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Experience sampling methodology study of anxiety and depression in adolescents with epilepsy: The role of metacognitive beliefs and perseverative thinking

James Temple [a,b], Mary Gemma Cherry [a,c,*], Victoria Gray [d], Andrew Jones [e], Peter Fisher [a,c]

[a] Department of Primary Care and Mental Health, University of Liverpool, Liverpool, UK
[b] Mersey Care NHS Foundation Trust, Liverpool, UK
[c] Clinical Health Psychology Service, Liverpool University NHS Foundation Trust, Liverpool, UK
[d] Psychological Services (Paediatrics), Alder Hey Children's NHS Foundation Trust, Liverpool, UK
[e] Department of Psychology, Liverpool John Moores University, Liverpool, L3 3AF, UK

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ABSTRACT

Emotional distress is common in young people with epilepsy (YPwE). According to the Self-Regulatory Executive Function (S-REF) model, maladaptive metacognitive beliefs and perseverative thinking are fundamental in the development and maintenance of emotional distress. As emotional distress and perseverative thinking can highly fluctuate over short intervals in YPwE, it is important to account for this variability when testing the utility of psychological models. Experience sampling methodology (ESM) was therefore used to explore the momentary relationship between metacognitive beliefs, perseverative thinking, and emotional distress in YPwE. Eighteen participants diagnosed with epilepsy (aged 12–17 years) completed the 10-day ESM period. Participants were prompted to complete the ESM assessment five times daily. The ESM assessment assessed participant's momentary levels of metacognitive beliefs, perseverative thinking (i.e., worry and rumination), and emotional distress (i.e., anxiety and depression). A series of multilevel regression analyses indicated that metacognitive beliefs were significantly positively associated with worry, rumination, anxiety and depression. After controlling for worry and rumination, respectively, metacognitive beliefs did not account for additional variance in anxiety or depression. Findings provide preliminary support for the utility of the S-REF model for emotional distress in YPwE. Metacognitive therapy, which is underpinned by the S-REF model, may be an appropriate intervention for emotional distress in YPwE. Future studies should assess the mediational relationship between metacognitive beliefs, perseverative thinking, and emotional distress using time-lagged models.

1. Introduction

Epilepsy is one of the most common neurological conditions in the world [1] affecting around 60,000 young people in the UK [2] and around 760,000 young people in North America [3]. Around 19 % and 14 % of young people with epilepsy (YPwE; aged ≤ 18 years) meet diagnostic criteria for anxiety and depressive disorders, respectively [4], 3–5 times higher than that reported in the general youth population [5]. Anxiety and depression in YPwE are associated with adverse antiseizure medication (ASM) effects [6] and have a larger negative impact on quality of life than seizure frequency or duration [7]. Anxiety and depression also increase suicide risk [8,9], the incidence being 2.3 times higher in YPwE than in the general youth population [10]. Providing effective psychological interventions for YPwE is therefore essential [4,11].

Cognitive behavioural therapy (CBT) is recommended for anxiety and depression in YPwE [12,13]. However, the existing evidence-base for the efficacy of CBT in YPwE is limited [14,15]. CBT achieves only modest treatment effects for young people and adults with physical health conditions [16,17], including adults with epilepsy [18]. This is perhaps because 'reality-testing' negative automatic thoughts (NATs), a defining feature of CBT, may be of limited benefit in a physical health context given that people's NATs are often realistic (e.g., 'I am unable to control my seizures') [17–20]. Moreover, a recent systematic review of psychosocial variables associated with emotional distress in YPwE found conflicting evidence regarding the association between emotional distress and illness appraisals [21], the modification of which is a central premise of CBT. Thus, psychological interventions that focus on how and why people respond negatively to NATs (as opposed to focusing on the

* Corresponding author.
E-mail address: gcherry@liv.ac.uk (M. Gemma Cherry).

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content of NATs) may be more useful for alleviating anxiety and depression in YPwE. One such intervention is Metacognitive Therapy (MCT) [22].

Preliminary evidence indicates that MCT may be an effective intervention for anxiety and depression in adults with physical health conditions [23–28], adolescents and young adults with cancer [29], and adolescents with common mental health disorders [30–34]. Prior to evaluating the efficacy of MCT in YPwE, the clinical utility of the psychological model underpinning MCT, the Self-Regulatory Executive Functioning (S-REF) model [35,36], needs to be established [37,38].

According to the S-REF model, anxiety and depression are maintained and intensified by a negative and continued response style called the cognitive attentional syndrome (CAS). The CAS includes perseverative thinking (i.e., worry and rumination), attentional focus on threat, and unhelpful coping behaviours (e.g., thought suppression, substance misuse, avoidance). The CAS is activated by stored metacognitive beliefs (i.e., beliefs about thinking, emotions, and conceptual processing strategies). While several metacognitive belief domains are highlighted in the S-REF model, they are often clustered into two general types: *positive metacognitive beliefs* and *negative metacognitive beliefs*. Positive metacognitive beliefs refer to beliefs about the benefits of, or need to, engage in perseverative thinking (e.g., "I must ruminate in order to find answers to my sadness", "worrying helps me cope"); whereas negative metacognitive beliefs refer to beliefs about the uncontrollability and danger of perseverative thinking (e.g., "my ruminating is uncontrollable", "worrying will make me go crazy").

Preliminary evidence supports the utility of the S-REF model for anxiety and depression in adults with epilepsy; positive and negative metacognitive beliefs are associated with anxiety and depression, with relationships partially mediated by perseverative thinking in the form of worry [39,40]. However, the relationship between metacognitive beliefs, perseverative thinking (i.e., worry and rumination), and anxiety and depression in YPwE has not been explored. Moreover, the two studies investigating the utility of the S-REF model in epilepsy have several methodological shortcomings. First, both studies relied solely on retrospective self-report measures, which are often affected by recall biases [41]. As people with epilepsy often experience memory problems [42], recall bias may be more likely in this population. Second, data were only collected at a single time-point. Emotional distress and engagement in perseverative thinking can highly fluctuate over short intervals [43]. Thus, collecting data at a single time-point increases risk of inaccuracy. Due to the unpredictability of seizures, the occurrence of which are associated with increased distress [44,45], perseverative thinking and emotional distress may be more likely to fluctuate in people with epilepsy than the general population. Moreover, retrospective and daily measures of psychological variables measure different constructs. For instance, one study found that retrospective measures of worry only accounted for a small amount of variance in daily worry [46]. Another study demonstrated that daily rumination predicted higher cortisol levels whereas retrospective measures of rumination did not [47]. Thus, an alternative methodology that overcomes the limitations of traditional retrospective self-report methods and accounts for the daily variability in perseverative thinking and emotional distress in YPwE is needed.

One such method is experience sampling methodology (ESM). ESM involves asking participants to complete a short assessment about their current 'momentary' experiences several times daily in everyday settings [48]. This minimises recall bias, enables the assessment of variability in experiences over time, and more accurately captures cause and effect relationships [41]. It also enables the assessment of emotional distress and related factors as they occur in their natural environment alongside daily tasks, increasing external validity [49].

ESM has been used to assess the relationship between metacognitive beliefs and perseverative thinking (i.e., worry and rumination) in adult populations [50–54]. However, to our knowledge, ESM has never been used to assess the relationship between metacognitive beliefs and perseverative thinking in YPwE. Therefore, the aims of this study were to assess the influence of metacognitive beliefs on perseverative thinking (i.e., worry and rumination) and emotional distress (i.e., anxiety and depression) in YPwE via ESM. More specifically, using ESM we explored whether metacognitive beliefs were associated with worry and rumination (aim 1), and anxiety and depression (aim 2); whether worry and rumination were respectively associated with anxiety and depression (aim 3); and whether metacognitive beliefs explained additional variance in anxiety and depression after respectively accounting for worry and rumination (aim 4). Given adolescence is often regarded as a particularly challenging time for young people involving numerous physical and psychological changes [55,56], the study will focus specifically on YPwE aged 12–17 years.

2. Method

2.1. Study design

A prospective cohort design including a baseline assessment and an ESM assessment period was used. There is little theoretical guidance on the appropriate frequency and duration of an ESM assessment period. The decision largely depends on the amount of data required to capture sufficient variability in the constructs being investigated. Findings regarding the impact of ESM assessment frequency on compliance rates are mixed. While some meta-analyses [57,58] have found compliance rates are associated with ESM assessment frequency, others [59,60] have found no association between ESM assessment frequency and compliance rates. A recent ESM study [61] assessing similar constructs to those assessed in our study (i.e., perseverative thinking and emotional distress) explored the optimal ESM assessment frequency and duration in a non-clinical sample of adults. Participants were prompted to complete eight assessments daily for 14 consecutive days. Based on inspection of data and participant feedback, the authors concluded that five assessments daily for ten days yielded the best 'trade-off' between participant burden and data quality and quantity. We therefore chose for ESM assessments to occur five times daily for ten consecutive days.

2.2. Participants

Participants were recruited through a National Health Service (NHS) children's hospital in North-West England and through advertisements on social media. To be eligible, participants needed to have a diagnosis of epilepsy (of any type), be aged 12–17 years old, and have access to a smartphone with an android or iOS operating system for the 10-day ESM period. Both participant and their parent/caregiver needed to sufficiently understand English and have capacity to provide informed assent and consent, respectively. We aimed to recruit as many participants as possible within our recruitment period.

2.3. ESM assessment protocol

ESM assessments were delivered by an application ('app') downloaded onto participants' smartphones (SEMA3) [62]. Prompts were delivered in pseudo-randomised blocks during participants average waking hours with each block lasting a minimum of 120 minutes. If assessments were not completed within 15 minutes of the prompt, they were no longer accessible.

2.4. Measures

2.4.1. Baseline assessment

Sociodemographic and epilepsy information were obtained via medical records (for those recruited through an NHS children's hospital) and self-report.

Metacognitive beliefs were assessed using the Metacognitions Questionnaire for Adolescents (MCQ-A) [63]; a 30-item self-report questionnaire measuring five metacognitive belief domains across five

subscales. The MCQ-A has been validated for use with adolescents and has acceptable internal consistency across the five subscales (α = 0.66-0.88) [63,64].

Anxiety and depression were assessed using the 25-item version of the Revised Child Anxiety and Depression Scale (RCADS-25) [65]. The RCADS-25 measures child and adolescent symptoms of anxiety (15-items) and depression (10-items). Both subscales have acceptable internal consistency (α = 0.79-0.82), test–retest reliability (intraclass correlation coefficient [ICC] = 0.70-0.73), and construct validity [65,66].

Worry and rumination were assessed using 5-item versions of the Penn State Worry Questionnaire (brief-PSWQ) [67] and the Ruminative Response Scale (brief-RRS) [67]. Both scales have been validated in adolescents, correlate highly with their respective full versions (brief-PSWQ: r = 0.91–0.94; brief-RRS: r = 0.88–0.91), and have acceptable to excellent internal consistency (brief-PSWQ: α = 0.84–0.91; brief-RRS: α = 0.78–0.81) [67].

2.4.2. ESM assessment

Where possible, we used validated and reliable ESM assessment measures (momentary anxiety, depression, worry, rumination). Otherwise, we developed our own measures based on validated and reliable retrospective self-report questionnaires (momentary positive and negative metacognitive beliefs about worry and rumination). Item selection was based on face validity. Members of a young person's advisory group were consulted about the ESM assessment measures and schedule. Split-half reliability coefficients were calculated for all ESM assessment measures (mean scores for each measure for the first half of the 10-day ESM period were compared with mean scores for the second half). Using the 'mlr' (multilevel reliability) function from the 'psych' package in R, reliability of within-person change and between-person change were also calculated for the main ESM outcome measures (i.e., anxiety and depression) [68]. ESM assessment measures assessed momentary levels of anxiety (3 items), depression (3 items), worry (1 item), rumination (1 item), positive metacognitive beliefs about worry and rumination (2 items), and negative metacognitive beliefs about worry and rumination (2 items). All ESM items were preceded by the phrase 'Right now' (see Appendix A for all included ESM items).

Anxiety and depression were assessed using an adapted version of the anxiety and depression subscales of the 15-item Profile of Mood States (POMS-15) scale [69]. Each subscale consists of 3 items assessing momentary anxiety and depression, respectively. Both subscales have been used in adolescents [70,71]. In the current sample, both subscales demonstrated excellent reliability (ICC = 0.97 for both subscales). Both subscales also demonstrated excellent within-person reliability (R_C = 0.87 for anxiety; R_C = 0.86 for depression) and between-person reliability (R_{KF} = > 0.99 for both subscales).

Worry and rumination were assessed using an adapted version of a 2-item scale developed by Kircanski et al. (2015) [72]. The 2-item scale measures momentary worry (1-item) and rumination (1-item). Both items demonstrate good convergent and discriminant validity with retrospective self-report questionnaires assessing worry and rumination, respectively [72]. In the current sample, both items demonstrated excellent reliability (ICC = 0.93 for both items).

Positive and negative metacognitive beliefs were assessed by adapting items from the CAS-1 [22] and the Metacognitions about Symptom Control Scale (MaSCS) [73] to represent momentary experiences. Two subscales were developed: *positive metacognitive beliefs about worry and rumination*; *negative metacognitive beliefs about worry and rumination*. The CAS-1 has good concurrent and predictive validity, and good internal consistency (α = 0.78) in an epilepsy population [74,75]. The MaSCS has good concurrent validity and internal consistency (α = 0.88-0.89 for subscales) in a physical health population [73]. In the current sample, the positive metacognitive beliefs about worry and rumination subscale demonstrated good reliability (ICC = 0.87) and the negative metacognitive beliefs about worry and rumination subscale demonstrated excellent reliability (ICC = 0.92).

2.5. Procedure

Ethical approval was obtained from the NHS Wales Research Ethics Committee 4 (reference: 21/WA/0072). Participants completed the baseline assessment measures online via Qualtrics. Next, participants and their parent/caregiver met with the researcher (JT) remotely via video-platform to set up the ESM app on their smartphone and choose a start date for the ESM assessment period. Participants who completed the study received a £15 gift voucher.

2.6. Statistical analysis

The analysis plan was preregistered in the AsPredicted database (https://aspredicted.org/8v7ga.pdf). No baseline data were missing. Participants completing less than one third of assessments were excluded from analyses [43,76,77]. Pearson correlation, *t*-tests, or ANOVAs assessed whether compliance rates (i.e., number of ESM assessments completed) significantly differed depending on emotional distress, sociodemographic information, or clinical characteristics. Spearman's correlation analysis using ESM average scores assessed the association between ESM measures and their corresponding baseline measure (clustering was not accounted for in these analyses). These analyses were conducted using SPSS version 28.0.1.1.

Multilevel modelling explored the relationship between momentary metacognitive beliefs, perseverative thinking (i.e., worry and rumination) and emotional distress (i.e., anxiety and depression). The data obtained via ESM was nested at three levels: assessments (level 1), days (level 2), individuals (level 3). As there was informative clustering within days (level 2) [78], a 2-level structure (assessments > individuals) was most appropriate.

To explore aim 1, multilevel regression models were constructed with worry and rumination as respective outcome variables. Initially, multilevel simple regression models assessed associations between each outcome variable (worry and rumination) and each independent variable (positive metacognitive beliefs about worry and rumination, negative metacognitive beliefs about worry and rumination). To assess the relative contribution of each independent variable to worry and rumination, multilevel multiple regression models were constructed with the independent variables significantly associated with worry and rumination in the simple regression models entered simultaneously.

To explore aim 2 and 3, multilevel regression models were constructed with anxiety and depression as respective outcome variables. Multilevel simple regression models explored associations between each outcome variable (anxiety and depression) and each independent variable (positive metacognitive beliefs about worry and rumination, negative metacognitive beliefs about worry and rumination, worry [for the outcome variable anxiety], rumination [for the outcome variable depression]). To explore aim 4, multilevel multiple regression models were constructed for each outcome variable (anxiety and depression) with the independent variables significantly associated with anxiety and depression in the simple regression models entered simultaneously.

A random intercept for each individual was used for each model. Unstandardized beta coefficients were calculated for each model. ICCs were calculated to explain the proportion of variability explained by clustering (i.e., within participants). Marginal and conditional R^2 values were calculated for the multilevel regression models to explain how much variance is accounted for by the independent variables without the variance explained by clustering (marginal R^2) and how much variance is explained by the independent variables and variation explained by clustering combined (conditional R^2). Multilevel modelling was conducted in R using the lme4 package.

Sensitivity analyses were conducted in which all multilevel multiple regression models were repeated with the inclusion of the corresponding baseline measure of the outcome variable investigated as an additional

independent variable (e.g., for the model in which momentary worry was the outcome variable, baseline worry was included as an additional independent variable).

3. Results

3.1. Descriptive statistics

Nineteen participants took part in the study. One participant completed less than third of the ESM assessments and was excluded from analyses. Thus, 18 participants were included in the analyses. Sample characteristics are summarized in Table 1. Participants had a mean age of 14.3 (range 12–17), were predominantly female (65 %), and all but one were White British. The most common seizure type was generalized (50 %), followed by focal (39 %). Most of the sample were on ASM monotherapy (61 %). The mean age of epilepsy diagnosis was 8.9 years and mean epilepsy duration was 5.4 years. The mean time elapsed between completing the baseline assessment and starting the 10-day ESM period was 13.3 days (median 9 days). Thirty-three percent and 22 % of the sample scored above the clinical threshold for anxiety and depression, respectively. Mean and median scores and standard deviations for all baseline and ESM assessment measures are shown in Table 2. Scores were relatively low across most measures.

Table 1
Sample characteristics (n = 18).

Variable	Category	n (% of participants)
Gender	Male	4 (22.2 %)
	Female	13 (72.2 %)
	Prefer not to say	1 (5.6 %)
Age: mean (range; SD)		14.33 (12–17; 1.24)
Ethnicity	White British	17 (94.4 %)
	White & Black African	1 (5.6 %)
Seizure type	Generalized	9 (50 %)
	Focal	7 (38.9 %)
	Unknown	2 (11.1 %)
ASM protocol	Monotherapy	11 (61.1 %)
	Polytherapy	7 (38.9 %)
Seizure severity	Mild	5 (27.8 %)
	Moderate	8 (44.4 %)
	Severe	0 (0 %)
	Unknown	5 (27.8 %)
Age of epilepsy diagnosis (n = 16): mean (range; SD)		8.88 (2–16; 4.49)
Epilepsy duration in years (n = 16): mean (range; SD)		5.38 (0–12; 4.36)
Comorbidity		
None		14 (77.8 %)
Another medical condition		4 (22.2 %)
Days between baseline assessment and starting ESM protocol: mean (range; SD)		13.33 (3–46; 11.54)
RCADS-25 Anxiety clinical threshold[a]		N/A: 1 (5/6%) [b]Below threshold: 10 (55.6 %) Borderline threshold: 1 (5.6 %) Above threshold: 6 (33.3 %)
RCADS-25 Depression clinical threshold[a]		N/A: 1 (5.6 %) [b]Below threshold: 12 (66.7 %) Borderline threshold: 1 (5.6 %) Above threshold: 4 (22.2 %)

Note. ASM = Antiseizure medication; ESM = Experience sampling method; RCADS = Revised Children's Anxiety & Depression Scale (25-item version); N/A = Not applicable; *SD* = standard deviation; [a]*T*-scores of 65–70 are classed as borderline clinical threshold for anxiety or depression, *T*-scores of > 70 are classed as above clinical threshold for anxiety or depression. See Appendix B for further information on how T-scores were calculated; [b]As identifying as male or female is required to calculate *T*-scores for the RCADS, this could not be calculated for one participant.

3.2. Compliance with ESM protocol

Of a possible 900 ESM assessments, participants completed 599 (66 %) within the 15-minute timeframe. Participant compliance rates of ESM assessments ranged from 40 to 88 % (mean 67 %). Compliance rates were not significantly associated with emotional distress, sociodemographic information, or clinical characteristics (see Appendix C).

3.3. Correlation analysis

Significant moderate-to-large correlations were found between most baseline measures (e.g., baseline worry was significantly correlated with most other baseline measures) and most ESM measures (e.g., worry measured via ESM was significantly correlated with most other ESM measures). Weak and non-significant correlations were found between positive metacognitive beliefs about worry and rumination (measured via ESM) and most other ESM measures. Thus, the positive metacognitive beliefs about worry and rumination subscale was separated

Table 2
Descriptive statistics for each study variable.

Variable	Mean (median; *SD*)	Score range
Baseline measures		
1) Anxiety (RCADS-25 anxiety subscale)	15.56 (14.5; 7.06)	0–60
2) Depression (RCADS-25 depression subscale)	12.67 (11.5; 6.04)	0–40
3) Worry (brief-PSWQ)	17.56 (18.5; 5.26)	5–25
4) Rumination (brief-RRS)	12.05 (11.5; 3.35)	5–20
5) Positive metacognitive beliefs about worry (MCQ-A-PBW subscale)	11.22 (11; 3.49)	6–24
6) Negative metacognitive beliefs about worry (MCQ-A-NBW subscale)	16.44 (15.5; 4.73)	6–24
ESM assessment measures		
7) Anxiety	0.96 (0.79; 0.92)	0–4
8) Depression	0.98 (0.55; 1.13)	0–4
9) Worry	1.19 (1.25; 0.98)	0–4
10) Rumination	1.01 (1.03; 0.97)	0–4
11) Positive metacognitive beliefs about worry[a]	0.73 (0.56; 0.76)	0–4
12) Positive metacognitive beliefs about rumination[a]	0.65 (0.36; 0.67)	0–4
13) Negative metacognitive beliefs about worry and rumination	3.16 (3.15; 2.28)	0–8

Note. brief-PSWQ = Penn State Worry Questionnaire (5-item version); brief-RRS = Ruminative Response Scale (5-item version); ESM = Experience sampling method; MCQ-A-PBW = Metacognitions Questionnaire for Adolescents–Positive Beliefs about Worry Subscale; MCQ-A NBW = Metacognitions Questionnaire for Adolescents–Negative Beliefs about Danger and Uncontrollability of Worry Subscale; RCADS = Revised Children's Anxiety & Depression Scale (25-item version). *SD* = standard deviation; [a]see 'Correlation analysis' section for explanation of why positive metacognitive beiefs about worry and rumination are presented as two distinct subscales.

Table 3
Spearman's rho correlations between study variables.

	Baseline measures						*ESM assessment measures*					
Variables	**1)**	**2)**	**3)**	**4)**	**5)**	**6)**	**7)**	**8)**	**9)**	**10)**	**11)**	**12)**	**13)**
Baseline measures													
1) Anxiety (RCADS-25 anxiety subscale)	–	0.68**	0.78***	0.81***	0.44	0.69**	0.35	0.09	0.14	0.14	-0.40	-0.23	-0.03
2) Depression (RCADS-25 depression subscale)		–	0.48*	0.75***	0.31	0.54*	0.44	0.23	0.15	0.21	-0.11	0.08	0.07
3) Worry (brief-PSWQ)			–	0.78***	0.33	0.70**	0.22	-0.07	0.09	0.01	-0.28	-0.26	0.18
4) Rumination (brief-RRS)				–	0.18	0.72***	0.26	0.05	0.15	0.13	-0.12	-0.03	0.20
5) Positive metacognitive beliefs about worry (MCQ-A-PBW subscale)					–	0.25	0.67**	0.40	0.38	0.47	-0.08	-0.10	-0.06
6) Negative metacognitive beliefs about worry (MCQ-A-NBW subscale)						–	0.44	0.14	0.45	0.30	-0.24	-0.01	0.47*
ESM assessment measures													
7) Anxiety							–	0.83***	0.85***	0.88***	0.05	0.37	0.47
8) Depression								–	0.84***	0.86***	0.31	0.48*	0.40
9) Worry									–	0.89***	0.19	0.41	0.64**
10) Rumination										–	0.29	0.57*	0.57*
11) Positive metacognitive beliefs about worry											–	0.78***	0.06
12) Positive metacognitive beliefs about rumination												–	0.31
13) Negative metacognitive beliefs about worry and rumination													–

Note. ESM = Experience sampling method; RCADS = Revised Children's Anxiety & Depression Scale (25-item version); brief-PSWQ = Penn State Worry Questionnaire (5-item version); brief-RRS = Ruminative Response Scale (5-item version); MCQ-A-PBW = Metacognitions Questionnaire for Adolescents–Positive Beliefs about Worry Subscale; MCQ-A NBW = Metacognitions Questionnaire for Adolescents–Negative Beliefs about Danger and Uncontrollability of Worry Subscale; * $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$ (two-tailed).

into two distinct subscales: *positive metacognitive beliefs about worry* and *positive metacognitive beliefs about rumination.*

Regarding correlations between ESM measures and their corresponding baseline measure, a significant moderate correlation was found between negative metacognitive beliefs about worry and rumination measured via ESM and baseline negative metacognitive beliefs about worry ($\rho = 0.47$, $p < 0.05$). Non-significant correlations were found between all other corresponding baseline and ESM measures (see Table 3).

3.4. Multilevel modelling

3.4.1. Aim 1

In the multilevel simple regression analyses for worry and rumination, all three metacognitive belief domains were significantly associated with worry ($\beta = 0.20$-0.31) and rumination ($\beta = 0.17$-0.31). The marginal R^2 and condition R^2 for the independent worry and rumination models ranged from 0.02 to 0.20 and 0.55 to 0.59, respectively, indicating that the models accounted for 2–20 % of the variance when variability due to clustering within-person was not accounted and 55–59 % of the variance when it was accounted. The ICCs for the independent worry and rumination models ranged from 0.45 to 0.54, indicating that approximately half of the variability in observed relationships between outcome variables and each significant independent variable was due to within-person variation (see Table 4).

When entered simultaneously into a multilevel multiple regression analysis, positive metacognitive beliefs about rumination ($\beta = 0.16$ for worry model; $\beta = 0.17$ for rumination model) and negative metacognitive beliefs about worry and rumination ($\beta = 0.17$ for worry model; $\beta = 0.20$ for rumination model) remained significantly independently associated with worry and rumination whereas positive metacognitive beliefs about worry did not. The overall model for worry accounted for 17 % (marginal $R^2 = 0.17$) of the variance when variability due to clustering within-person was not accounted and 56 % (conditional $R^2 = 0.56$) of the variance when it was accounted. The overall model for rumination accounted for 21 % (marginal $R^2 = 0.21$) of the variance when variability due to clustering within-person was not accounted and 60 % (conditional $R^2 = 0.60$) of the variance when it was accounted. The ICCs for the multilevel multiple regression models for worry and rumination were 0.47 and 0.50, respectively (see Table 5).

3.4.2. Aim 2 & 3

The results of the multilevel simple regression analyses for anxiety and depression are shown in Table 6. Positive metacognitive beliefs about rumination ($\beta = 0.19$), negative metacognitive beliefs about worry and rumination ($\beta = 0.08$), and worry ($\beta = 0.46$) were significantly associated with anxiety. Positive metacognitive beliefs about rumination ($\beta = 0.13$), negative metacognitive beliefs about worry and rumination ($\beta = 0.07$), and rumination ($\beta = 0.32$) were significantly associated with depression. The marginal R^2 and condition R^2 for the

Table 4
Multilevel simple regression for the outcome variables (worry, rumination) and each independent variable.

Worry	***β***	**95 % CI**	***p***	**ICC**	**Marginal R^2/Conditional R^2**
Positive metacognitive beliefs about worry	0.22	0.12 - 0.32	< 0.001	0.54	0.03/0.55
Positive metacognitive beliefs about rumination	0.31	0.21 - 0.42	< 0.001	0.53	0.05/0.55
Negative metacognitive beliefs about worry and rumination	0.20	0.16 - 0.25	< 0.001	0.45	0.17.55
Rumination	***β***	**95 % CI**	***p***	**ICC**	**Marginal R^2/Conditional R^2**
Positive metacognitive beliefs about worry	0.17	0.07 - 0.27	< 0.01	0.54	0.02/0.55
Positive metacognitive beliefs about rumination	0.31	0.20 - 0.41	< 0.001	0.54	0.05/0.56
Negative metacognitive beliefs about worry and rumination	0.22	0.18 - 0.26	< 0.001	0.49	0.20/0.59

Note. CI = confidence interval; ICC = intraclass correlation coefficient; β = unstandardized beta coefficient

Table 5
Multilevel multiple regression for the outcome variables (worry, rumination) with independent variables entered simultaneously.

Worry	***β***	**95 % CI**	***P***
Intercept	0.50	0.08 - 0.92	< 0.05
Positive metacognitive beliefs about worry	0.07	-0.04 -0.18	ns
Positive metacognitive beliefs about rumination	0.16	0.04 - 0.28	< 0.01
Negative metacognitive beliefs about worry and rumination	0.17	0.12 - 0.21	< 0.001
Model summary			
Assessments: 589			
ICC = 0.47			
Marginal R^2 / Conditional R^2 = 0.17 / 0.56			
Rumination	***β***	**95 % CI**	***P***
Intercept	0.28	-0.13 -0.69	ns
Positive metacognitive beliefs about worry	0.01	-0.10 - 0.11	ns
Positive metacognitive beliefs about rumination	0.17	0.06 - 0.28	< 0.01
Negative metacognitive beliefs about worry and rumination	0.20	0.15 - 0.24	< 0.001
Model summary			
Assessments: 589			
ICC = 0.50			
Marginal R^2 / Conditional R^2 = 0.21 / 0.60			

Note. CI = confidence interval; ICC = intraclass correlation coefficient; ns = non-significant; β = unstandardized beta coefficient

Table 6
Multilevel simple regression for the outcome variables (anxiety, depression) and each independent variable.

Anxiety	***β***	**95 % CI**	***p***	**ICC**	**Marginal R^2/Conditional R^2**
Worry	0.46	0.41 - 0.51	< 0.001	0.44	0.37/0.65
Positive metacognitive beliefs about worry	0.05	-0.04 - 0.13	ns	–	0.00/0.62
Positive metacognitive beliefs about rumination	0.19	0.10 - 0.28	< 0.001	0.62	0.02/0.63
Negative metacognitive beliefs about worry and rumination	0.08	0.05 - 0.12	< 0.001	0.59	0.04/0.60
Depression	***β***	**95 % CI**	***p***	**ICC**	**Marginal R^2/Conditional R^2**
Rumination	0.32	0.26 - 0.38	< 0.001	0.66	0.13/0.70
Positive metacognitive beliefs about worry	-0.01	-0.09 - 0.07	ns	–	0.00/0.72
Positive metacognitive beliefs about rumination	0.13	0.05 - 0.22	< 0.01	0.72	0.01/0.62
Negative metacognitive beliefs about worry and rumination	0.07	0.03 - 0.10	< 0.001	0.71	0.02/0.72

Note. CI = confidence interval; ICC = intraclass correlation coefficient; ns = non-significant; β = unstandardized beta coefficient

variables significantly associated with anxiety and depression ranged from 0.02 to 0.37 and 0.60 to 0.72, respectively, indicating that the models accounted for 2–37 % of the variance when variability due to clustering within-person was not accounted and 60–72 % of the variance when it was accounted. Positive metacognitive beliefs about worry were not significantly associated with anxiety or depression. ICCs for the independent anxiety and depression models ranged from 0.44 to 0.72.

3.4.3. Aim 4

The results of the multilevel multiple regression analyses for anxiety and depression are shown in Table 7. After respectively controlling for worry and rumination (and other metacognitive beliefs domains respectively significantly associated with anxiety and depression in the multilevel simple regression analyses), none of the metacognitive belief domains were significantly independently associated with anxiety or depression. The overall model for anxiety accounted for 36 % (marginal R^2 = 0.36) of the variance when variability due to clustering within-person was not accounted and 65 % (conditional R^2 = 0.65) of the variance when it was accounted. The overall model for depression accounted for 13 % (marginal R^2 = 0.13) of the variance when variability due to clustering within-person was not accounted and 70 % (conditional R^2 = 0.70) of the variance when it was accounted. The ICCs for the multilevel multiple regression models for anxiety and depression were for 0.45 and 0.66, respectively.

Table 7
Multilevel multiple regression for the outcome variables (anxiety, depression) with independent variables entered simultaneously.

Anxiety	***β***	**95 % CI**	***P***
Intercept	0.42	0.14 - 0.69	< 0.01
Worry	0.45	0.40 - 0.51	< 0.001
Positive metacognitive beliefs about rumination	0.05	-0.02 - 0.13	ns
Negative metacognitive beliefs about worry and rumination	-0.01	-0.04 - 0.02	ns
Model summary			
Assessments: 589			
ICC = 0.45			
Marginal R^2 / Conditional R^2 = 0.36 / 0.65			
Depression	***β***	**95 % CI**	***P***
Intercept	0.65	0.22 – 1.08	< 0.01
Rumination	0.31	0.25 - 0.38	< 0.001
Positive metacognitive beliefs about rumination	0.04	-0.04 - 0.12	ns
Negative metacognitive beliefs about worry and rumination	-0.00	-0.04 - 0.03	ns
Model summary			
Assessments: 589			
ICC = 0.66			
Marginal R^2 / Conditional R^2 = 0.13 / 0.70			

Note. CI = confidence interval; ICC = intraclass correlation coefficient; ns = non-significant; β = unstandardized beta coefficient

3.5. Sensitivity analysis

The inclusion of the corresponding baseline measure of the outcome variable in the multilevel multiple regression models had little impact on the results. None of baseline measure were significantly independently associated with the relevant outcome variables (see Appendix D).

4. Discussion

This is the first study to explore the potential utility of the S-REF model [35,36] for explaining emotional distress (i.e., anxiety and depression) in YPwE. It is also the first study to explore the fluctuating nature of emotional distress and theoretically related constructs in YPwE by using an innovative methodology, ESM.

The first aim of our study was to explore whether metacognitive beliefs were associated with worry and rumination. All three metacognitive beliefs domains (i.e., positive metacognitive beliefs about worry, positive metacognitive beliefs about rumination, negative metacognitive beliefs about worry and rumination) were significantly positively associated with worry and rumination, accounting for 55–59 % of the variance in worry and rumination when variability due to clustering within-person was accounted for. These findings are in-line with predictions of the S-REF model and build on previous findings in adults with epilepsy and adults with other physical health populations [19,39,79,80].

To assess the independent association between each metacognitive belief domain and worry and rumination, multilevel multiple regression models were constructed. Negative metacognitive beliefs about worry and rumination were independently positively associated with worry. Positive metacognitive beliefs about rumination and negative metacognitive beliefs about worry and rumination were independently positively associated with rumination. Collectively, all three metacognitive belief domains respectively accounted for 56 % and 60 % of the variance in worry and rumination when variability due to clustering within-person was accounted for. These findings support the utility of the S-REF model in YPwE and are the first to demonstrate that metacognitive beliefs about rumination are associated with rumination in a physical health population.

Positive metacognitive beliefs about worry *were not* independently associated with worry but positive metacognitive beliefs about rumination *were* independently associated with worry. This is unexpected according to the S-REF model. While worry and rumination are both central components of the CAS, positive metacognitive beliefs about worry are primarily expected to lead to worry whereas positive metacognitive beliefs about rumination are primarily expected to lead to rumination. While worry and rumination have traditionally been conceptualised as distinct concepts, they correlate highly and share common processes [81–83]. As such, worry and rumination have been conceptualised as part of a wider transdiagnostic construct named 'repetitive negative thinking' [84,85]. If worry is part of a wider transdiagnostic construct, this would explain why positive metacognitive beliefs about rumination were associated with worry. However, this does not explain why positive metacognitive beliefs about worry were not independently associated with worry. This finding could be due to positive metacognitive beliefs about worry being involved in the worry process at a different stage. According to the S-REF model, positive metacognitive beliefs about worry are primarily involved in initiating worry as opposed to maintaining it. Associations between variables in our study were only compared within the same assessment period. This lack of association between worry and positive metacognitive beliefs about worry when assessed at the same time-point has been found in other studies [39,53,80]. Unfortunately, our sample size precluded us from investigating the relationship between variables at different time-points (i.e., whether metacognitive beliefs about worry at assessment 1 predict worry at a later assessment point).

The second aim of our study was to explore whether metacognitive beliefs are associated with anxiety and depression. In-line with predictions of the S-REF model, two of the three metacognitive belief domains (positive metacognitive about rumination, negative metacognitive beliefs about worry and rumination) were significantly positively associated with anxiety and depression, accounting for 60–72 % of the variance in anxiety and depression when variability due to clustering within-person was accounted for. However positive metacognitive beliefs about worry were not significantly associated with anxiety or depression. This is not in line with predictions of the S-REF model. The same methodological considerations described earlier regarding the time-point at which positive metacognitive beliefs are important may apply.

The final two aims of our study were to explore whether worry and rumination were respectively associated with anxiety and depression (aim 3); and whether metacognitive beliefs explained additional variance in anxiety and depression after accounting for worry and rumination, respectively (aim 4). Worry and rumination were significantly positively associated with anxiety and depression. The strongest associations were found between worry and anxiety, and rumination and depression (as would also be expected according to the S-REF model). These findings are in-line with the well-established evidence-base demonstrating that perseverative thinking in the form of worry and rumination are prominent in those presenting with anxiety and depression [86–89].

After respectively accounting for worry and rumination, none of the metacognitive belief domains explained additional variance in anxiety and depression. The S-REF model proposes that the relationship between positive metacognitive beliefs about worry and rumination and anxiety and depression should be fully mediated by worry and rumination. Thus, it is unsurprising that positive metacognitive beliefs about worry and rumination did not account for additional variance in anxiety and depression after controlling for worry and rumination. Alternatively, the S-REF proposes that the relationship between negative metacognitive beliefs about worry and rumination and anxiety and depression should only be partially mediated by worry and rumination - negative metacognitive beliefs about worry and rumination lead to anxiety and depression both directly and indirectly (via worry and rumination). Thus, it is surprising that negative metacognitive beliefs about worry and rumination did not account for additional variance in anxiety and depression after controlling for worry and rumination.

One reason for this finding could be due to the low levels of anxiety and depression reported by the sample, as this may have resulted in insufficient variance in anxious and depressive symptoms (leading to floor effects). Moreover, the S-REF model was primarily designed to explain emotional distress in clinically distressed samples. While it is not uncommon for individuals who do not meet diagnostic criteria for clinical levels of anxiety and depression to hold positive metacognitive beliefs about worry and rumination [39], according to the S-REF model, it is less common for such individuals to hold negative metacognitive beliefs about worry and rumination. While holding positive metacognitive beliefs would be expected to lead to increased anxiety and depression (indirectly via worry and rumination), individuals are more likely to terminate worry and rumination if they believe such processes are controllable (i.e., if they do not hold negative metacognitive beliefs). Thus, while the S-REF model asserts that negative metacognitive beliefs are of most direct importance in the maintenance and exacerbation of anxiety and depression, negative metacognitive beliefs may only arise in individuals who meet diagnostic criteria for anxiety or depression. This notion is supported by the relatively low scores on the ESM scale measuring negative metacognitive beliefs about worry and rumination in our sample (see Table 2). It is also supported by findings in previous studies. Benedetto et al., (2022) [90] found that negative metacognitive beliefs about worry were associated with anxiety in a clinical sample of adolescents but not in a non-clinical sample of adolescents. Moreover, Papageorgiou & Wells (2003) [91] tested the fit of the S-REF model in depressed and non-depressed participants. While the S-REF model fitted

the data well for the depressed sample, it did not fit well for the non-depressed sample.

Another reason for this finding could be due to amalgamating negative metacognitive beliefs about worry and rumination into a unitary scale. According to the S-REF model, negative metacognitive beliefs about worry would be expected be more closely related to anxiety whereas negative metacognitive beliefs about rumination would be expected to be more closely related to depression. If a participant held strong negative metacognitive beliefs about worry but not rumination, this may have been masked. Due to the wording of items in the ESM assessment, it was not possible to separate out these constructs (i.e., both items assessing negative metacognitive beliefs referred to metacognitive beliefs about both worry and rumination; see Appendix A). To assess the S-REF model more accurately, future studies may benefit from recruiting a sample of YPwE who are clinically anxious or depressed and separating out negative metacognitive beliefs about worry and rumination into two distinct constructs.

4.1. Strengths and limitations

The use of ESM enabled the assessment of the relationship between metacognitive beliefs, perseverative thinking (i.e., worry and rumination) and emotional distress (anxiety and depression) in YPwE at a momentary level. This enabled the identification of more complex and dynamic relationships between variables that may be unaccounted for using traditional research designs. The high level of within-person variability for the relationships between metacognitive beliefs, perseverative thinking, and emotional distress demonstrates that such variables appear to fluctuate over short intervals, providing support for the use of more nuanced methodologies which account for this variability, such as ESM. However, future studies would benefit from combining different assessment methods (i.e., baseline and ESM measures) into multilevel analyses. This would require a larger sample size.

ESM assessment compliance rates were high, suggesting the use of ESM is a feasible approach in YPwE. Several strategies were implemented to increase compliance. First, the 10-day ESM schedule was informally piloted on eleven members of a young person's advisory group (Generation-R). Based on their feedback, the language used in the ESM assessment was revised (e.g., wording of questions was changed from second-person to first-person and a uniform five-point Likert scale was implemented for responses across all ESM items). Second, as recommended by Hektner et al. (2007) [92], we ensured participants understood the questions in the ESM assessment, we let them complete an example ESM assessment on their smartphone to familiarise themselves with the ESM procedure, and we arranged a 'check-in' call with each participant during the study (to ensure there were no technical issues and to answer any questions regarding the ESM procedure). We also offered to provide participants with a letter about the study which they could provide to their school or college to increase their likelihood of being able to complete prompts during the day. Delivering the ESM assessments via an app on participants smartphones opposed to other common delivery methods (i.e., paper-pen method or providing participants with a palmtop device) also may have increased compliance due to ease of access.

Apart from negative metacognitive beliefs about worry and rumination, the relationship between ESM measures and their corresponding baseline measures were non-significant. This may have been due to type II error. As the correlation analysis was based on average ESM scores, the current sample size (n = 18) was likely underpowered [93]. However, this does not explain the weak correlation coefficients between most ESM measures and their corresponding baseline measures. The weak coefficients could suggest that the ESM assessments were not validly assessing what they were intending. Yet, significant moderate-to-large correlations were found between most ESM measures and theoretically related constructs (measured via ESM), demonstrating convergent validity of the ESM measures. Alternatively, it could be that baseline and ESM measures were capturing different aspects of such constructs, which has been demonstrated in prior research (i.e., retrospective measures of worry only account for a small amount of variance in daily worry) [46]. There was also a high level of within-person variability for most variables. Thus, using an average score for ESM measures in the correlation analysis may not be appropriate. Assessment of the reliability and validity of ESM measures is difficult [94–96]. Until well-established methods for assessing reliability and validity of ESM measures are developed, future studies would benefit from asking participants to what extent they think ESM items represents a certain construct [97].

Our analysis was constrained to cross-sectional associations (i.e., whether metacognitive beliefs at assessment 1 are associated with perseverative thinking at assessment 1). Therefore, causality cannot be assumed. Future studies should assess the mediational relationship between metacognitive beliefs, perseverative thinking (i.e., worry and rumination) and emotional distress (i.e., anxiety and depression) using time-lagged models (e.g., whether metacognitive beliefs at assessment 1 predict anxiety at assessment 3, and whether this relationship is mediated by worry at assessment 2). However, this would require a larger sample size.

Due to our small sample size (n = 18) the generalizability of our findings to the wider epilepsy population is unknown. None of the participants were experiencing 'severe' seizures, all but one participant identified as White British, and most of the sample (72 %) were female which may indicate potential nonresponse bias. As participants had to have access to a smartphone to participate, this also may have led to a biased sample. While no participants were excluded due to inaccessibility to a smartphone, it is possible that potential participants did not volunteer due to this requirement. To understand more about potential nonresponse bias, future studies would benefit from recording the proportion of participants screened ineligible and the proportion of participants who decide not to participate (along with reasons why).

Finally, we only assessed two metacognitive beliefs domains – positive and negative metacognitive beliefs. Thus, future studies would benefit from assessing other metacognitive beliefs domains, namely cognitive confidence (i.e., the belief that one's memory is unreliable). According to the S-REF model, reduced cognitive confidence may lead to increased worry and rumination and strengthen negative metacognitive beliefs [22]. Reduced cognitive confidence is associated with anxiety and depression in adults with epilepsy [39,40] and may be particularly important in YPwE given memory concerns reported [42]. Likewise, as we only assessed one aspect of the CAS - perseverative thinking (i.e., worry and rumination) - future studies would benefit from considering other CAS processes such as attentional focus on threat and unhelpful coping behaviours.

4.2. Clinical implications

Findings of this study provide support for the utility of the S-REF model for emotional distress in YPwE. Thus, MCT [22], which is underpinned by the S-REF model [35,36], may be an effective psychological intervention for YPwE experiencing anxiety and depression. It may also be an effective preventative intervention for those at risk of developing anxiety and depression; modifying maladaptive positive metacognitive beliefs may prevent YPwE from developing maladaptive negative metacognitive beliefs and subsequent anxiety or depressive disorders. Preliminary evidence indicates that MCT, which primarily aims to modify maladaptive positive and negative metacognitive beliefs, is an acceptable and effective intervention to reduce anxiety and depression in adults with physical health conditions [23–28], adolescents and young adults with cancer [29], and adolescents with common mental health disorders [30–34]. Prior to examining the effectiveness of MCT for anxiety and depression in YPwE, a better understanding of the temporal and mediational relationships between metacognitive beliefs, perseverative thinking, and emotional distress in a clinically distressed

sample of YPwE is recommended.

4.3. Conclusion

This study provides promising initial evidence for the utility of the S-REF model to understand emotional distress in YPwE. This suggests that MCT, which is underpinned by the S-REF model, may be an appropriate intervention for anxiety and depression in YPwE. To enable more fine-grained testing of the S-REF model for emotional distress in YPwE, replication of this study with a larger sample is warranted.

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CRediT authorship contribution statement

James Temple: Formal analysis, Investigation, Methodology, Writing – original draft, Writing – review & editing, Conceptualization. **Mary Gemma Cherry:** Conceptualization, Methodology, Supervision, Writing – review & editing. **Victoria Gray:** Conceptualization, Investigation. **Andrew Jones:** Formal analysis, Software, Writing – review & editing. **Peter Fisher:** Conceptualization, Methodology, Supervision, Writing – review & editing.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.yebeh.2023.109599.

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