Article:
Nielsen, G. orcid.org/0000-0001-6053-5670, Stone, J. orcid.org/0000-0001-9829-8092, Lee, T.C. orcid.org/0009-0009-0917-860X et al. (68 more authors) (2024) Specialist physiotherapy for functional motor disorder in England and Scotland (Physio4FMD): a pragmatic, multicentre, phase 3 randomised controlled trial. The Lancet Neurology, 23 (7). pp. 675-686. ISSN 1474-4422

https://doi.org/10.1016/s1474-4422(24)00135-2

Keywords: shame; seizures; anxiety; depression; somatic symptoms

1. Introduction

Shame is a distressing emotion characterized by self-perceptions of being flawed and worthless, and accompanied by behavioural impulses to retreat, withdraw, and vanish [1]. This complex emotion is intricately linked to our social identity and evolves from infancy through our interactions with others, commencing with our early attachments to primary caregivers [2]. Caregivers who lack emotional attunement can make children more vulnerable to developing a predisposition for maladaptive shame [1, 2]. Moreover, exposure to discrimination, stigma, and traumas can contribute to high levels of shame in people at any stage in life [3, 4]. If shame is triggered too easily, frequently or intensely, it can increase associated maladaptive behavioural tendencies, which in turn can lead to impaired psychological functioning [5]. Corresponding to this, maladaptive shame has been linked to low self-esteem [6], interpersonal problems [2], various mental health difficulties such as anxiety, depression, borderline personality disorder [7-9], as well as physical health difficulties [10]. Shame has been considered as a transdiagnostic treatment target in psychological therapeutic approaches, such as in Compassionate Focused Therapy [11, 12].

Despite previous findings that individuals with seizure disorders often experience factors associated with shame, such as high levels of stigmatization [3, 13] and trauma [14, 15], there has been a lack of research examining the role of shame in these disorders. More specifically, it has recently been argued that maladaptive shame may be a key factor in the development and maintenance of functional seizures [5, 16]. This highlights the need for further investigation into the experiences of individuals with seizures and the potential impact of shame on their well-being. Therefore, this study was designed to examine levels of shame and mental health difficulties (MHDs) in persons with functional (PWFS) and epileptic seizures (PWE); as well as to explore whether shame is associated with MHDs and seizure frequency or severity in the two different seizure disorders.

1.2. Functional seizures (FS) and shame

Functional seizures (FS, also known as psychogenic non-epileptic seizures or dissociative seizures) are a subtype of functional neurological symptom disorder [17]. FS are episodes of disruption of normal functioning involving the temporary reduction of consciousness and self-control and a range of involuntary motor, sensory and mental manifestations [18]. FS superficially resemble epileptic seizures (ES) or syncope, but unlike these states, FS are not associated with pathophysiological abnormalities sufficient to explain the alteration of neurological functioning.

The underlying mechanisms of FS are complex and contentious. Brown and Reuber [18] developed the Integrative Cognitive Model, which posits that functional seizures are automatised behavioural responses to physiological and mental arousal. They describe that the key vulnerability to functional seizures includes inhibitory dysfunction arising from chronic stress, whilst potential trigger factors include emotions associated with physiological arousal [18]. Since the majority of people with functional seizures have experienced traumatic experiences [19] and subsequent stigma related to their condition [20] this may lead to them experiencing increased levels of shame. Given that shame is frequently an overwhelming emotion [5], it has the potential to intensify symptoms of psychopathology, elevate arousal levels, and impede the efficacy of behavioural inhibition capable of halting functional seizures. Moreover, shame can directly serve as a trigger for functional seizures [5][18].

1.3. Epileptic seizures and shame

While a secondary emotion like shame would not be considered as a primary cause of epileptic seizures, shame may still play an important role in epileptic seizure disorders as well. Similarly to people with functional seizures, PWE often face stigma [3] and experienced traumatic events [21]. Moreover, chronic stress has been linked to an increased likelihood of developing or triggering epileptic seizures [22, 23]. Whilst maladaptive shame has not yet been examined in PWE, previous research found that PWE experience high levels of self-disgust [24].

1.4 Mental health difficulties in people with ES and FS

Since the ictal manifestations of functional seizures resemble epileptic seizures and patients often present to clinicians in similar ways, much previous research has compared levels of MHDs in people with these two types of seizures. In these studies, PWFS tend to report more traumatic experiences, have

experienced greater levels of childhood abuse and neglect, are more likely to have fearful attachment styles and report more somatic complaints and dissociative experiences, than people with epilepsy and healthy controls [14, 19, 25]. However, to the authors' knowledge there has not yet been a study, that measured and compared levels of shame between PWFS and PWE.

1.5 Measuring shame, a complex emotion

When measuring shame in people with seizures, it is important to consider that it a complex and multifaceted emotion [26]. Most previous research has focused on *shame proneness*, defined as the tendency to experience shame readily and intensely across different situations [27-31]. However, more recent studies have also examined *shame aversion*; that is, the appraisal of shame as especially painful and intolerable [32]. Schoenleber and Berenbaum [32] proposed that those who are more averse to experiencing shame may be more likely to engage in unhelpful behaviors to regulate their shame, compared to those who are simply prone to feeling shame. In support of this, they found that shame aversion was a stronger predictor of a range of mental health difficulties, than shame proneness [33-35]. Hence, these two dimensions of shame will be considered in this study.

Additionally, it is worth considering that demographic and social factors that can affect levels of shame. Lower socio-economic status has been associated with feelings of "internalised inferiority" and shame [36]. This is not surprising, as shame is an emotion that is triggered by threats to one's social self or status [12]. Furthermore, previous studies have consistently shown that shame proneness is higher in females [26, 37], while older individuals tend to have lower levels of maladaptive shame [37].

1.6 Research questions

The observations on the significant impact of shame on health and mental health provide [7-10] a rationale for studying shame in both epileptic and functional seizure disorders and for exploring whether shame contributes to current (adulthood) MHDs. The study was designed to test the hypothesis, that 1) both PWE and PWFS would experience high levels of shame, but PWFS would experience more shame proneness, shame aversion, somatic symptoms, anxiety, and depression, than PWE; and 2) shame aversion and shame proneness would predict anxiety, depression, somatic symptoms, seizure severity and seizure frequency in both groups, but we expected the association to be stronger in the functional seizure group.

2. Methods

This comparative cross-sectional online study included 138 participants, PWFS and PWE. Ethical and Health Research Authority approval (Reference number: 22/YH/0213) was obtained for the study. Participation was voluntary, and participants provided written informed consent before taking part.

2.1. Participants

Participants were recruited from outpatient neurology clinics at a South Yorkshire hospital in the United Kingdom by Consultant Neurologists. Membership-led organizations for individuals experiencing epileptic or functional seizures also advertised the study on their online platforms. Recruitment took place between December 2022 and March 2023.

Participants were included if they were a) 18 years old or older; b) could read and write in English c) could complete the online questionnaire without help; d) had a self- declared diagnosis of functional OR epileptic seizures as confirmed by a physician (e.g., neurologist or psychiatrist). Participants who had both epileptic and functional seizures were excluded.

2.2 Power analysis

A priori power analysis was conducted using the 'G*Power 3' software [38] to attain the minimum sample size required to find an effect, suggesting a total sample size of 134 to find a small effect (f2 = 10) with 80% power.

2.3 Procedure

The survey ran on Qualtrics [39], an online data collection platform. Participants could complete the online survey in their home environment after accessing link from researchers or from the study advertisement sheet. First, participants were presented with an information sheet, a consent form, and a screening questionnaire. Thereafter, eligible participants could complete the questionnaire battery.

2.4 Measures

2.4.1 Demographic questionnaire

Information about participants' age, gender, ethnicity, educational and employment status was collected.

2.4.2 Perceived socioeconomic status (PSES)

2.4.3 PSES was measured as a visual scale which asks participants to indicate where they think they stand on the socioeconomic ladder [40] in comparison to others. The instructions were as follows: "At the top of the ladder are the people who are the best off, those who have the most money, most education, and best jobs. At the bottom are the people who are the worst off, those who have the least money, least education, worst jobs, or no job. Please place an 'X' on the rung that best represents where you think you stand on the ladder." The higher participants placed themselves on the ladder the higher they scored.Shame proneness

The short version of the Test of Self-Conscious Affect Scale (TOSCAS-3) [41] was used to measure internalised and global shame. The short version of TOSCA-3 includes 11 negative scenarios that yield six subscales of guilt-proneness, shame-proneness, detachment, and externalisation. Items are scored on a 5-point Likert-type scale, with higher scores represent higher shame proneness. The mean shame proneness score is calculated by the average shame proneness total score of participants divided by 11. The Cronbach's alpha for the TOSCA-3 was good (α =.80).

2.4.4 Shame aversion

The Shame-Aversive Reactions Questionnaire (ShARQ) [8] assessed aversion to shame. The ShARQ uses a 7-point Likert-type scale and includes 14 items. Higher mean scores on the ShARQ indicated higher levels of shame aversion. The ShARQ had good internal consistency (Cronbach's α = .89).

2.4.5 Seizure severity

The Liverpool Seizure Severity Scale (LSSS-2) [42] was used to measure recent seizure severity. Those participants who did not have seizures in the last 4 weeks could not complete the rest of the LSSS-2 questionnaire (functional group N = 9; epilepsy group N = 34). All 12 items on the LSSS-2 are scored on a Likert scale, with higher scores indicating greater seizure severity. The LSSS-2 had good internal consistency (Cronbach's α = .76)

2.4.6 Seizure frequency

This scale required participants to choose between five options that best describe the frequency of their seizures over the last year. The options were: 1) I have not had any seizures in the last year; 2) I usually have more than one seizure per year but fewer than one seizure per month; 3) I usually have more than one seizure per month but fewer than one seizure per week; 4) I usually have more than one seizure per week but fewer than one seizure per day; 5) I usually have more than one seizure per day. Participants were assigned a score from 1 (least frequent) to 5 (most frequent), with higher scores indicating higher frequency of seizures.

2.4.7 Depression.

The 8-item version of the Patient Health Questionnaire (PHQ-8) [43] was used to assess depression severity. Items were scored from 0 (not at all) to 3 (nearly every day); higher scores indicate higher levels of depression. The PHQ-8 can be used as a depression measure for population-based studies, with a score of ≥10 suggesting current clinical depression [43]. Internal consistency of the item PHQ-8 was excellent (Cronbach's α of α = 0.90).

2.4.8 Anxiety

The 7-item Generalized Anxiety Disorder scale (GAD-7) [44] assesses overall severity of anxiety. Each item is scored on a four-point Likert scale (0–3), with total scores ranging from 0 to 21, with higher scores reflecting greater anxiety severity. A GAD-7 score of ≥10 suggests clinical levels of Generalised Anxiety Disorder [44]. Internal consistency of the GAD-7 was excellent (Cronbach α = .92)

2.4.9 Somatic symptoms

The 8-item Somatic Symptom Scale-8 (SSS-8) [45] assesses somatic symptom burden, with a 5 point Likert-type scale ranging from 0 (not at all) to 4 (very much). The total score ranges from 0 to 32, with higher scores indicating a greater somatic symptom burden. Cut-off scores indicate no or minimal (0-3 points), low (4- 7 points), medium (8-11 points), high (12-15 points), and very high (16-32 points) somatic symptom burden. The SSS-8 had good internal consistency (Cronbach α = 0.81).

2.5 Data analyses

IBM SPSS Statistics for Windows (Version 28; 2021) was utilised for the statistical analyses. A preliminary analysis was carried out to test differences between groups on demographic and condition-specific variables using t-tests (for continuous parametric variables), Mann-Whitney U tests (for continuous non-parametric variables), or Chi-Square test (for categorical variables) (Table 1.). Preliminary Pearson's and Spearman's correlation analyses were performed to test the strength of association between variables (Table 2). Hypothesis 1. was tested using five Student's t-tests analyses (Table 3). The variables shame proneness

and shame aversion showed a negatively skewed distribution. Therefore, both variables were transformed by reflecting and raising them to the square root. After that, all assumptions for the t-tests were met. Hypothesis 2, was tested with five multiple hierarchical regressions (Table 4, 5). All assumptions for the hierarchical regression analyses were met. For each five hierarchical regression analyses, in Step 1, socioeconomic status, age, and gender were added as covariates. In Step 2, the variables shame aversion, shame proneness, and 'group membership' were added (the latter being a dummy coded variable involving epilepsy d = 0 or functional d = 1 seizure groups). Finally, two moderators were entered in Step 3. The moderator variables were created from the product of the standardised independent variables (shame aversion, shame proneness) and the standardised dichotomous dummy coded group variable (epilepsy vs functional seizure group). To reduce multicollinearity, all moderator variables and independent variables were standardised (except dummy variables) [46].

3. Results

Of 167 eligible participants who began to complete the questionnaires, 29 were excluded as missed at least one questionnaire (other than being excluded from the seizure severity questionnaire). See Table 1. for the included (n = 138) participant characteristics.

3.1. Demographic and Condition Specific variables

In total, 138 participants were included in the study (age range 18-67; mean age 38.7; SD = 12.2). Twelve participants (7.2%) were recruited from hospital clinics and the rest through charities. Most participants were female (82.6%) and white (90%). In the ES group (n = 70, male = 16, female = 54), most participants were British (92.9%), whereas the FS group (n = 68, male = 8; female = 59, non-binary = 1) was about half British (54.4%) and half international (45.6%). All international participants came from either English speaking (USA, Australia, Canada, New-Zealand) or high-income European countries. All participants had at least secondary education, and about half had a university degree. In the ES group, 71.4% indicated being employed or studying full-time, and 12.9% indicated being on sickness/disability leave, whereas the corresponding rates were 30% and 57.3% in the FS group, respectively. The FS group perceived their socioeconomic status as significantly lower than the ES group ($p < .001$). The FS group had significantly more frequent seizures than the ES group ($p < .001$). In the last four weeks, 69% of participants in the FS group had at least one seizure, in contrast to 51% in the ES group. People with ES reported significantly stronger seizures, than people with FS ($p < .001$). Table 2 shows that, the FS group mean score met the diagnostic cut-off level for anxiety and depression, and the ES group mean score the diagnostic cut-off for depression. People with ES reported high, and the FS group very high levels of somatic symptoms.

3.2. Hypothesis 1. T-tests to compare levels of MHDs and shame between group

The FS group had significantly higher depression ($t (136) = -3.61$; $p < .001$) and somatic symptoms scores than the ES group ($t (136) = -5.31$; $p < .001$) (Table 2). However, t-tests showed no differences between groups in the levels of anxiety ($t (136) = -1.29$; $p = .1$), shame proneness ($t (136) = .35$;.36) and shame aversion ($t (136) = -.14$; $p = .45$).

3.3. Correlation analyses

A preliminary correlation analysis was performed to test the strength of associations between study variables for both groups combined. Shame aversion correlated significantly positively with depression ($r = .491$; $p < .01$), anxiety ($r = .527$; $p < .01$), somatic symptoms ($r = .260$; $p < .01$), seizure severity ($r = .238$; $p = .02$), but not with seizure frequency ($r = -.043$) (Table 3). There was a significant positive correlation between shame proneness and depression ($r = .389$; $p < .01$), anxiety ($r = .396$; $p < .01$), and somatic symptoms ($r = .280$; $p < .01$), but shame proneness did not correlate significantly with seizure frequency ($r = .028$) and severity ($r = .109$). There was a strong positive association between shame aversion and shame proneness ($r = .628$; $p < .01$).

3.4. Hypothesis 2. Moderated hierarchical regression analyses testing the association between shame, and MHDs & seizures

To examine whether shame predicted MHDs, seizure frequency and seizure severity six moderated hierarchical regressions were performed (Table 4, 5). These analyses revealed that shame aversion predicted significant variance in depression ($\beta = .39$, $p < .01$) and anxiety ($\beta = .47$, $p < .001$) after controlling for perceived socioeconomic status, age, and gender variables. However, shame aversion did not predict variance of somatic symptoms ($\beta = .15$, $p = .13$), seizure frequency ($\beta = -.11$, $p = .27$), and seizure severity ($\beta = 0.15$, $p = .25$). Shame proneness did not predict variance beyond that was explained by perceived socioeconomic status for depression ($\beta = .07$, $p = .50$), anxiety ($\beta = .15$, $p = .16$), somatic symptoms ($\beta = .13$, $p = .23$), seizure severity ($\beta = -.04$, $p = .77$), and seizure frequency ($\beta = .00$, $p = .97$).

The moderation analyses indicated that there was no interaction effect between 'group and shame proneness' and 'group and shame aversion' variables for any of the dependent variables (See Step 3 analyses Beta results in Table 4 & 5). This means the strength of the relationship between shame variables and MHDs or seizure variables was not stronger in the PWFS, than the PWE group.

Perceived socioeconomic status, a control variable, was a significant predictor of depression (β = -.44, p < .01), anxiety (β = -.26, p < .01), somatic symptoms (β = -.40, p < .01), and seizure frequency (β= -.37, p < .01), but not of seizure severity (β = -.15, p = .13). Being younger (β = -.23, p = 0.03) and male (β=-.22, p = .04) was also associated with greater seizure severity (Table 4 & 5).

Table 1
Demographic and Condition Specific Variable

	Epilepsy (*N* = 70)	Functional (*N* = 68)	95% Significance (two-tailed)
Demographic variables			
Age	M = 38.1	M = 39.3	*p* = .49[a]
	(SD = 11.6)	(SD = 12.9)	
Gender	77% Female	88% Female 1 non-binary	*p* = .52[b]
Ethnicity			*p* = .93[b]
White	64	60	
Asian	1	2	
Black	1	1	
Mixed	4	3	
Latina	0	1	
Native American	0	1	
Country			*p* < .001[b]
British	92.9%	54.4%	
International	7.1%	45.6%	
Perceived Socioeconomic Status	M = 5.23	M = 3.94	*p* < .001[c]
	(SD = 1.9)	(SD = 2)	
Employment			*p* < .001[b]
Paid work/full-time study	71.4%	30.8%	
Sickness/Disability leave	12.9%	57.3%	
Other	15.7%	11.9%	
Education			*p* = .43[b]
Secondary/vocational	21.4%	27.9%	
Post-secondary certificate	22.9%	13.2%	
University degree	50%	54.4%	
Other	5.7%	4.5%	
Condition variables			
Seizure severity	N= 36 M = 61.94 (SD = 18.03)	N=59 M = 50.89 (SD = 16.21)	*p* = .03[c]
Seizure frequency	M = 2.47 (SD = 1.22) Median = 2	M = 3.75 (SD = 1.75) Median = 4	*p* < .001[c]

Note: *[a]Mann-Whitney test; [b] Chi Square test; [c] T-test*

Table 2

Group Differences Between Dependent Variables

	Epilepsy (N = 70)	Functional (N = 68)	Test values	95% Significance level (one-tailed)
Depression	*M* = 12.34 *SD* = 6.77	*M*=16.51 *SD* = 6.77	*t* (136) = -3.61	*p* < .001 *d* = -.62
Anxiety	*M* = 9.21 *SD* = 5.86	*M* = 10.54 *SD* = 6.24	*t* (136) = -1.29	*p* = 0.1 *d* = -.22
Somatic symptoms	*M* = 13.41 *SD* = 6.71	*M* = 19.29 *SD* = 6.29	*t* (136) = -5.31	*p* < .001 *d* = -.55
Shame proneness	*M* = 3.50 *SD* = 0.85	*M* = 3.58 *SD* = 0.70	*t* (136) = .35	*p* =.36 *d* = .06
Shame aversion	*M* = 4.73 *SD* = 0.95	*M* = 4.67 *SD* =1.08	*t* (136) = -.14	*p* = .45 *d* = -.02

Table 3

Correlations Among Study Variables

Variable	1	2	3	4	5	6	7	8	9	10
1. Age	-									
2. Perceived SES*	.013	-								
3. Gender	-.259**	-.170*								
4. Depression	-.093	-.445**	.125	-						
5. Anxiety	-.142	-.224**	.082	.731**	-					
6. Somatic symptoms	-.012	-.391**	.074	.607**	.493**	-				
7. Shame aversion	-.250**	-.369**	.258**	.491**	.527**	.260**	-			
8. Shame proneness	-.206*	-.417**	.382**	.389**	.396**	.280**	.628**	-		
9. Seizure frequency	.051	-.348**	-.017	.240**	.086	.296**	-.043	.028	-	
10. Seizure severity	-.174	-.129	-.134	.131	.136	.070	.238*	.109	.058	-

Note. SES = Socioeconomic status; For gender (male code =1; female code =2) Spearman's rho was calculated. Pearson's correlation was calculated for all variables.
*p< .05 **p<. 01

Table 4

Regression Analyses of Group membership as Moderator of the Relation Between Shame Aversion and Shame Proneness and Seizure Severity and Seizure Frequency

Predictor	ΔR^2	ΔF	*Beta [95%CI]*
3. Depression as dependent variable			
Step 1	.20	F (3, 132)=11.5**	
Age			-.08 [-.14, .05]
Gender			.02 [-.37,.50]
Perceived SES			-.44 [-.59, -.28]**
Step 2	.37	F (6, 129)=12.73**	
SP			.07 [-.13, .26]
SA			.39 [.21, .58]**
Group			.24 [.19, .79]**
Step 3	.38	F (8, 127)=9.69**	
SP * group			-.12 [-.57, .20]
SA * group			.00 [-.37, .37]
4. Anxiety as dependent variable			
Step 1	.07	F (3, 132)=3.33*	
Age			-.14[.31, -.03]
Gender			-.01 [-.49, .44]
Perceived SES			-.26 [-.39, -.06]**
Step 2	.31	F (6, 129)=9.69**	
SP			.15 [-.06, .35]
SA			.47 [.28, .67]**
Group			.14 [-.03, .60]
Step 3	.31	F (8, 127)=7.20**	
SP * group			-.07 [-.48, .30]
SA * group			.02 [-.38, .44]
5. Somatic symptoms as dependent variable			
Step 1	.16	F (3, 132)= 8.240**	
Age			-.01 [-.17,.16]
Gender			.00[-.43,.45]
PSS			-.40[-.55,-.23]**
Step 2	.29	F (6, 129)= 8.665**	
SP			.13[-.08, .33]
SA			.15 [-.04, .34]
Group			.36[.39,1.02]**
Step 3	.30	F (8, 127)= 6.691**	
SP * group			.11 [-.23, .60]
SA * group			-.19 [-.65, .14]

Note. Note. PSS = Perceived Socioeconomic Status; SA = Shame aversion; SP=Shame proneness. Gender is dummy coded variable, with male d=0 and female d=1.

*p< .05 **p<. 01

Table 5
Regression Analyses of Group Membership as Moderator of the Relation Between Shame Aversion and Shame Proneness and Seizure Severity and Seizure Frequency

Predictor	ΔR^2	ΔF	*Beta (95% CI)*
	1. Seizure Severity		
Step 1	.10	F (3, 89)= 2.97*	
Age			-.23 [-.44, -.02]*
Gender			-.22 [-1.13,-.03]*
Perceived SES			-.15 [-.37,.05]
Step 2	.20	F (6, 86)= 3.68**	
SP			-.04 [-.32,.24]
SA			.15 [-.11,.41]
Group			-.32 [-1.07,-.23]**
Step 3	.22	F (8, 84)= 2.97**	
SP * group			.22 [-.84,.21]
SA * group			.26 [-.19, .84]
	2. Seizure frequency		
Step 1	.11	F (3, 132)= 6.03**	
Age			.04 [-.13, .20]
Gender			-.08[-.67,.23]
PSS			-.37[-.53, -21]**
Step 2	.28	F (6, 129)= 9.537**	
SP			.00 [-.20, .21]
SA			-.11 [-.30,.09]
Group			.41[.51; 1.14]**
Step 3	.27	F (8, 127)= 7.202**	
SP * group			.02[-.37,.45]
SA * group			.09[-.27, .51]

Note. Note. PSS = Perceived Socioeconomic Status; SA = Shame aversion; SP=Shame proneness. Gender is dummy coded variable, with male d=0 and female d=1.
*p< .05 **p<. 01

5. Discussion

The current study 1) examined levels and differences between the PWFS and PWE in terms of shame and mental health difficulties (MHDs); and 2) hypothesised that shame proneness and aversion would predict MHDs and seizures in both groups, and that this association would be stronger in the functional seizure group.

In line with our expectations, we found that both groups of seizure patients experienced high levels of shame, as the mean levels of these measures were more than a standard deviation higher in our PWFS and PWE groups, than in non-clinical populations of previous studies [32, 34]. However, in contrast to our predictions, no differences were found between the two groups in terms of levels of shame proneness or shame aversion, and anxiety. Still, in support with our first hypothesis, PWFS experienced higher levels of depression and somatic symptoms, than people with PWE.

In accordance with our second hypothesis, shame aversion predicted levels of depression and anxiety after controlling for perceived socioeconomic status (PSS), age, and gender in both groups. In contrast, shame proneness was not a significant predictor of any MHDs or seizure variables after controlling for PSS, age and gender. Furthermore, the strength of the associations between shame and MHDs did not differ between the PWFS and PWE groups.

However, an important ancillary finding emerged from our data: PSS was a significant predictor of depression, anxiety, somatic symptoms, seizure frequency; and was negatively associated with shame aversion, and shame proneness. Furthermore, PSS was significantly lower in the PWFS group, than in the PWE group.

5.1. Levels of shame and psychopathology in people with FS and ES

Shame proneness and aversion levels were equivalently high in both patient groups. This finding may reflect true similarities between groups regarding shame processes. Previous studies have shown that both patient groups experience high levels of stigma [3] and elevated levels of psychopathology compared to non-clinical populations [47]. These factors have previously been linked to elevated shame and could impact people with either seizure disorder [5, 48].

Indeed, in our study both patient groups reported clinical levels of depression and somatic symptoms, with symptom levels significantly higher in PWFS than in PWE. Anxiety levels were also higher in both groups, than would be expected in the general population. The mean anxiety symptom level in the PWFS met diagnostic threshold for clinical anxiety. Whilst anxiety did not reach diagnostic threshold for PWE, there was no significant difference observed in anxiety levels between groups. Our findings on levels of the heightened levels of anxiety, depression and somatic symptoms align with findings of previous studies with people with seizures [14, 47]. Hence, the results of the current study indicate the need of strategies to prevent and reduce social stigma and stress related to seizure disorders.

5.2. Shame as a predictor of MHDs

In this study shame aversion predicted anxiety and depression in both FS and ES, even after controlling for PSES, age, and gender. This finding contributes to previous evidence suggesting that shame aversion might be an important underlying process contributing to MHDs even more strongly than shame proneness [33-35]. Whilst shame is a painful emotion, perhaps it is not the experience of it, but how we respond to it matters the most in terms of its contribution to psychopathology. According to mindfulness-based theories, suffering relates less to emotions themselves, than to our unhelpful reactions to them, such as aversion to negative emotions and craving for positive ones [49, 50]. This finding therefore supports the rationale for emotional regulation approaches adopted by mindfulness-based therapies (such as Acceptance and Commitment Therapy, Compassion Focused Therapy, and Mindfulness-Based Stress Reduction [51]), that focus on promoting one's acceptance of inner states, instead of responding to them with aversion, in order to achieve mental well-being.

It is important to highlight that this study examined the role of shame in clinical populations with high levels of somatic symptoms, whilst controlling for significant demographic variables, and found that neither shame aversion nor proneness were associated with somatic symptoms in either group. Previous studies have found a weak relationship between shame and somatization, but they involved non-clinical samples and did not control for demographic variables [31, 52]. This could suggest that our study findings may be more representative of the experiences of people with clinically high levels of somatic symptoms, rather than those with lower levels.

5.3. Perceived socioeconomic status, an influential variable

Interestingly, PSS predicted depression, anxiety, somatic symptoms, and seizure frequency. This finding is not unique to this sample, as several previous studies found that perceived and actual social deprivation [53-55] and social inequalities [56] are associated with various physical and mental health difficulties. More specifically, a data linkage study found evidence that increased deprivation was linked to increased rates of epilepsy [57]. Although our study only assessed individuals' self-perceived socioeconomic status, the FS group's notably low employment rates provide additional support for the accuracy of the self-assessment of socioeconomic status. The results of this study highlight how social and demographic factors can impact psychological factors, which should be an important consideration for future studies, when studying clinical populations [58].

5.4. Limitations and future recommendations

One limitation of the study is its cross-sectional design, which means that we cannot establish true causal relationships between the variables. Our findings provide a rationale for longitudinal research capable of further exploring the impact of shame on the life outcomes of people with seizures. The second limitation of the study is the lack of a non-clinical control group, which limits our ability to draw firmer conclusions in terms of the levels of shame in our sample. Furthermore, this study included a volunteer sample with self-declared diagnosis, which means that participants may have not been fully representative and that neurological diagnoses may not always have been secure. Future research might use consecutive recruitment of participants with video-EEG confirmed diagnoses. However, it is uncertain whether this would make the sample more representative, as patients with infrequent seizures or less refractory seizure disorders might then be underrepresented. Furthermore, the study's generalisability is limited, as most of the study participants

identified as being white and living in high-income Western countries. It is uncertain whether our findings apply to people from various non-Western cultural backgrounds and from developing countries, where there are higher prevalence rates of epilepsy and poverty [59] . Our study had a good acceptability with 83% of the participants finishing the study, but we acknowledge the limitation of not knowing the reason for attrition of the remaining participants.

The study's strengths included a large clinical sample, which yielded sufficient power to detect an effect. Furthermore, the data analysis involved hierarchical modelling, which allowed the assessment of the unique contribution of each study variable, after controlling for social and demographic variables. To the authors' knowledge, this was the first study to assess the role of shame in people with epileptic and functional seizures, which could provide preliminary evidence for future studies.

6. Conclusion

The study results call for the clinical recognition that PWFS and PWE experience high levels of shame and mental health difficulties (MHDs), with shame aversion contributing to MHDs in both groups. These observations demonstrate that care providers should offer access to specialised psychological assessment and treatment for PWFS and PWE [60]. More specifically, our observations suggest that therapies that focus on promoting helpful shame regulation through self-compassion [12] and mindfulness or acceptance strategies [61] could be trialled as potentially suitable interventions for people with seizures.

The study findings also highlight the importance of socioeconomic status for mental health and seizure outcomes in both of our patient groups, underlining the importance of the social dimension of a bio-psycho-social approach to the treatment of patients with seizures [62].

Acknowledgements

We would like to thank the charities (Epilepsy Action, Epilepsy Research UK, FND Hope, FND Northern Ireland), consultant neurologists (Dr Richard Grunevald, Dr Varduhi Cahill, Dr Jon Dickson), Torie Robinson (from Epilepsy Sparks), and Dr Rhys Thomas, who helped us to recruit participants. We are also grateful for Epilepsy Action and FND Hope charities for offering feedback on our study protocol and questionnaires.

Declaration of Interest

None.

Funding

None.

Data Statement

The data is not made available due to confidentiality.

References

1. Jacoby, M., *Shame and the origins of self-esteem: A Jungian approach*. 2016: Taylor & Francis.
2. Gilbert, P., S. Allan, and K. Goss, *Parental representations, shame, interpersonal problems, and vulnerability to psychopathology.* Clinical Psychology & Psychotherapy: An International Journal of Theory and Practice, 1996. **3**(1): p. 23-34.
3. Mayor, R., et al., *Experiences of stigma in people with epilepsy: A meta-synthesis of qualitative evidence.* Seizure, 2022. **94**: p. 142-160.
4. Saraiya, T. and T. Lopez-Castro, *Ashamed and afraid: A scoping review of the role of shame in post-traumatic stress disorder (PTSD).* Journal of clinical medicine, 2016. **5**(11): p. 94.
5. Reuber, M., et al., *Shame in patients with psychogenic nonepileptic seizure: A narrative review.* Seizure, 2022. **94**: p. 165-175.
6. Budiarto, Y. and A.F. Helmi, *Shame and Self-Esteem: A Meta-Analysis.* Europe's Journal of Psychology, 2021. **17**(2): p. 131-145.
7. Manjrekar, E., M. Schoenleber, and W. Mu, *Shame aversion and maladaptive eating-related attitudes and behaviors.* Eating Behaviors, 2013. **14**(4): p. 456-459.
8. Schoenleber, M. and H. Berenbaum, *Shame aversion and shame-proneness in Cluster C personality disorders.* Journal of Abnormal Psychology, 2010. **119**(1): p. 197.

9. Schoenleber, M., P.I. Chow, and H. Berenbaum, *Self-conscious emotions in worry and generalized anxiety disorder.* British Journal of Clinical Psychology, 2014. **53**(3): p. 299-314.
10. Dickerson, S.S., T.L. Gruenewald, and M.E. Kemeny, *When the social self is threatened: Shame, physiology, and health.* Journal of personality, 2004. **72**(6): p. 1191-1216.
11. Craig, C., S. Hiskey, and A. Spector, *Compassion focused therapy: a systematic review of its effectiveness and acceptability in clinical populations.* Expert Rev Neurother, 2020. **20**(4): p. 385-400.
12. Gilbert, P., *Shame in psychotherapy and the role of compassion focused therapy.* 2011.
13. Annandale, M., G. Vilyte, and C. Pretorius, *Stigma in functional seizures: A scoping review.* Seizure - European Journal of Epilepsy, 2022. **99**: p. 131-152.
14. Brown, R.J. and M. Reuber, *Psychological and psychiatric aspects of psychogenic non-epileptic seizures (PNES): A systematic review.* Clinical Psychology Review, 2016. **45**: p. 157-182.
15. Arnold, L.M. and M.D. Privitera, *Psychopathology and Trauma in Epileptic and Psychogenic Seizure Patients.* Psychosomatics, 1996. **37**(5): p. 438-443.
16. Myers, L., et al., *Shame in the treatment of patients with psychogenic nonepileptic seizures: The elephant in the room.* Seizure, 2022. **94**: p. 176-182.
17. Association, A.P., *DSM-5 Classification*. 2016: American Psychiatric Association.
18. Brown, R.J. and M. Reuber, *Towards an integrative theory of psychogenic non-epileptic seizures (PNES).* Clin Psychol Rev, 2016. **47**: p. 55-70.
19. Holman, N., et al., *Adult attachment style and childhood interpersonal trauma in non-epileptic attack disorder.* Epilepsy research, 2008. **79**(1): p. 84-89.
20. Rawlings, G.H., et al., *Written accounts of living with psychogenic nonepileptic seizures: A thematic analysis.* Seizure, 2017. **50**: p. 83-91.
21. Lu, E., et al., *Systematic literature review of psychiatric comorbidities in adults with epilepsy.* Journal of clinical neurology (Seoul, Korea), 2021. **17**(2): p. 176.
22. Espinosa-Garcia, C., H. Zeleke, and A. Rojas, *Impact of Stress on epilepsy: Focus on neuroinflammation—A mini review.* International journal of molecular sciences, 2021. **22**(8): p. 4061.
23. Novakova, B., et al., *The role of stress as a trigger for epileptic seizures: a narrative review of evidence from human and animal studies.* Epilepsia, 2013. **54**(11): p. 1866-1876.
24. Mayor, R., et al., *An exploration of the experiences of self-disgust in people with epilepsy.* Epilepsy & Behavior, 2022. **126**: p. 108435.
25. Gerhardt, C., et al., *Insecure and unresolved/disorganized attachment in patients with psychogenic nonepileptic seizures.* Journal of the Academy of Consultation-Liaison Psychiatry, 2021. **62**(3): p. 337-344.
26. Tangney, J.P. and R.L. Dearing, *Shame and guilt*. 2003: Guilford press.
27. Tangney, J.P., P. Wagner, and R. Gramzow, *Proneness to shame, proneness to guilt, and psychopathology.* Journal of abnormal psychology, 1992. **101**(3): p. 469.
28. Cândea, D.M. and A. Szentagotai-Tătar, *Shame-proneness, guilt-proneness and anxiety symptoms: A meta-analysis.* J Anxiety Disord, 2018. **58**: p. 78-106.
29. Porter, A.C., et al., *Self-evaluation and depressive symptoms: A latent variable analysis of self-esteem, shame-proneness, and self-criticism.* Journal of Psychopathology and Behavioral Assessment, 2019. **41**: p. 257-270.

30. Rüsch, N., et al., *Shame and implicit self-concept in women with borderline personality disorder.* American journal of psychiatry, 2007. **164**(3): p. 500-508.
31. Fritch, H.M., *Examining the Role of Shame in Somatic Pain and Symptoms*. 2018: Liberty University.
32. Schoenleber, M. and H. Berenbaum, *Shame aversion and shame-proneness in Cluster C personality disorders.* Journal of Abnormal Psychology, 2010. **119**(1): p. 197-205.
33. Schoenleber, M. and H. Berenbaum, *Shame regulation in personality pathology.* Journal of Abnormal Psychology, 2012. **121**(2): p. 433.
34. Schoenleber, M., P.I. Chow, and H. Berenbaum, *Self-conscious emotions in worry and generalized anxiety disorder.* Br J Clin Psychol, 2014. **53**(3): p. 299-314.
35. Schoenleber, M., et al., *Association between shame aversion and ruminative retribution: Evidence for moderation by externalization of blame and control.* Motivation and emotion, 2021. **45**: p. 798-808.
36. Bosma, H., et al., *Low socioeconomic status and perceptions of social inadequacy and shame: findings from the Dutch SMILE study.* Eur J Public Health, 2015. **25**(2): p. 311-3.
37. Orth, U., R.W. Robins, and C.J. Soto, *Tracking the trajectory of shame, guilt, and pride across the life span.* Journal of personality and social psychology, 2010. **99**(6): p. 1061.
38. Faul, F., Erdfelder, E., Lang, A.-G., & Buchner, A. , *G*Power 3: A flexible statistical power analysis program for the social, behavioral, and biomedical sciences.* Behavior Research Methods, 2007. **39** p. 175–191.
39. Qualtrics, *Qualtrics* 2020: Provo, UT, USA.
40. Adler, N.E., et al., *Relationship of subjective and objective social status with psychological and physiological functioning: preliminary data in healthy white women.* Health Psychol, 2000. **19**(6): p. 586-92.
41. Alton, K.L., *Exploring the Guilt-Proneness of Non-Traditional Students*, in *Educational Psychology*. 2012, Southern Illionis University.
42. Scott-Lennox, J., et al., *Reliability, validity and responsiveness of a revised scoring system for the Liverpool Seizure Severity Scale.* Epilepsy research, 2001. **44**(1): p. 53-63.
43. Kroenke, K., et al., *The PHQ-8 as a measure of current depression in the general population.* Journal of affective disorders, 2009. **114**(1-3): p. 163-173.
44. Spitzer, R.L., et al., *A brief measure for assessing generalized anxiety disorder: the GAD-7.* Archives of internal medicine, 2006. **166**(10): p. 1092-1097.
45. Gierk, B., et al., *The somatic symptom scale–8 (SSS-8): a brief measure of somatic symptom burden.* JAMA internal medicine, 2014. **174**(3): p. 399-407.
46. Aiken, L.S., S.G. West, and R.R. Reno, *Multiple Regression: Testing and Interpreting Interactions*. 1991: SAGE Publications.
47. Diprose, W., F. Sundram, and D.B. Menkes, *Psychiatric comorbidity in psychogenic nonepileptic seizures compared with epilepsy.* Epilepsy & Behavior, 2016. **56**: p. 123-130.
48. Leaffer, E.B., D.C. Hesdorffer, and C. Begley, *Psychosocial and sociodemographic associates of felt stigma in epilepsy.* Epilepsy & Behavior, 2014. **37**: p. 104-109.
49. Teasdale, J.D. and M. Chaskalson, *How does mindfulness transform suffering? I: the nature and origins of dukkha*, in *Mindfulness*. 2013, Routledge. p. 89-102.
50. Teasdale, J.D. and M. Chaskalson, *How does mindfulness transform suffering? II: the transformation of dukkha*, in *Mindfulness*. 2013, Routledge. p. 103-124.

51. Baer, R.A. and J. Krietemeyer, *Overview of mindfulness-and acceptance-based treatment approaches.* Mindfulness-based treatment approaches: Clinician's guide to evidence base and applications, 2006: p. 3-27.
52. Pineles, S.L., A.E. Street, and K.C. Koenen, *The differential relationships of shame–proneness and guilt–proneness to psychological and somatization symptoms.* Journal of Social and Clinical Psychology, 2006. **25**(6): p. 688-704.
53. Kivimäki, M., et al., *Association between socioeconomic status and the development of mental and physical health conditions in adulthood: a multi-cohort study.* The Lancet Public Health, 2020. **5**(3): p. e140-e149.
54. Mishra, S. and R.N. Carleton, *Subjective relative deprivation is associated with poorer physical and mental health.* Social Science & Medicine, 2015. **147**: p. 144-149.
55. Visser, K., et al., *Neighbourhood deprivation effects on young people's mental health and well-being: A systematic review of the literature.* Social Science & Medicine, 2021. **270**: p. 113542.
56. Pickett, K.E. and R.G. Wilkinson, *Income inequality and health: a causal review.* Social science & medicine, 2015. **128**: p. 316-326.
57. Pickrell, W.O., et al., *Epilepsy and deprivation, a data linkage study.* Epilepsia, 2015. **56**(4): p. 585-591.
58. Bronfenbrenner, U., *Ecological systems theory*, in *Six theories of child development: Revised formulations and current issues.* 1992, Jessica Kingsley Publishers: London, England. p. 187-249.
59. Neligan, A. and J. Sander, *The incidence and prevalence of epilepsy.* London: UCL Institute of Neurology, 2009.
60. Glen, E.T., et al., *Integrative care models in neuropsychology: A National Academy of Neuropsychology education paper.* Archives of Clinical Neuropsychology, 2019. **34**(2): p. 141-151.
61. Lundgren, T., J. Dahl, and S.C. Hayes, *Evaluation of mediators of change in the treatment of epilepsy with acceptance and commitment therapy.* Journal of behavioral medicine, 2008. **31**: p. 225-235.
62. Elliott, J.O. and V.E. Richardson, *The biopsychosocial model and quality of life in persons with active epilepsy.* Epilepsy & Behavior, 2014. **41**: p. 55-65.