details**. Please ensure everyone meeting the International Committee of Medical Journal Editors (ICJME) requirements for authorship is included as an author of your paper. All authors of a manuscript should include their full name and affiliation on the cover page of the manuscript. Where available, please also include ORCiDs and social media handles (Facebook, Twitter or LinkedIn). One author will need to be identified as the corresponding author, with their email address normally displayed in the article PDF (depending on the journal) and the online article. Authors' affiliations are the affiliations where the research was conducted. If any of the named co-authors moves affiliation during the peer-review process, the new affiliation can be given as a footnote. Please note that no changes to affiliation can be made after your paper is accepted. Read more on authorship.
2. A structured **abstract** of no more than 200 words. A structured abstract should cover (in the following order): the *purpose* of the article, its *materials and methods* (the design and methodological procedures used), the *results* and conclusions (including their relevance to the study of disability and rehabilitation). Read tips on writing your abstract.
3. You can opt to include a **video abstract** with your article. Find out how these can help your work reach a wider audience, and what to think about when filming.
4. 5-8 **keywords**. Read making your article more discoverable, including information on choosing a title and search engine optimization.
5. A feature of this journal is a boxed insert on **Implications for Rehabilitation**. This should include between two to four main bullet points drawing out the implications for rehabilitation for your paper. This should be uploaded as a separate document. Below are examples:
   Example 1: Leprosy

- Leprosy is a disabling disease which not only impacts physically but restricts quality of life often through stigmatisation.
- Reconstructive surgery is a technique available to this group.
- In a relatively small sample this study shows participation and social functioning improved after surgery.

Example 2: Multiple Sclerosis

- Exercise is an effective means of improving health and well-being experienced by people with multiple sclerosis (MS).
- People with MS have complex reasons for choosing to exercise or not.

- Individual structured programmes are most likely to be successful in encouraging exercise in this cohort.

6. **Acknowledgement**. Please supply all details required by your funding and grant-awarding bodies as follows: *For single agency grants*: This work was supported by the under Grant . *For multiple agency grants*: This work was supported by the under Grant ; under Grant ; and under Grant .
7. **Declaration of Interest**. This is to acknowledge any financial interest or benefit that has arisen from the direct applications of your research. Further guidance on what is a declaration of interest and how to disclose it.
8. **Data availability statement.** If there is a data set associated with the paper, please provide information about where the data supporting the results or analyses presented in the paper can be found. Where applicable, this should include the hyperlink, DOI or other persistent identifier associated with the data set(s). Templates are also available to support authors.
9. **Data deposition.** If you choose to share or make the data underlying the study open, please deposit your data in a recognized data repository prior to or at the time of submission. You will be asked to provide the DOI, pre-reserved DOI, or other persistent identifier for the data set.
10. **Supplemental online material.** Supplemental material can be a video, dataset, fileset, sound file or anything which supports (and is pertinent to) your paper. We publish supplemental material online via Figshare. Find out more about supplemental material and how to submit it with your article.
11. **Figures.** Figures should be high quality (1200 dpi for line art, 600 dpi for grayscale and 300 dpi for colour). Figures should be saved as TIFF, PostScript or EPS files.
12. **Tables.** Tables should present new information rather than duplicating what is in the text. Readers should be able to interpret the table without reference to the text. Please supply editable files.
13. **Equations**. If you are submitting your manuscript as a Word document, please ensure that equations are editable. More information about mathematical symbols and equations.
14. **Units.** Please use SI units (non-italicized).

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You must obtain the necessary permission to reuse third-party material in your article. The use of short extracts of text and some other types of material is usually permitted, on a limited basis, for the purposes of criticism and review without securing formal permission. If you wish to include any material in your paper for which you do not hold copyright, and which is not covered by this informal agreement, you will need to obtain written permission from the copyright owner prior to submission. More information on requesting permission to reproduce work(s) under copyright.

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Please include a declaration of interest statement, using the subheading "Declaration of interest." If you have no interests to declare, please state this (suggested wording: *The authors report no conflicts of interest*). For all NIH/Wellcome-funded papers, the grant number(s) must be included in the disclosure of interest statement. Read more on declaring conflicts of interest.

Clinical Trials Registry

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Please ensure that all research reported in submitted papers has been conducted in an ethical and responsible manner, and is in full compliance with all relevant codes of experimentation and legislation. All papers which report *in vivo* experiments or clinical trials on humans or animals must include a written statement in the Methods section. This should explain that all work was conducted with the formal approval of the local human subject or animal care committees (institutional and national), and that clinical trials have been registered as legislation requires. Authors who do not have formal ethics review committees should include a statement that their study follows the principles of the Declaration of Helsinki.

Consent

All authors are required to follow the ICMJE requirements on privacy and informed consent from patients and study participants. Please confirm that any patient, service user, or participant (or that person's parent or legal guardian) in any research, experiment, or clinical trial described in your paper has given written consent to the inclusion of material pertaining to themselves, that they acknowledge that they cannot be identified via the paper; and that you have fully anonymized them. Where someone is deceased, please ensure you have written consent from the family or estate. Authors may use this Patient Consent Form, which should be completed, saved, and sent to the journal if requested.

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Authors are further encouraged to cite any data sets referenced in the article and provide a Data Availability Statement.

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Appendix I: Ethical approval for the 'Testing for Emotionalism After Recent Stroke' (TEARS) study

Scotland A Research Ethics Committee

Research Ethics Service
2nd Floor Waverley Gate
2-4 Waterloo Place
Edinburgh
EH1 3EG
Telephone: 0131 465 5680
www.hra.nhs.uk

Dr Niall M Broomfield
Consultant Psychologist, Lead for Stroke Neuropsychology
NHS Greater Glasgow and Clyde
Department of Psychology
Level 3 G Block
Western Infirmary
Glasgow
G11 6NT

Date: 17 February 2015
Your Ref.:
Our Ref.: 14/SS/1103
Enquiries to: Walter Hunter
Extension: 35680
Direct Line: 0131 465 5680
Email: walter.hunter@nhslothian.scot.nhs.uk

Dear Dr Broomfield

Study title: **Testing emotionalism after recent stroke (TEARS)**
REC reference: **14/SS/1103**
IRAS project ID: **157483**

Thank you for your letter of 13 February 2015, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Vice-Chair.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this favourable opinion letter. The expectation is that this information will be published for all studies that receive an ethical opinion but should you wish to provide a substitute contact point, wish to make a request to defer, or require further information, please contact the REC Manager, Mr Walter Hunter, Walter.Hunter@nhslothian.scot.nhs.uk . Under very limited circumstances (e.g. for student research which has received an unfavourable opinion), it may be possible to grant an exemption to the publication of the study.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation [as revised], subject to the conditions specified below.

Adults with Incapacity (Scotland) Act 2000

I confirm that the Committee has approved this research project for the purposes of the Adults with Incapacity (Scotland) Act 2000. The Committee is satisfied that the requirements of section 51 of the Act will be met in relation to research carried out as part

of this project on, or in relation to, a person who lacks capacity to consent to taking part in the project.

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at http://www.rdforum.nhs.uk.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of approvals from host organisations

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database. This should be before the first participant is recruited but no later than 6 weeks after recruitment of the first participant.
There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to request a deferral for study registration within the required timeframe, they should contact hra.studyregistration@nhs.net. The expectation is that all clinical trials will be registered, however, in exceptional circumstances non registration may be permissible with prior agreement from NRES. Guidance on where to register is provided on the HRA website.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Ethical review of research sites

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document	*Version*	*Date*
Covering letter on headed paper		26 November 2014
Covering letter on headed paper: Justification for including adults lacking capacity		10 February 2015
GP letter: Baseline	Two	02 February 2015
GP letter: Six months	Two	02 February 2015
GP letter: 12 months	Two	02 February 2015
GP letter: Advise 6 months follow-up	One	02 February 2015
GP letter: Advise 12 months follow-up	One	02 February 2015
Letter from funder		04 July 2014
Letter from statistician	2.0	10 October 2014
Non-validated questionnaire		
Non-validated questionnaire		
Letter from funder		03 January 2014
Other	One	04 February 2015
Participant consent form: Aphasia Cognitive Impair	2	19 November 2014
Participant consent form: Aphasia Cognitive Impair-to continue	2	19 November 2014
Participant consent form: Carer-to continue	4	04 February 2015
Participant consent form: Carer	4	04 February 2015
Participant consent form: Participant	Four	04 February 2015

Participant consent form: Participant-to continue	Four	04 February 2015
Participant consent form: Welfare guardian/nearest relative	Four	04 February 2015
Participant information sheet: Participant	Four	04 February 2015
Participant information sheet: Participant-to continue	Four	04 February 2015
Participant information sheet: Welfare Guardian/nearest relative	Four	04 February 2015
Participant information sheet: Welfare Guardian/nearest relative-to continue	Four	02 February 2015
Participant: Carer	Four	04 February 2015
Participant information sheet: Carer-to continue	Four	04 February 2015
REC Application Form		26 November 2014
Referee's report or other scientific critique report		
Research protocol or project proposal	2	02 October 2014
Research protocol	Eight	04 February 2015
Summary CV for Chief Investigator (CI)		
Summary, synopsis or diagram (flowchart) of protocol in non technical language	Eight	29 January 2015

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document *"After ethical review – guidance for researchers"* gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website: http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/

HRA Training

We are pleased to welcome researchers and R&D staff at our training days – see details at http://www.hra.nhs.uk/hra-training/

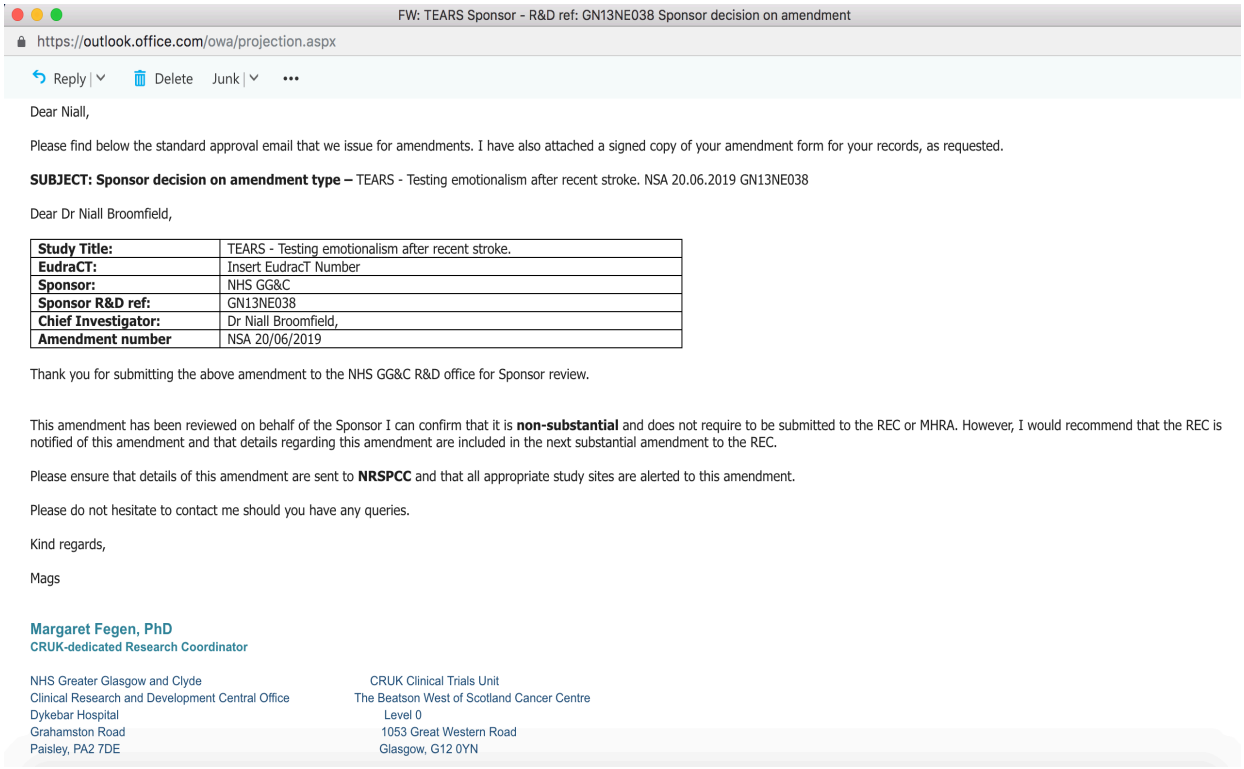
Thank you for responding to the Committee's request for further information on the above research, and enclosing the following revised documents:

REC reference number: 14/SS/1103-Please quote this number on all correspondence

Yours sincerely

Dr Colin Selby
Committee Vice Chairman
cc: Dr Erica Packard

Appendix J: Non-substantial amendment form agreed by study sponsor of the TEARS study



FW: TEARS Sponsor - R&D ref: GN13NE038 Sponsor decision on amendment

https://outlook.office.com/owa/projection.aspx

Reply | Delete Junk | •••

Dear Niall,

Please find below the standard approval email that we issue for amendments. I have also attached a signed copy of your amendment form for your records, as requested.

SUBJECT: Sponsor decision on amendment type – TEARS - Testing emotionalism after recent stroke. NSA 20.06.2019 GN13NE038

Dear Dr Niall Broomfield,

Study Title:	TEARS - Testing emotionalism after recent stroke.
EudraCT:	Insert EudracT Number
Sponsor:	NHS GG&C
Sponsor R&D ref:	GN13NE038
Chief Investigator:	Dr Niall Broomfield,
Amendment number	NSA 20/06/2019

Thank you for submitting the above amendment to the NHS GG&C R&D office for Sponsor review.

This amendment has been reviewed on behalf of the Sponsor I can confirm that it is **non-substantial** and does not require to be submitted to the REC or MHRA. However, I would recommend that the REC is notified of this amendment and that details regarding this amendment are included in the next substantial amendment to the REC.

Please ensure that details of this amendment are sent to **NRSPCC** and that all appropriate study sites are alerted to this amendment.

Please do not hesitate to contact me should you have any queries.

Kind regards,

Mags

Margaret Fegen, PhD
CRUK-dedicated Research Coordinator

NHS Greater Glasgow and Clyde
Clinical Research and Development Central Office
Dykebar Hospital
Grahamston Road
Paisley, PA2 7DE

CRUK Clinical Trials Unit
The Beatson West of Scotland Cancer Centre
Level 0
1053 Great Western Road
Glasgow, G12 0YN

Partner Organisations:
Health Research Authority, England NIHR Clinical Research Network, England
NHS Research Scotland NISCHR Permissions Co-ordinating Unit, Wales
HSC Research & Development, Public Health Agency, Northern Ireland

Notification of Non-Substantial/Minor Amendments(s) for NHS Studies

This template **must only** be used to notify NHS/HSC R&D office(s) of amendments, which are **NOT** categorised as Substantial Amendments.
If you need to notify a Substantial Amendment to your study then you MUST use the appropriate Substantial Amendment form in IRAS.

Instructions for using this template

- For guidance on amendments refer to http://www.hra.nhs.uk/research-community/during-your-research-project/amendments/
- This template should be completed by the CI and optionally authorised by Sponsor, if required by sponsor guidelines.
- This form should be submitted according to the instructions provided for NHS/HSC R&D at http://www.hra.nhs.uk/research-community/during-your-research-project/amendments/which-review-bodies-need-to-approve-or-be-notified-of-which-types-of-amendments/ . If you do not submit your notification in accordance with these instructions then processing of your submission may be significantly delayed.

1. Study Information

Full title of study:	Testing Emotionalism After Recent Stroke
IRAS Project ID:	157483
Sponsor Amendment Notification number:	One
Sponsor Amendment Notification date:	June 19th 2019
Details of Chief Investigator:	
Name [first name and surname]	Professor Niall M Broomfield
Address:	Department of Clinical Psychology Norwich Medical School University of East Anglia University of East Anglia Norwich Research Park Norwich, UK
Postcode:	NR4 7TJ
Contact telephone number:	Tel: 01603 591217 07973 375705
Email address:	
Details of Lead Sponsor: **NHS Greater Glasgow and Clyde Research and Development**	

Partner Organisations:
Health Research Authority, England NIHR Clinical Research Network, England
NHS Research Scotland NISCHR Permissions Co-ordinating Unit, Wales
HSC Research & Development, Public Health Agency, Northern Ireland

Name:	**Margaret Fegen**
Contact email address:	Margaret.Fegen@ggc.scot.nhs.uk
Details of Lead Nation:	
Name of lead nation *delete as appropriate*	Scotland, although UEA as research site is England
If England led is the study going through CSP? *delete as appropriate*	Yes / No
Name of lead R&D office:	NHS Greater Glasgow and Clyde

Partner Organisations:
Health Research Authority, England NIHR Clinical Research Network, England
NHS Research Scotland NISCHR Permissions Co-ordinating Unit, Wales
HSC Research & Development, Public Health Agency, Northern Ireland

2. Summary of amendment(s)

This template **must only** be used to notify NHS/HSC R&D office(s) of amendments, which are **NOT** categorised as Substantial Amendments.
If you need to notify a Substantial Amendment to your study then you MUST use the appropriate Substantial Amendment form in IRAS.

No.	Brief description of amendment *(please enter each separate amendment in a new row)*	Amendment applies to *(delete/ list as appropriate)*		List relevant supporting document(s), including version numbers *(please ensure all referenced supporting documents are submitted with this form)*		R&D category of amendment *(category A, B, C) For office use only*
		Nation	**Sites**	**Document**	**Version**	
1	I am Chief Investigator for TEARS, a study of post-stroke emotionalism prevalence which recruited across five health boards in Scotland. The study commenced when I was Consultant Clinical Psychologist for stroke in NHS Greater Glasgow and Clyde and Hon Clinical Associate Professor at University of Glasgow I am now in a new post at University of East Anglia Medical School. We plan that a part of the data analysis on the TEARS study will be undertaken at the University of East Anglia, under my supervision, involving two doctoral students (ClinPsyD) and one undergraduate student. The details of this are as follows: - We will receive at UEA already anonymised data from Robertson Centre for Biostatistics - This data will be sent to us securely via a password protected server set up by RCB - This will be for the purposes of already planned and approved data analysis - This will be completed by myself and the students - With the help and advice of the Leeds statistics	England	University of East Anglia	TEARS Final Protocol Feb 2015 TEARS SAP (Statistical Analysis Plan)	8 2	
		Northern Ireland	n/a			
		Scotland	n/a			
		Wales	n/a			

	team (Prof West being our study statistician, he also has access to all data) - The students will be closely supervised in their work on the anonymised data and will write this up for their doctoral research and/or for publication - Their work will cover pre-designated areas as set out in the study protocol and study analysis plan - [redacted] (doctoral student) and [redacted] (undergraduate intern) will work on the secondary quantitative analyses of the neuropsychological, quality of life, social participation and functional data (see Statistical Analysis Plan Section 9.2 p. 13 and study protocol 4.2.2 p.26 and p.27/8) - Sophie Fitzgerald (doctoral trainee) will work on transcription and qualitative analysis of the TEARS-IV and TEARS-IV audio recorded interviews (see study protocol 8.8 p. 29) - I am CI for TEARS, and I will be PI for the new UEA site and thus closely supervise the students, and with assistance/advice from Leeds Statistical team (Prof West)					
2						
3						
4						
5						

[Add further rows as required]

Partner Organisations:
Health Research Authority, England NIHR Clinical Research Network, England
NHS Research Scotland NISCHR Permissions Co-ordinating Unit, Wales
HSC Research & Development, Public Health Agency, Northern Ireland

3. Declaration(s)

Declaration by Chief Investigator

- I confirm that the information in this form is accurate to the best of my knowledge and I take full responsibility for it.
- I consider that it would be reasonable for the proposed amendment(s) to be implemented.

Signature of Chief Investigator:

Print name: Prof Niall M Broomfield

Date: 19 June 2019

Optional Declaration by the Sponsor's Representative (as per Sponsor Guidelines)

The sponsor of an approved study is responsible for all amendments made during its conduct.

The person authorising the declaration should be authorised to do so. There is no requirement for a particular level of seniority; the sponsor's rules on delegated authority should be adhered to.

- I confirm the sponsor's support for the amendment(s) in this notification.

Signature of sponsor's representative: …  ……

Print name: *Dr Margaret Fegen*……

Post: ……Research Coordinator……

Organisation:……NHS Greateer Glasgow & Clyde……

Date:……20/06/2019……

Appendix K: University of East Anglia's Faculty of Medicine and Health Sciences (FMH) Research Ethics Committee approval

Faculty of Medicine and Health Sciences Research Ethics Committee

University of East Anglia

Sophie Fitzgerald
Department of Clinical Psychology
Norwich Medical School
University of East Anglia
Norwich
Research Park
Norwich NR4 7TJ

NORWICH MEDICAL SCHOOL
Bob Champion Research & Educational Building
James Watson Road
University of East Anglia
Norwich Research Park
Norwich NR4 7UQ

Email fmh.ethics@uea.ac.uk
www.med.uea.ac.uk

4th October 2019

Dear Sophie

Post-stroke emotionalism (PSW): A qualitative longitudinal study exploring individuals' experience with PSE **Reference: 161**

The submission of your research proposal was discussed at the Faculty Research Ethics Committee meeting on 26th September 2019.

The Committee were happy to approve your application in principle but have the following concern which they would like you to address and amend accordingly:

- The researcher will be transcribing recordings and will have access to demographic data. Please clarify at what point the data will become anonymous and how security of this data will be ensured.

Please write to me once you have resolved/clarified the above. I require documentation confirming that you have complied with the Committee's requirements. The Committee has requested that you detail the changes below the relevant point on the text in this letter and also include your amendments as a tracked change within your application/proposal. The revisions to your application can be considered by Chair's action, rather than a further committee meeting, which means that you can resubmit the above documentation at any time. Please send your revisions to me as an attachment in an email as this will speed up the decision making process.

As your project does not have ethics approval until the above issues have been resolved, I want to remind you that you should not be undertaking your research project until you have ethical approval by the Faculty Research Ethics Committee. Planning on the project or literature based elements can still take place but not the research involving the above ethical issues. This is to ensure that you and your research are insured by the University and that your research is undertaken within the University's 'Guidelines on Good Practice in Research' approved by Senate in July 2015.

Yours sincerely

Prof Alastair Forbes
Chair
FMH Research Ethics Committee

Faculty of Medicine and Health Sciences Research Ethics Committee

University of East Anglia

NORWICH MEDICAL SCHOOL
Bob Champion Research & Educational Building
James Watson Road
University of East Anglia
Norwich Research Park
Norwich NR4 7UQ

Email: fmh.ethics@uea.ac.uk
www.med.uea.ac.uk

Sophie Fitzgerald
Department of Clinical Psychology
Norwich Medical School
University of East Anglia
Norwich
Research Park
Norwich NR4 7TJ

14th November 2019

Dear Sophie

Project title: Post-stroke emotionalism (PSE): A qualitative longitudinal study exploring individuals experience with PSE

Reference: 2019/20-161

Thank you for your email of 31st October 2019 notifying us of the amendments you would like to make to your above proposal. These have been considered and I can confirm that your amendments have been approved.

Please can you ensure that any further amendments to either the protocol or documents submitted are notified to us in advance, and that any adverse events which occur during your project are reported to the Committee.

Approval by the FMH Research Committee should not be taken as evidence that your study is compliant with GDPR and the Data Protection Act 2018. If you need guidance on how to make your study GDPR compliant, please contact your institution's Data Protection Officer.

Please can you arrange to send us a report once your project is completed.

Yours sincerely

Prof Alastair Forbes
Chair
FMH Research Ethics Committee

Appendix L: General Data Protection Regulation (GDPR) approval from University of East Anglia's Data Protection Information Compliance Specialist

FW: TEARS GDPR

From: "Paul Cutting (ITCS - Staff)" <Paul.Cutting@uea.ac.uk> on behalf of Data Protection <dataprotection@uea.ac.uk>
Date: Thursday, 2 April 2020 at 11:51
To: "Niall Broomfield (MED - Staff)" <N.Broomfield@uea.ac.uk>
Subject: RE: TEARS GDPR

Dear Niall,

Further to my email yesterday, I've discussed this with Ellen.

We think the current data protection picture of this research can be summarised as follows:

1. We still don't have a clear picture of who is responsible for the data at each stage of the research process
2. UoG have told us that they do have a contract in place with GG&C and have shared the data with UEA on the basis of that contract
3. We are unclear as to whether the data is anonymised or pseudonymised, however based on our understanding and previous conversations we believe the position is that it would be difficult if not impossible to identify individuals from the data
4. Based on the privacy notice we've seen and subsequent conversations with UoG we do feel that there are still some gaps

If we were at the start of this project the university might handle this differently from a Data Protection point of view. However given the nature of the data, the fact that there are agreements and privacy notices in place and that all parties are happy to proceed; we believe there is a low risk to the individuals and feel that it is acceptable to proceed.

Kind regards

Paul

Paul Cutting
Information Compliance Specialist
Information Compliance
Room 1.24, The Library
University of East Anglia, Norwich Research Park, Norwich, NR4 7TJ
Tel: 01603 591143

Appendix M: TEARS clinical interview schedule

Testing for Emotionalism After Recent Stroke-Case Interview (TEARS-IV)

******SWITCH ON DIGITAL RECORDER NOW******

A. Introduction

Hello. Stroke can sometimes cause changes to emotions (that is, how we feel inside) or emotional expression (that is, how we show our emotions - in particular through crying and laughter). In this interview, I am going to ask some screening questions first about crying, then about laughter, to see if this has been a problem for you since the stroke. If it has, I will also ask you some more detailed questions, is that OK?

B. Post-Stroke Crying: Screen Questions

OK, so first I am going to ask you some YES/NO questions about crying since the stroke.

(i) Have you felt more tearful **in the past two weeks** than before the stroke? ◯NO ◯YES**

(ii) Have you actually cried more **in the past two weeks** than before the stroke, not just felt like it?

◯NO◯YES**

***** If response is YES on <u>either</u> item (i) or item (ii), complete sections C-E.***

If response is NO to both items (i) and (ii), go to section F.

C. Post-Stroke Crying: Case Characteristics

I will now ask you more about your emotions since the stroke and about how you express those emotions especially in tears - any changes to these, how this affects you and what you do.

If any questions are especially upsetting and you do not wish to continue, please say.

I will record your responses on this sheet, to help me remember. I will also record the interview using this hand-held digital recorder. Is that Okay?

To start, let's talk about your emotions and emotional expression, in particular crying, since the stroke, in more detail.

I. What have your emotions and emotional expression, in particular crying, been like since the stroke?

Prompt: So have you changed in any way then? What is that like? Does it make sense, this idea of how we feel inside being different sometimes from how we express our feelings?

II. Have you been more tearful since the stroke than you were beforehand?

Prompt: What is that like? Can you say how often? Was this a pattern just following the stroke, but not now?
Can you tell me more?

III. Have you actually cried more in the past month? (not just felt like it?)

Prompt: You said sometimes. How often is that? What is that like? Can you tell me more?

IV. What are the crying episodes actually like? Does the weepiness come suddenly, at times when you aren't expecting it? Suddenly means with only a few minutes no warning, not after several minutes trying to control yourself.

Prompt: Does that happen?
What is that like?
How often does that happen?
If you have episodes of crying, do you tend to sob loudly?
Can you tell me more?

V. If you feel the tears coming on, or if they have started, can you control yourself to stop them?

Prompt: So can you sometimes put it off by trying to control it? What helps?
So for instance, have you been able to stop yourself crying in front of other people? What did you do?
What was that like? Can you tell me more?

OK, thinking again about the past two weeks:

has the crying come on suddenly, with only a few seconds or no warning? ◯NO◯YES

has the crying occurred at times you were not expecting it? ◯NO◯YES

has the crying come on even if you did not feel sad at the time ◯NO ◯YES

And again thinking about the past two weeks:

when the crying occurred, could you control yourself to stop it? ◯NO◯YES

did the crying come on in situations you would not have cried in before the stroke? ◯NO◯YES

D. Frequency and Impact of Crying

OK, now I am going to ask more about the frequency and impact of the crying episodes

Thinking about the past two weeks:

(i) Exactly how often have you experienced crying episodes?

◯ rare- less than once per week

◯ sometimes- at least once per week

◯ often- several times per week but less than every day

◯ very often- more than once per day

(ii) Approximately how long have the crying episodes lasted?

◯ just a few seconds

◯ up to one minute

◯ several minutes

◯ ten minutes or more

(iii) How distressing have you found the crying episodes?

◯ minimal/not distressing

◯ moderately distressing

◯ very distressing

◯ very severely distressing

◯ unsure

(iv) How embarrassing have you found the crying episodes?

◯ minimally/not embarrassing

◯ moderately embarrassing

◯ very embarrassing

◯ very severely embarrassing

◯ unsure

(v) Again, thinking about the past two weeks, have any of the following events set off/triggered the crying episodes:

Happy news item ◯NO◯YES

Sad news item ◯NO◯YES

Distressing news item ◯NO◯YES

Emotional news item ◯NO◯YES

Sentimental news item ◯NO◯YES

Having happy memories in mind ◯NO◯YES

Having sad memories in mind ◯NO◯YES

Having distressing memories in mind ◯NO◯YES

Having emotional memories in mind ◯NO◯YES

Having sentimental memories in mind ◯NO◯YES

Talking about the stroke ◯NO◯YES

Talking about crying since the stroke ◯NO◯YES

A kind word from others ◯NO◯YES

Have any factors not listed here set off the crying episodes, please specify:

(vi) Again, thinking of the past two weeks, have the crying episodes stopped you from doing anything?

◯NO◯YES

If Yes, which three activities has the crying episodes most stopped you doing (e.g. meeting new people, meeting familiar people, going out, answering the telephone?)

Specify:

1.

2.

3.

(vi) Again, thinking of the past two weeks, have crying episodes caused you to avoid people or situations?

◯NO◯YES

If Yes, which three aspects have you most avoided because of the crying episodes?

Specify:

1.

2.

3.

E. Coping Strategies for Crying
OK, now I am going to ask what you and others do to cope with the crying episodes.

Thinking about the crying episodes in the past two weeks:

(i) Have you used any of the following strategies to try and stop/limit the crying episodes?

Looking away	◯NO◯YES
Hiding behind a book or newspaper	◯NO◯YES
Leaving a room	◯NO◯YES
Trying to think of something else	◯NO◯YES
Controlling your breathing	◯NO◯YES
Trying to relax	◯NO◯YES
Counting to ten	◯NO◯YES
Covering your face with your hands	◯NO◯YES

If Yes, can you describe which three main methods you use most:

1.

2.

3.

(iii) Of the strategies that you have used to try and stop crying episodes, have any actually helped?

Looking away	◯ Yes, helpful	◯ No, unhelpful	◯ Not tried
Hiding behind the newspaper	◯ Yes, helpful	◯ No, unhelpful	◯ Not tried
Leaving the room	◯ Yes, helpful	◯ No, unhelpful	◯ Not tried

Thinking of something else ◯ Yes, helpful ◯ No, unhelpful ◯ Not tried

Controlling your breathing ◯ Yes, helpful ◯ No, unhelpful ◯ Not tried

Relaxing ◯ Yes, helpful ◯ No, unhelpful ◯ Not tried

Counting to ten ◯ Yes, helpful ◯ No, unhelpful ◯ Not tried

Covering your face with hands ◯ Yes, helpful ◯ No, unhelpful ◯ Not tried

Which other strategies, not mentioned above, have you found helpful to stop/limit crying episodes?

1.

2.

3.

(iv) Have other people used any of the following to try and stop/limit the crying episodes?

Ignoring you are crying ◯ Yes, others tried ◯ No, others not tried

Talking about something else ◯ Yes, others tried ◯ No, others not tried

Distracting you off the crying ◯ Yes, others tried ◯ No, others not tried

Comforting you ◯ Yes, others tried ◯ No, others not tried

Reassuring you ◯ Yes, others tried ◯ No, others not tried

(v) Which of these strategies tried by other people have helped?

Ignoring you are crying ◯ Yes, helpful ◯ No, unhelpful ◯ Not tried

Talking about something else ◯ Yes, helpful ◯ No, unhelpful ◯ Not tried

Distracting you off the crying ◯ Yes, helpful ◯ No, unhelpful ◯ Not tried

Comforting you ◯ Yes, helpful ◯ No, unhelpful ◯ Not tried

Reassuring you ◯ Yes, helpful ◯ No, unhelpful ◯ Not tried

Which strategies used by others, not mentioned above, have been helpful to stop/limit the crying episodes?

1.

2.

3.

F. Post-Stroke Laughter: Screen Questions

It may seem unusual, because stroke is something people usually find upsetting, but some people find that they might laugh more than they did before the stroke, or switch suddenly between laughing and crying.

(i) Have you noticed difficulty controlling laughter outbursts **in the past two weeks**, more than before the stroke? ◯NO ◯YES**

(ii) Have you actually suddenly laughed out loud **in the past two weeks**, not just felt like it?

◯NO◯YES**

***** If response is YES on <u>either</u> item (i) or item (ii), complete section G.***

If response is NO to both items (i) and (ii), go to section H.

G. Post-Stroke Laughter: Case Characteristics

I will now ask you more about your emotions since the stroke and about how you express those emotions in laughter - any changes to these, how this affects you and what you do.

OK, thinking again about the past two weeks:

has the laughing come on suddenly, with only a few seconds or no warning? ◯NO◯YES

has the laughing occurred at times you were not expecting it? ◯NO◯YES

has the laughing come on even if you did not feel amused at the time ◯NO◯YES

Again thinking about the past two weeks:

when the laughing occurred, could you control yourself to stop it? ◯NO◯YES

did the laughing come on in situations you would not have laughed in before the stroke?

◯NO◯YES

Again thinking about the past two weeks:

(i) Exactly how often have you experienced laughter episodes?

◯ rare- less than once per week

◯ sometimes- at least once per week

◯ often- several times per week but less than every day

◯ very often- more than once per day

(ii) Approximately how long have the laughter episodes lasted?

◯ just a few seconds

◯ up to one minute

◯ several minutes

◯ ten minutes or more

(iii) How distressing have you found the laughter episodes?

◯ minimal/not distressing

◯ moderately distressing

◯ very distressing

◯ very severely distressing

◯ unsure

(iv) How embarrassing have you found the laughter episodes?

◯ minimally/not embarrassing

◯ moderately embarrassing

◯ very embarrassing

◯ very severely embarrassing

◯ unsure

(v) Again, thinking about the past two weeks, have any of the following events set off/triggered the laughter episodes:

Happy news item ◯NO◯YES

Sad news item ◯NO◯YES

Distressing news item ◯NO◯YES

Emotional news item ◯NO◯YES

Sentimental news item ◯NO◯YES

Having happy memories in mind ◯NO◯YES

Having sad memories in mind ◯NO◯YES

Having distressing memories in mind ◯NO◯YES

Having emotional memories in mind ◯NO◯YES

Having sentimental memories in mind ◯NO◯YES

Talking about the stroke ◯NO◯YES

Talking about laughter since the stroke ◯NO◯YES

A kind word from others ◯NO◯YES

Have any factors not listed here set off the laughter episodes, please specify:

(vi) Again, thinking of the past two weeks, have the laughter episodes stopped you from doing anything?

◯NO◯YES

If Yes, which three activities has the crying episodes most stopped you doing (e.g. meeting new people, meeting familiar people, going out, answering the telephone?)

Specify:

1.

2.

3.

(vi) Again, thinking of the past two weeks, have laughter episodes caused you to avoid people or situations?

◯NO◯YES

If Yes, which three aspects have you most avoided because of the laughter episodes?

Specify:

1.

2.

Again thinking about any laughter episodes in the past two weeks:

(i) Have you used any of the following strategies to try and stop/limit the laughter episodes?

Looking away ◯NO◯YES

Hiding behind a book or newspaper ◯NO◯YES

Leaving a room ◯NO◯YES

Trying to think of something else ◯NO◯YES

Controlling your breathing ◯NO◯YES

Trying to relax ◯NO◯YES

Counting to ten ◯NO◯YES

Covering your face with your hands ◯NO◯YES

If Yes, can you describe which three main methods you use most:

1.

2.

3.

(iii) Of the strategies that you have used to try and stop crying episodes, have any actually helped?

Looking away ◯ Yes, helpful ◯ No, unhelpful ◯ Not tried

Hiding behind the newspaper ◯ Yes, helpful ◯ No, unhelpful ◯ Not tried

Leaving the room ◯ Yes, helpful ◯ No, unhelpful ◯ Not tried

Thinking of something else ◯ Yes, helpful ◯ No, unhelpful ◯ Not tried

Controlling your breathing ◯ Yes, helpful ◯ No, unhelpful ◯ Not tried

Relaxing ◯ Yes, helpful ◯ No, unhelpful ◯ Not tried

Counting to ten ◯ Yes, helpful ◯ No, unhelpful ◯ Not tried

Covering your face with hands ◯ Yes, helpful ◯ No, unhelpful ◯ Not tried

I. Case Summary ** COMPLETED BY ASSESSOR **

(i) Considering the participants presentation and everything they have said **regarding the past two weeks**, how do they classify:

◯ No evidence of post-stroke emotionalism

◯ Evidence of post-stroke emotionalism, crying component only

◯ Evidence of post-stroke emotionalism, laughter component only

◯ Evidence of post-stroke emotionalism, crying and laughter components

******SWITCH OFF DIGITAL RECORDER NOW******

Appendix N: TEARS Participant Information Sheet

INFORMATION SHEET FOR PARTICIPATION IN CLINICAL RESEARCH PROJECT

Title of project

Testing for Emotionalism After Recent Stroke (TEARS)

You are being invited to take part in a study of uncontrollable crying or laughing due to stroke. We think emotionalism after stroke is common, but we cannot be sure as few studies have looked at it.

Before you decide, it is important that you fully understand why the research is being done and what is involved. The study is funded by the Stroke Association and is led by XXX and XXX.

Please take time to read the following information carefully. Feel free to ask the Stroke Research Nurse any questions if anything is unclear.

Thank you.

Purpose of study

You have been diagnosed as having had a stroke. Sometimes stroke causes changes to emotions (that is, how we feel inside) or emotional expression (that is, how we show our emotions - in particular through crying and laughter). This is known as Post-Stroke Emotionalism. We think Emotionalism after stroke is common, but we cannot be sure because only a few studies have looked at it.

To find out, we are studying a large group of people with stroke starting up to two weeks after their illness, and following them up on two further occasions, at six months and one year. On each occasion participants will complete assessments of mood and emotions, quality of life, thinking skills and social/physical functioning. A relative, nominated by you, will also complete some assessments. This will help us work out how common emotionalism is, what contributes to persistent emotionalism, and how emotionalism can impact quality of life, mood and thinking skills.

Why have I been chosen?

All patients diagnosed with a stroke will be considered for participation. People who agree to

Figure 1: Flowchart displaying participants who continue to have post-stroke emotionalism (PSE) at subsequent time-points and new participants who are identified as having PSE.

from X

D

N : part you will be given this information sheet to keep and asked to sign a consent form. For any person given study information to consider taking part who is then discharged home, we would telephone them to see if they want to take part, if they agree to this call.

You are free to withdraw at any time and do not have to give a reason. This will not affect the standard of care you receive. If you decide to withdraw, any test scores obtained from you would be kept, unless you did not wish this information to be included in the final study analysis.

The researchers are not involved with your general care. The results of the tests will be shared with the team looking after you in the stroke ward and with your Family Doctor.

If during the study, we felt that you would no longer be able to decide for yourself whether to continue taking part, we would ask a relative or friend to consent on your behalf to remain in the study. We would ask them to set aside their own views and consider your

interests, wishes and feelings. Any advance decisions you may have made that they are aware of would take precedence.

What will happen to me if I take part?

This study will involve three assessments, each comprising a sound (audio) recorded interview, questionnaires and paper and pencil tests of mood and emotions, quality of life, thinking skills and social/physical functioning.

The assessments will occur on three separate occasions across one year, with a Stroke Research Nurse. The first assessments will take place face-to-face up to two weeks after the stroke while you are still in hospital and include a sound (audio) recorded interview. Some participants will be asked to complete a second face-to-face interview about mood and emotions since the stroke, whilst in hospital.

The next set of assessments will be six months after face to face, again at the hospital and including a sound recorded interview or at home if you cannot manage to the hospital. After the six months assessment, you will be given one short questionnaire to complete again at home. The nurse will call you to remind of this. The final assessment will be twelve months after, face to face. We will pay your taxi fares to make these hospital trips, and provide light refreshments.

We will also ask you nominate a close relative/friend to answer questions about your memory and thinking skills before the stroke, about their perspective of any emotionalism symptoms you may have and about caring for you. These will take around 15 minutes. If your relative has consented to help but we cannot locate them, we may leave you some questionnaires in a stamped addressed envelope to give to them, so they can be completed at home and sent back.

The results of the first assessment will be shared with the ward team and your family doctor. The results of the other assessments with be shared with your family doctor.

Assessment

The study will involve assessments of mood and emotions, quality of life, thinking skills and social/physical functioning.

During each assessment, we will interview you, asking questions about your emotions since the stroke and how controllable these are. Interviews will be sound recorded. There will also be a questionnaire about your emotions and emotional control, and a questionnaire about mood and anxiety. There will be a questionnaire about your quality of life since the stroke. There will be a questionnaire about your physical functioning since the stroke. We will ask about your participation in social activities. There will be a paper and pencil test of thinking and memory. Specific instructions for the questionnaires and tests will be given when they are administered.

In total the assessment sessions will take approximately 80 minutes. We will break things into two sessions of 40 minutes, with refreshments. All assessments will be administered by the Stroke Nurse.

A relative, nominated by you, would also answer questions about your memory/thinking skills before the stroke, about any emotionalism symptoms and about caring for you. This will take them around 15 minutes.

To check for any additional medical problems or changed medicines you may have, the stroke nurse will contact your Family Doctor around the time of the six and twelve month assessments and ask you about your medicines and any medical events that may have happened.

What do I have to do?

The study will involve assessments of mood/emotions, quality of life, thinking skills and social/physical functioning. If you agree to participate you will complete the assessments on three occasions across a one year period. If you participate you will complete the first interview/set of assessments whilst in hospital and the following two interviews/sets of assessments back at the hospital. There is the option that the nurse can visit you at home to do the assessments, if you cannot manage into the hospital. We will pay all taxi fares for you, for research visits to and from the hospital. There are no other requirements.

What are the possible benefits to taking part?

There are no direct benefits to you of taking part. However by taking part you will help us improve our understanding of how common emotionalism is after, how it affects mood, thinking skills, quality of life and physical/social functioning. We hope the study will also improve our management of emotionalism problems in the future using talking therapies (psychology) rather than pills.

What if something goes wrong?

If any problems are encountered during assessment that cannot be resolved for example you suddenly become unwell, assessment will be ended and where necessary a member of the treating clinical team will be immediately notified. You have the right to withdraw from the study at anytime without providing a reason and with no impact to the standard of care you receive. If you are unhappy about any aspect of the study and wish to make a formal complaint, please contact the lead researcher in the first instance but the normal NHS complaints mechanism is also available to you.

What will happen to the results of the study?

The results of your assessments will be shared with the ward clinical team and your family doctor. We will publish the final results in a scientific journal and discuss the study at stroke professional meetings. Your personal details will not be available in any of these materials. If you are interested in the results when the study is complete, details can be posted to you.

Confidentiality

Your scores on the assessments will be shared with the ward clinical team and your Family Doctor. All Personal information collected by the research team will be anonymised and stored in a secure way.

Part of the study assessment includes asking about depression. If we suspected severe depression or suicidal thoughts during the assessments at hospital, the assessment could be stopped if you wished. We would let your Stroke Doctor know, so they could check on your mood and ensure you were safe and had any supports needed. They may choose to refer you for specialised help, to get additional support. If we suspected severe depression or suicidal thoughts during the telephone assessment, the assessment could be stopped if you wished. We would let your Family Doctor know, so they could check on your mood and ensure you were safe and had any supports needed. Your Family Doctor may choose to refer you for specialised help, to get additional support.

Who is organising and funding the study?

The study is funded by the Stroke Association and organised by the Institute of Cardiovascular and Medical Sciences University of Glasgow. The researchers will receive no remuneration for including you in the study.

Who has reviewed this study?
This study has been reviewed and approved by an independent research ethics committee, the Scotland A Research Ethics Committee.

SUMMARY
You are being invited to take part in a clinical research study of uncontrollable crying or laughing due to stroke. If you agree to participate you will complete assessments of your mood and emotions, quality of life, thinking skills and social/physical functioning, on three occasions in a one year period. A relative, nominated by you, would answer questions about your emotions, memory and impact of caring for you.

Name of Researchers
XXX
XXX

Name of sponsors
NHS Greater Glasgow and Clyde

Appendix O: TEARS Consent Forms

[redacted]

NHS Greater Glasgow and Clyde

Name of Researchers

Dr Niall M Broomfield, Consultant Psychologist in Stroke NHS Greater Glasgow and Clyde and Honorary Clinical Teacher, University of Glasgow.

[redacted]

Name of sponsors

NHS Greater Glasgow and Clyde

Testing for Emotionalism After recent Stroke (TEARS): CONSENT FORM

Study number:

Subject Number:

Ward Recruited (tick one): [redacted] ☐ [redacted] ☐ [redacted] ☐

[redacted] ☐ [redacted] ☐ [redacted] ☐

[redacted] ☐

Please read the information below, sign and date if in agreement **Please initial the BOX**

1. I confirm that I have read and understood the information sheet version 7 dated 20/05/16 for the above study and have had the opportunity to ask questions. ☐

2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason and without my medical care or rights being affected. ☐

3. I understand that sections of my medical notes and data collected during the study may be looked at by the researchers named above and/ or by the study sponsor or the regulatory authorities where it is relevant to my taking part in this research. ☐

4. I understand and agree to the interview component of the assessment being sound (audio) recorded. ☐

[redacted]

NHS Greater Glasgow and Clyde

5. I understand that results will be used for research purposes and shared with the clinical team. ☐

6. I agree to my GP or other care professional being informed of my participation in the study ☐

7. I understand that in the event that suicidal thoughts or concerns regarding severe depression are disclosed to the researcher then the clinical team will be immediately informed regardless of patient preference. ☐

8. I agree to take part in the above study. ☐

Name of Patient	Date	Signature
Name of person taking consent	Date	Signature
Name of person taking consent (If different from the researcher)	Date	Signature

1 copy to the patient, 1 copy to the Stroke Research Nurse, 1 Original for the patient's notes

[redacted]

NHS Greater Glasgow and Clyde

Testing for Emotionalism After Recent Stroke (TEARS)

CONSENT FORM FOR GUARDIAN, WELFARE ATTORNEY, OR NEAREST RELATIVE

Study number:

Subject Number:

Ward Recruited (tick one): [redacted] ☐ [redacted] ☐ [redacted] ☐

[redacted] ☐ [redacted] ☐ [redacted] ☐

[redacted] ☐

Please read the information below, sign and date if in agreement	**Please initial the BOX**
1. I confirm that I have read and understood the information sheet version 6 dated 20/05/16 for the above study and have had the opportunity to ask questions.	☐
2. I understand that consent is voluntary and that I am free to withdraw it at any time without giving any reason and without medical care or rights being affected.	☐
3. I understand that sections of my family member/friends medical notes and data collected during the study may be looked at by the researchers named above and/ or by the study sponsor or the regulatory authorities where it is relevant to my family member/friend taking part in this research.	☐
4. I understand that results will be used for research purposes and shared with the clinical team.	☐
5. I agree to my family member/friends GP or other care professional being informed of their participation in the study	☐
6. I understand that in the event that suicidal thoughts or concerns regarding severe depression are disclosed to the research nurse then the clinical team will be immediately informed regardless of my family member/friends preference.	☐
7. I agree to my family member/friend taking part in the above study.	☐

[redacted]

NHS Greater Glasgow and Clyde

Relationship to patient _____

Name of person giving consent	Date	Signature
Name of person taking consent	Date	Signature
Name of person taking consent (If different from the researcher)	Date	Signature

1 copy to the patient, 1 copy to the researcher, 1 Original for the patient's notes

[redacted]

NHS Greater Glasgow and Clyde

Testing for Emotionalism After Recent Stroke (TEARS)

CONSENT FORM FOR CARER

Study number:

Subject Number (Participant):

Ward Recruited (tick one): [redacted] ☐ [redacted] ☐ [redacted] ☐

[redacted] ☐ [redacted] ☐ [redacted] ☐

[redacted] ☐

Please read the information below, sign and date if in agreement **Please initial the BOX**

1. I confirm that I have read and understood the information sheet version 5 dated 16/11/2015 for the above study and have had the opportunity to ask questions. ☐

2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason and without my family member/friend's medical care or rights being affected. ☐

3. I understand that results will be used for research purposes and shared with the clinical team. ☐

4. I agree to take part in the above study. ☐

| --- | --- | --- |
Name of carer/friend/relative	Date	Signature

[redacted]

NHS Greater Glasgow and Clyde

Name of person taking consent	Date	Signature
---	---	---
Name of person taking consent (If different from the researcher)	Date	Signature

[redacted]

NHS Greater Glasgow and Clyde

Consent Form

TEARS Measuring uncontrolled crying after a stroke.

Principal Investigator: Dr Niall Broomfield, Clinical Psychologist in Stroke

	Yes 	No 
I have been given **information** about the study and read information sheet version 4 dated 20/05/2016.		
I have **talked** to _____ about the study.		
My **questions** have been **answered.**		
I **understand** what is involved.		
I understand my **name** will **not** be **used**.		
I understand the interview will be **sound** recorded.		
I **agree** to take part in the study/tests involved.		
I understand I can **stop** the study/tests at any time.		

[redacted]

Name:……

Signed ……

Witness: ……

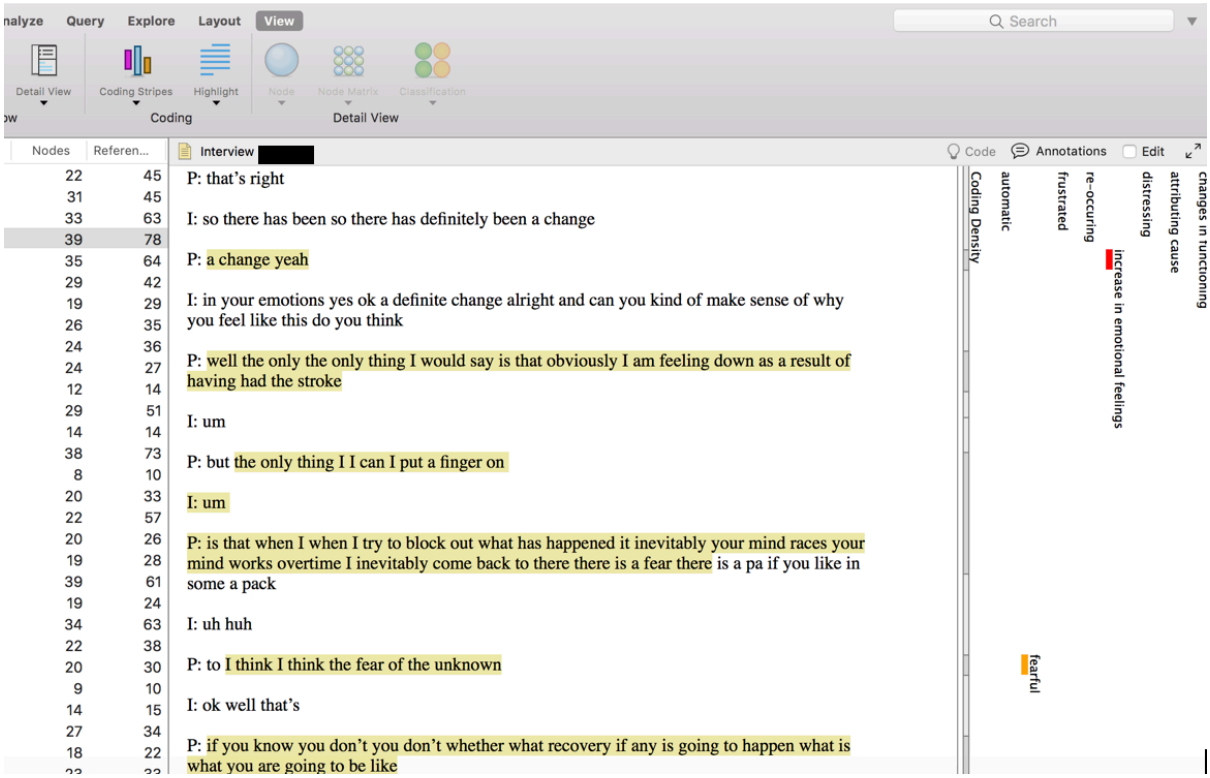
Signed (researcher): …… Date: ……

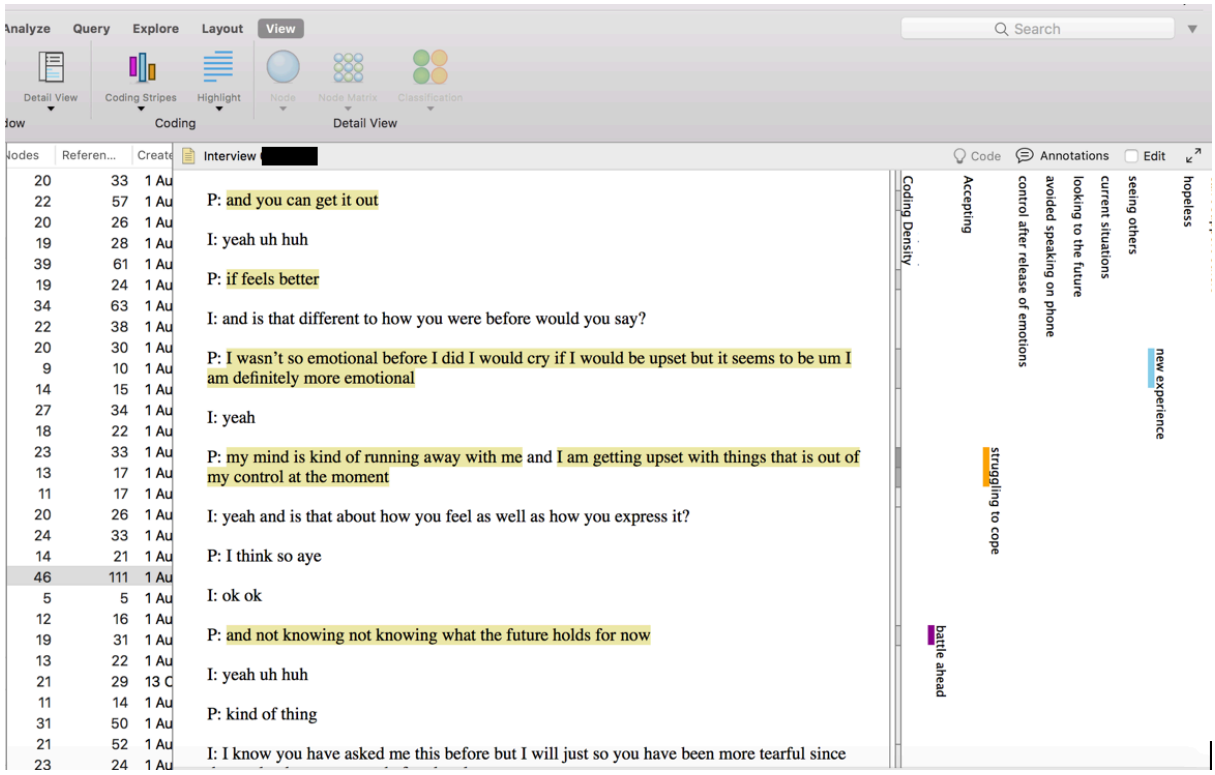
Appendix P: Overview of changes in the frequencies of codes overall for each sub-theme

Table 7. Overview of changes in frequency of references overall for each theme and sub-theme.

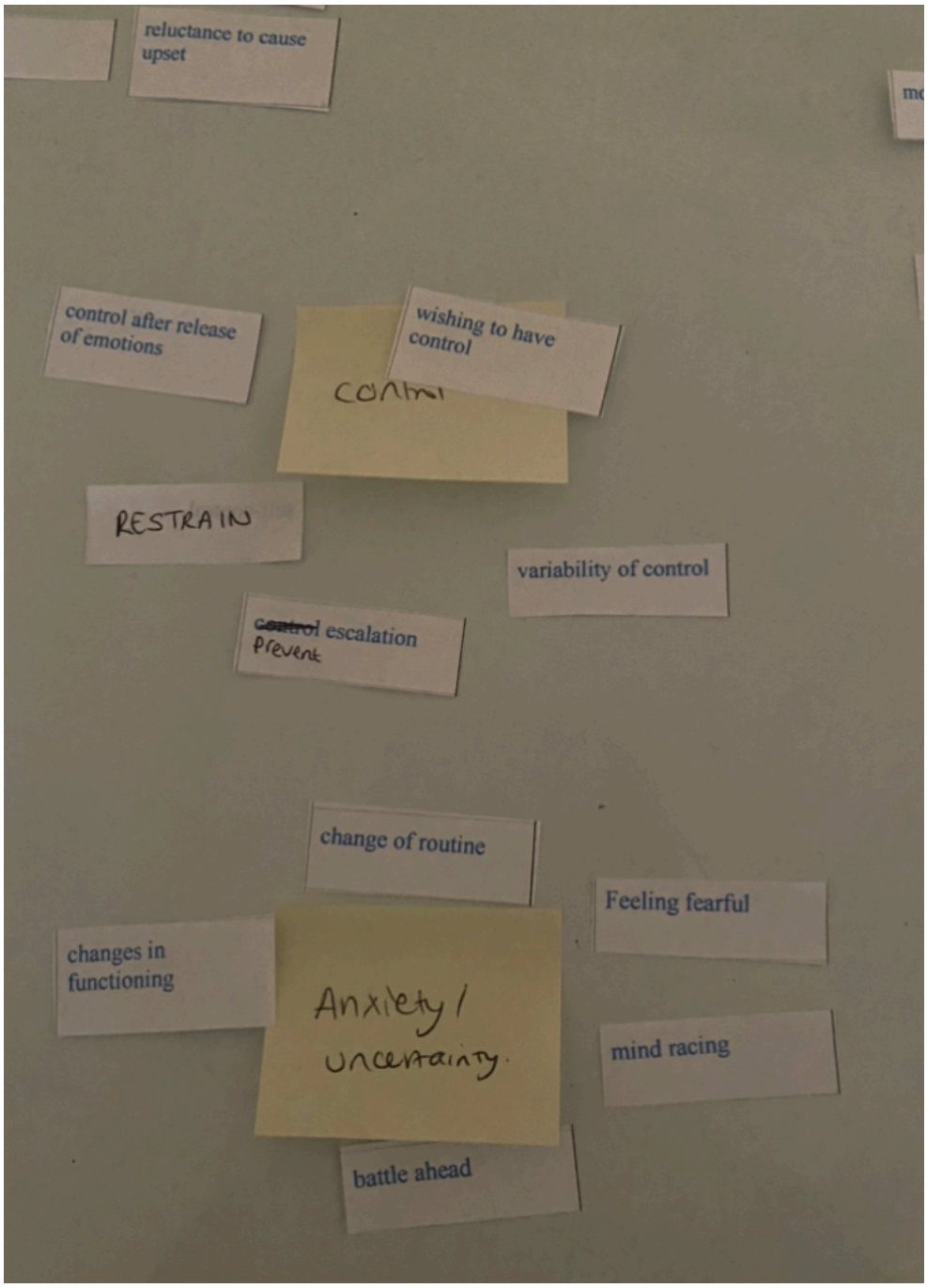
Themes and sub-themes	**2-weeks N=52**		**6-months N=25**		**12-months N=23**	
	Frequency of references	Unique participants n=	Frequency of references	Unique participants n=	Frequency of references	Unique participants n=
1. In the moment						
a. Characteristics	136	47 (90%)	49	18 (72%)	67	19 (82%)
b. Control	35	22 (42%)	27	13 (25%)	16	11 (47%)
c. Barriers to control	95	35 (67%)	44	14 (56%)	35	15 (65%)
d. External triggers	177	45 (86%)	46	15 (60%)	30	18 (78%)
e. Internal triggers	72	33 (63%)	20	9 (36%)	23	11 (47%)
f. Predictability	16	12 (23%)	9	6 (24%)	24	11 (47%)
g. Unpredictability	34	24 (46%)	15	10 (40%)	22	11 (47%)
2. Ways of coping						
a. Acceptance	43	24 (46%)	18	9 (36%)	18	9 (39%)
b. Avoidance of triggers	53	26 (50%)	39	12 (48%)	65	16 (69%)
c. Concealing emotionalism from others	42	25 (48%)	17	10 (40%)	34	17 (73%)
d. Behavioural attempts at symptom management	54	26 (50%)	49	13 (52%)	58	15 (65%)
e. Cognitive attempts at symptom management	82	30 (57%)	43	17 (68%)	28	8 (34%)
f. Support from others	65	29 (55%)	12	6 (24%)	18	7 (30%)
3. Impact						
a. Changes to self	106	37 (71%)	48	16 (64%)	72	20 (86%)
b. Emotional impact	93	38 (73%)	66	17 (68%)	63	19 (82%)
c. Trying to make sense	30	19 (36%)	5	5 (20%)	12	7 (30%)
d. Uncertainty	67	28 (53%)	37	12 (48%)	19	8 (34%)

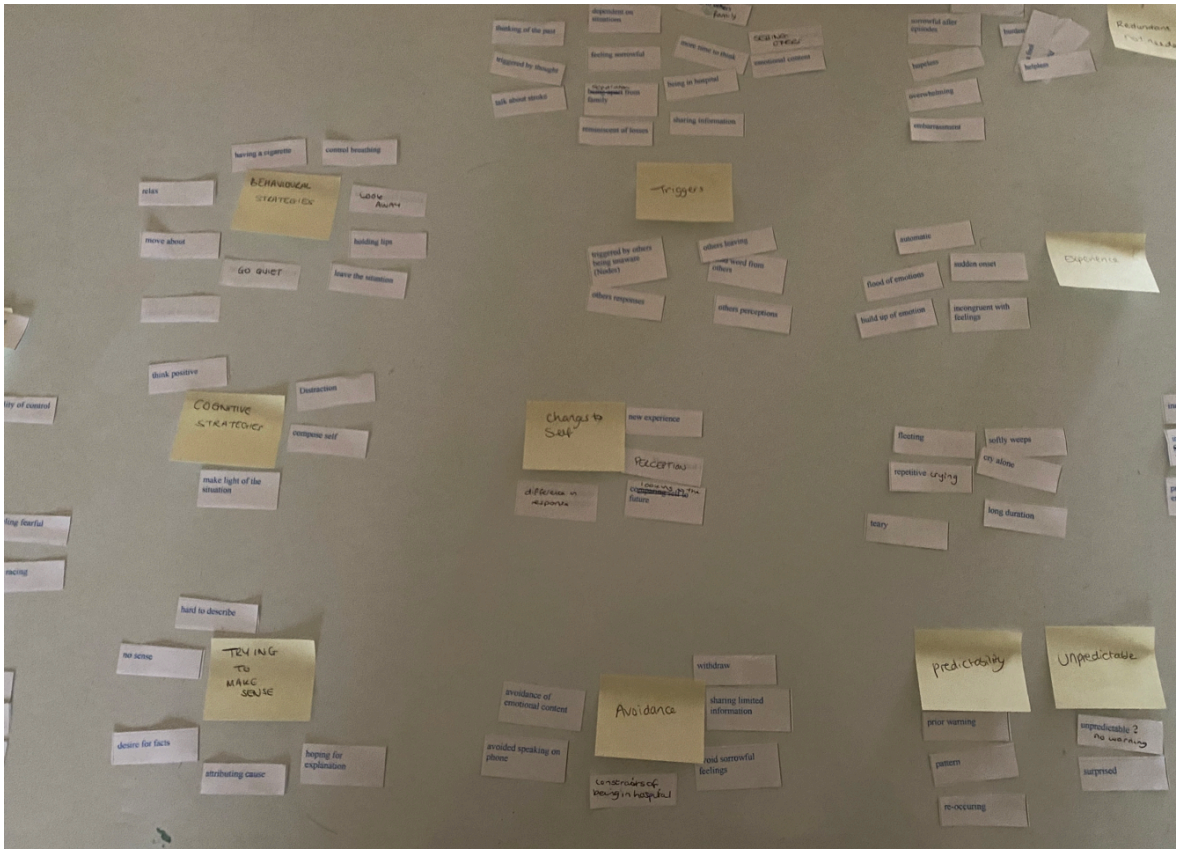
Appendix Q: Example of coding interview transcripts





Appendix R: Example of provisional compilation of codes into sub-themes and major themes





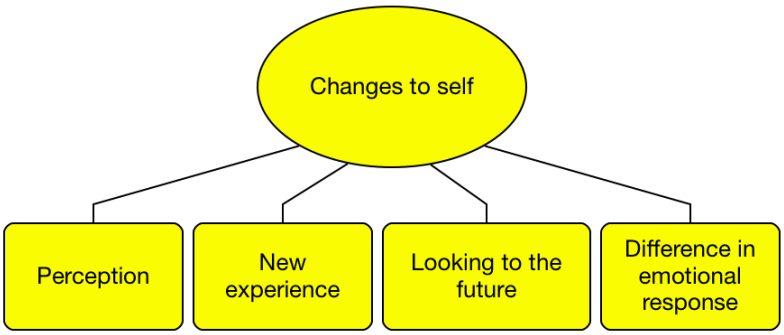


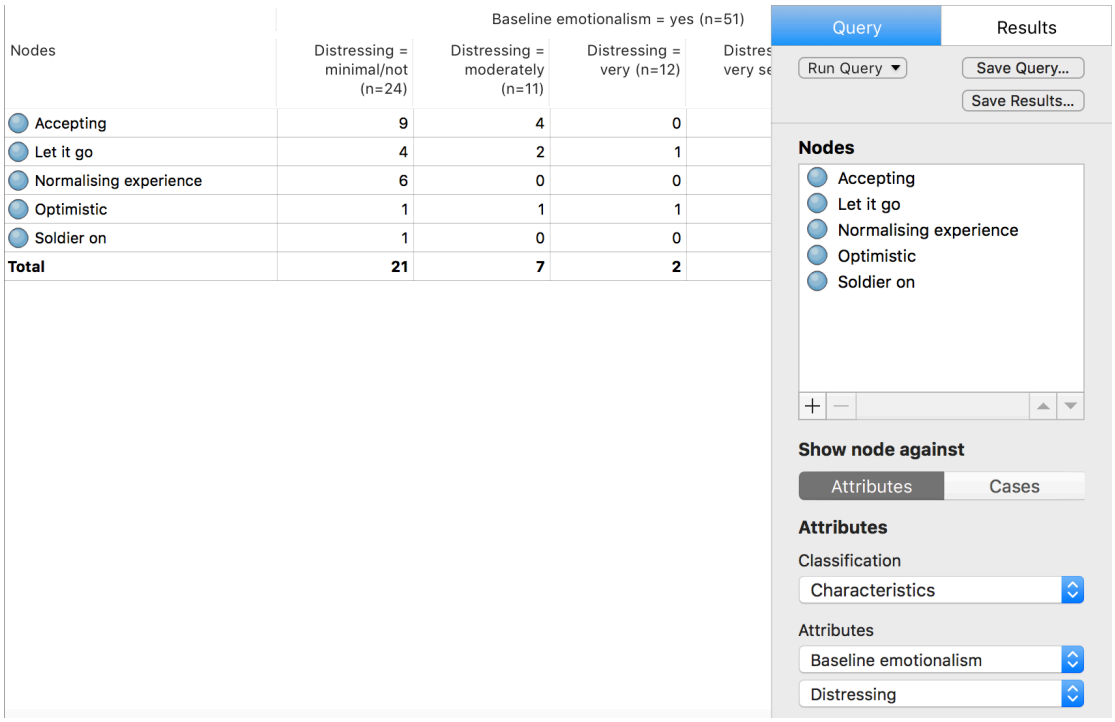
Figure 1: Example major theme with sub-themes

Appendix S: Developing an analytical framework which was applied to several transcripts

Table 8: Example of the developing framework for the theme 'barriers to control' which was starting to be applied to transcripts

	Difficult to regulate	Out of the blue	Struggling to cope
Participant 012	*"It's just stopping it once it starts it is quite difficult"*		*"I get myself into a state"*
Participant 016	*"I can't do anything to stop"*	*"I am just sitting on a bus and before I know it I am starting to cry for no reason at all"*	
Interview 029	*"I just started crying and I couldn't stop myself"*	*"Get taken unaware…I am not prepared for it"*	*"I am in a state…I just fell apart… but now I just can't seem to cope with the situations"*

Appendix T: Trajectory analysis of interview data



Nodes	Baseline emotionalism = yes (n=51)			
	Distressing = minimal/not (n=24)	Distressing = moderately (n=11)	Distressing = very (n=12)	Distres very se
Accepting	9	4	0	
Let it go	4	2	1	
Normalising experience	6	0	0	
Optimistic	1	1	1	
Soldier on	1	0	0	
Total	**21**	**7**	**2**	