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Review

Emotion dysregulation in patients with psychogenic nonepileptic seizures: A systematic review based on the extended process model

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ABSTRACT

Psychogenic nonepileptic seizures (PNES) are characterized by paroxysmal alterations in motor and sensory functions resembling epileptic seizures, but are not caused by epileptiform activity. In recent years, there has been increasing scientific interest in emotion dysregulation in patients with PNES (pwPNES), but the literature has not yet been interpreted within a broader model of emotion dysregulation. The aim of this review was therefore to synthesize the existing literature on emotion dysregulation in pwPNES within the extended process model (EPM) of emotion regulation.

Methods: PubMed and Web of Science were searched for studies relevant to emotion dysregulation as defined by the EPM. These studies were subjected to a bespoke quality appraisal tool. Studies of acceptable quality were categorized to the different stages of the EPM and critically appraised.

Results: Studies of emotion regulation in pwPNES were generally of low quality — a finding largely driven by small sample sizes. However, there was evidence of emotion dysregulation characterized by deficits in the identification of patients' own emotional states, as well as the selection and implementation of maladaptive regulatory strategies, and altered exteroceptive emotional processing. However, heterogeneity in findings suggests that emotion dysregulation is likely linked to other psychological factors and not common to all pwPNES.

Significance: This review suggests that while pwPNES are likely to experience emotion dysregulation as defined by the EPM, there is variability in the distribution of regulatory deficits in this patient population, and a person-centered approach should be taken when working with these patients. There is a need for more high quality and better-powered studies in this area.

1. Introduction

Psychogenic nonepileptic seizures (PNES) are assumed to be behavioral and experiential responses to aversive triggers [1]. Superficially resembling epileptic seizures but unassociated with epileptiform activity, PNES are relatively common, associated with long-term disability and a heavy economic burden [2]. Current biopsychosocial models attribute PNES to a complex interaction of predisposing, precipitating, perpetuating, and triggering factors — several of which relate to an individual's capacity to regulate their own emotions [3]. Relevant etiological factors include previous exposure to trauma, dissociation, coping, alexithymia, and insecure attachment [1]. While there is a growing trend for research on emotion dysregulation (ED) in patients with PNES (pwPNES), and an integrative psychological model has been proposed (e.g., [4]), observations of ED in pwPNES have not previously been placed within a more general theory of emotion regulation. We propose the extended process model (EPM) of emotion regulation as a potential model structure [5].

1.1. A summary of the extended process model of emotion regulation

Emotion regulation can be defined as the process by which a person modifies or controls what emotions they are experiencing, when they have them, and the nature in which emotions are experienced or expressed [6]. The EPM conceptualizes emotion regulation as a series of 'valuations' across three stages: identification, selection, and implementation. Each valuation also consists of three substeps, beginning with a representation of the internal or external environment (perception substep). The representation is compared against a goal–

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state (valuation substep), and if there is a sufficient discrepancy between the environment and the goal–state, the action substep is triggered. The functions of these substeps are constrained by the stage of the EPM that the valuation system is within. During identification, an ongoing emotion is identified (e.g., disgust), and a decision is made whether or not to regulate the emotion based on the discrepancy between the current emotion and a goal emotional state. During selection, a general emotion regulation strategy (e.g., attentional deployment) is decided upon. This triggers the implementation stage, during which the general regulatory strategy is translated into specific tactics suitable for the current situation (e.g., distraction from the disgust-eliciting stimulus). In the case of successful emotion regulation, the process cycles through again until the regulatory goal is reached (e.g., the individual is no longer feeling disgusted), in a continuous, multimodal, and iterative process. Emotion dysregulation can be thought of as a disruption at any substep of these stages.

The EPM is the most recent iteration of the widely used and tested Process Model of Emotion Regulation [7] and has already been used to conceptualize ED in a variety of populations and contexts including (but not limited to) psychopathology in general [8]. The potential value in interpreting studies of ED in pwPNES within an explicit theoretical framework such as the EPM is that i) it may allow for a more precise understanding of how and why ED occurs, and ii) it may allow for a more direct comparison against ED in other populations. The aim of this review was therefore to synthesize the existing literature on ED in pwPNES according the EPM [5].

2. Methods

A systematic review was conducted. Data were reported according to the Preferred Reporting Items for Systematic Reviews and Meta-analysis (PRISMA) checklist [9]. The definition of ED was based on the EPM [5]. The diagnosis of PNES does not map neatly onto any one of the nosological categories of the Diagnostic and Statistical Manual of Mental Disorders, 5th Edition: DSM-5, but usually fulfils the diagnostic criteria of Functional Neurological Symptom (Conversion) Disorder (FND) [10] as a Conversion Disorder falling under the subtype of "with attacks or seizures". The diagnostic process clinically defining PNES has been outlined in a consensus paper by the International League Against Epilepsy [11].

2.1. Search strategy

The electronic databases PubMed and Web of Science were searched on 18th January 2018 (years 1894–2017). Search terms relating to PNES were taken from a recent review article on PNES [12]. Search terms relating to ED were taken from or synonymous with keywords in the EPM [5]. Please see additional web content for more details of the search terms used. Further articles were identified from the reference list of papers identified during the electronic database search.

2.2. Study selection

Article titles and abstracts were screened for relevance to the review topic and compared against inclusion and exclusion criteria by IW. Only peer-reviewed original research reports were included. All other types of publications and articles not written in English were excluded from the review. Studies not directly relevant to the mechanism of ED in pwPNES only (i.e., explicitly defined case groups with mixed FND or with comorbid epilepsy) were also excluded at this stage. Studies of pediatric populations or treatment for PNES, and studies that focused on patients' support networks were considered outside the scope of the review. The remaining full-text articles were read in full by IW and MR. Articles were excluded if dependent variables were not quantitative standardized psychological measures and were therefore incompatible with the quality rating system [13, 14] or if the methodologies used did not directly relate to a stage of the EPM [15, 16]. Finally, each article was clustered to one or more specific stages of the EPM according to the methodologies or measures used. The categorization of each study into stages of the EPM was proposed by IW and confirmed by LL.

2.3. Quality assessment

Eligible articles were rated according to an appraisal tool designed specifically for quantitative psychological research in this field [1], which clarifies whether i) all diagnoses were video-EEG confirmed (yes/no), ii) epilepsy was explicitly ruled out in the group with PNES (yes/no), iii) there was reference to a procedure ensuring PNES were not misdiagnosed panic attacks (yes/no), iv) recruitment was consecutive (yes/no), and v) dependent variables were standardized (yes/no). Number, type, and gender ratio of control groups (where appropriate) were recorded to ensure groups were matched and did not have PNES (yes/no) (a difference in gender ratio of <10% or mean age difference of <five years between-groups was considered matched). Very few studies presented formal power calculations justifying sample sizes; therefore, we rated sample size according to the power and effect size conventions proposed by Cohen [17] and used in a previous systematic review of PNES [1]. Sample sizes for studies were rated as being very poor (<15 participants in each group; i.e., <80% power to detect a very large effect size, Cohen's $d = 1.1$), poor (<26 participants in each group; i.e., <80% power to detect a large effect size, $d = 0.8$), moderate (26–63 participants in each group; i.e., ≥80% power to detect a large effect size, $d = 0.8$), or good (≥64 participants in each group; i.e., ≥80% power to detect a medium effect size, $d = 0.5$), assuming a two-tailed independent *t*-test with alpha = 0.05. Study quality was calculated from these eight different quality criteria and sample power [1]. To establish interrater reliability, each article was rated by IW and MR. Any disagreements on ratings were resolved following discussion. Studies rated as 'unacceptable' were subsequently excluded from the review.

3. Results

3.1. Quality assessment

Fifty one papers were identified in the search (Fig. 1), all of which were included in the quality assessment (Appendix 1). 'Unacceptable' ratings were given in four (7.8%), 'low' in 24 (47.1%), 'medium' in 20 (39.2%), and 'high' in 3 (5.9%) studies. The median case group size was 30, but only 52% of studies (case–control design) were adequately powered (defined as moderate or good power). Sixteen of the 24 low quality studies would have been rated medium quality if they had a sample size ≥26. Likewise, the four excluded studies were deemed inadmissible because of a sample size <15; all of these studies would have been classed moderate to high quality on the basis of the other quality criteria. In terms of individual quality rating criteria, all dependent variables were standardized in 92%; an explicit reference to epilepsy being ruled out was made in 77%; all PNES cases were confirmed with video-EEG in 71%; anxiety attacks were ruled out in 51%, and patients were recruited consecutively in 37% of studies subjected to the quality review. Forty-seven studies were included in the final review.

3.2. Categorization of studies

Twenty-three studies were categorized as relevant to the identification stage of the EPM because the measures and methodologies used captured participants' identification of their own emotions (Appendix 2). Thirty-three studies were deemed relevant to the selection and implementation stages.