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Is there a creative functional paradoxical facilitation in juvenile myoclonic epilepsy?

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ABSTRACT

Objective: In patients with juvenile myoclonic epilepsy (JME), a specific personality profile suggestive of frontal lobe dysfunctions has been described. From a neurobiological point of view, the frontal lobe seems to be crucial for creative processes, although the exact role remains unclear. The theory of creative paradoxical functional facilitation (PFF) assumes that disinhibited frontal lobe function can enhance creative abilities. The aim of the current study was to explore our hypothesis that JME is associated with higher artistic creativity based on the theory of PFF.

Methods: We assessed 25 patients with JME aged 18 to 40 years in regard to neuropsychological creativity testing. Results were compared with those of 25 age-, sex-, and level of education-matched healthy control subjects (HC) and patients with temporal lobe epilepsy (TLE). Creative abilities were assessed using two validated and standardized tests: 1) nonverbal: the incomplete figure task of *Torrance Test of Creative Thinking* and 2) verbal: *verbal creativity test*. Additionally, a basic assessment of fluid intelligence (test for problem solving) and frontal lobe function (*trail-making test*) was administered to all participants.

Results: Verbal creativity was impaired in both groups with epilepsy compared with that in HC (specific score: JME vs. HC, $p = 0.008$; TLE vs. HC, $p = 0.003$). In regard to nonverbal creative abilities, both groups with epilepsy exhibited fair performance. Level of fluid intelligence was even in all groups ($p = 0.433$). Only patients with JME showed deficits in the frontal lobe test of psychomotor speed (time in seconds: 67.7 JME vs. 54.6 TLE vs. 52.8 HC; $p = 0.045$).

Conclusions: Overall, our study did not reveal increased creativity in JME. The current findings provide insights into creative abilities in two different epilepsy syndromes. Knowledge on specific neuropsychological strengths or deficits in patients with epilepsy may be useful for treatment or counseling.

1. Introduction

Juvenile myoclonic epilepsy (JME) is a common, well-defined subsyndrome of idiopathic generalized epilepsy accounting for up to 10% of all epilepsies [1]. Seizures commonly follow a circadian rhythm with preponderance upon awakening; the electroencephalogram (EEG) typically shows bilateral polyspike waves with a frontocentral emphasis [2]. A specific personality profile associated with JME was first anecdotally described in early reports by Janz & Christian, including social immaturity, disinhibition, and lack of endurance [3]. According to these early observations, further neuropsychological studies reported subtle cognitive impairments of frontal functions such as general cognitive abilities; attention; episodic, working and prospective memory; and executive functions in patients with JME [4–7]. Additionally, brain imaging studies in patients with JME demonstrated supportive findings, e.g., structural abnormalities and glucose hypometabolism involving the prefrontal cortex (PFC) [8–10]. This particular frontal lobe structure also plays a crucial role in creative processes.

Creativity is a multifaceted construct and can be defined as "the ability to produce a work that is both novel (i.e., original, unexpected) and appropriate (i.e., useful, adaptive concerning task constrains)" [11]. Based on theoretical approaches to creative thinking, this ability relies on cognitive skills such as problem solving, mental flexibility, or working memory [12], all of which are mostly attributed to the PFC [13]. Frontal lobe activation during creativity tasks has been indeed demonstrated in several neuroimaging studies [14–19]. In keeping with this hypothesis, some human experimental studies showed impaired creativity in connection with frontal lobe injury [20–22]. A

Abbreviations: AED, antiepileptic drug; EF, executive function; FTLD, frontotemporal lobar degeneration; HC, healthy controls; IGE, idiopathic generalized epilepsy; JME, juvenile myoclonic epilepsy; PFF, paradoxical functional facilitation; SD, standard deviation; TLE, temporal lobe epilepsy; TMT, trail-making test; TTCT, Torrance Test of Creative Thinking; VKT, verbal creativity test.

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study on 17 patients with frontotemporal lobar degeneration (FTLD) reported worse results in most creative abilities, but these patients also exhibited some responses that were more original. The authors discussed the increased originality as a possible result of social disinhibition in affected patients, as they showed more sexually oriented responses than healthy controls. The more sexually oriented answers of FTLD patients appeared as a higher originality in creativity testing [23]. Additionally, a series of clinical observations reported striking improvement in creative abilities or even de novo artistic creativity (e.g., painting, writing, music) in patients with medical conditions affecting the frontal lobe. These observations mainly concerned neurodegenerative diseases or stroke [12]. In this context, the theoretical concept of a *paradoxical functional facilitation* (PFF) was proposed. This describes the phenomenon that a lesion evokes gains of functions – by a release of inhibition – rather than causing deficits [24–26].

In summary, there are two opposed theories in regard to the frontal lobe's role in creativity: one considering that creative abilities require an unimpaired frontal lobe function and another one assuming that disinhibited frontal lobe function can enhance creative abilities. Concerning these two theories, we considered that the systematic assessment of creative abilities in patients with JME with presumed frontal lobe dysfunction might be of interest. Therefore, our hypothesis was that patients with JME exhibit, in line with the theory of PFF, greater creativity or originality compared with healthy controls. To control for adverse effects of antiepileptic drugs or other epilepsy-related factors such as burden of recurrent seizures or social stigma, we also tested patients with temporal lobe epilepsy (TLE).

2. Methods

2.1. Sample and procedures

Three different groups were included in the study: the index group of 25 patients with JME and, furthermore, 25 healthy controls and 25 patients with TLE representing the epilepsy control group. Patients with epilepsy were identified in the outpatient database of our institution. Healthy controls and patients with TLE were matched to the index group of patients with JME in regard to age, sex, and educational level. Thirteen patients with TLE had a left-sided focus, 3 patients had a right-sided focus, and 4 patients had bilateral seizure onset zones. In 5 patients, lateralization of the seizure focus was not possible. Patients with epilepsy exhibited semiological and EEG features typical of JME or TLE and had been diagnosed by experienced epileptologists according to the criteria of the *International League Against Epilepsy* [27]. Healthy controls did not suffer from any neurological or psychiatric disorders. As creativity may vary over the lifespan, subjects' age was limited to 18–40 years. Additionally, participants had to be German native speakers in order to exclude a language barrier that may confound results in verbal creativity testing.

All subjects signed informed consent before testing. The study was approved by the Institutional Review Board of *Charité–Universitätsmedizin Berlin*.

2.2. Neuropsychological evaluation: fluid intelligence, frontal lobe

All participants underwent a short neuropsychological assessment of fluid intelligence (a specific German-speaking test for problem solving with geometric figures termed *Leistungsprüfungssystem-Untertest 3*) in order to examine general intellectual abilities. The *trail-making test* (TMT) assessed psychomotor speed and mental flexibility. The intelligence test with geometric figures is scored by counting the number of correct responses that is transferred into a standard value. For the TMT, the total time (in seconds) that is needed to complete the trail is used to assess part A (psychomotor speed) and part B (psychomotor speed and mental flexibility).

2.3. Creativity testing

The validated German *verbal creativity test* (VKT) was designed to quantify verbal productivity [28]. In nine different tasks, subjects were requested to write down as many answers as possible. In these subtasks, the principle of diverted thinking was used to measure creative thinking; one given task could result in many different correct answers (e.g., alternative uses of an empty can or different consequences of a utopian situation). The VKT quantifies the verbal productivity of divergent thinking by counting the number of responses. It provides six subscales which form the *verbal creativity index* by the arithmetic mean.

The *Torrance Test of Creative Thinking* (TTCT) is the most widely used and well-known measure of creativity [29]. The incomplete figure task of the TTCT was used to study nonverbal creative abilities. Subjects were asked to complete ten different stimulus figures by sketching or drawing an object. The instruction contained an explicit demand to create something novel, specifically something that presumably nobody drew before. In the incomplete figure task of the TTCT, five general creative abilities were scored for each drawing: *originality*, *fluency*, *elaboration*, *resistance to premature closure*, and *abstractness of titles*. Additionally, a *checklist of creative strength* was also evaluated, including the following items: *emotional expressiveness*, *storytelling articulateness*, *movement or action*, *expressiveness of titles*, *unusual visualization*, *internal visualization*, *breaking boundaries*, *humor*, *richness of imagery*, *colorfulness of imagery*, and *fantasy*. Two author-proposed strengths were not used in our scoring called *synthesis of incomplete figures* and *synthesis of lines or circles*, as they did not occur in any given responses of the participants.

In order to avoid a subjective bias in the evaluation, the TTCT's measures were scored by four independent judges, all of whom were blind to the group the participant belongs to. For statistical analysis, we used the mean scores of all four jury members.

2.4. Statistical analysis

Data were analyzed using SPSS, Version 21.0 (IBM Corp. Armonk, NY). As the sample size of our groups ($n = 25$) did not fulfill the conditions of the central limit theorem and we therefore could not assume a homogeneity of variance, we analyzed the data by using non-parametric tests. For group comparisons, chi-square and Kruskal–Wallis tests were used. Pairwise comparison was calculated using Mann–Whitney U comparison. In the case of multiple comparisons, Bonferroni correction was conducted. If not stated otherwise, the level of significance was set at $p < 0.05$. In the group of patients with JME, the associations of clinical variables (e.g., age, gender, presence of anticonvulsant therapy, and mono- vs. polytherapy) with the creative ability in our testing were calculated using Mann–Whitney U test. The association of the duration of epilepsy with the results in the VKT was analyzed by Spearman correlation coefficient r. Univariate general linear model was used to analyze further group differences in creativity regarding certain clinical variables (e.g., education).

We further analyzed if results of the VKT correlated with findings of the frontal lobe test and the intelligence test for problem solving by using Spearman correlation.

3. Results

3.1. General data on study population

Clinical and demographic data of the subjects included are displayed in Table 1. Groups did not differ significantly in regard to age, sex, or educational level. Antiepileptic medication was noted for the time point of testing in order to detect possible confounders. In the group with JME, significantly more patients were treated with AEDs than patients with TLE ($p = 0.037$). Additionally, the duration of epilepsy was