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Parental stress in pediatric epilepsy after therapy withdrawal

Francesca Felicia Operto [a,b,*], Roberta Mazza [a], Grazia Maria Giovanna Pastorino [b,c], Stella Campanozzi [a], Lucia Margari [a], Giangennaro Coppola [b]

[a] Child Neuropsychiatry Unit, Department of Basic Medical Sciences, Neuroscience and Sense Organs, University of Bari "Aldo Moro", Piazza Giulio Cesare 11, Bari, Italy
[b] Child and Adolescent Neuropsychiatry, Medical School, University of Salerno, Via Giovanni Paolo II, 132, Fisciano, SA, Italy
[c] Department of Mental and Physical Health and Preventive Medicine, University of Campania, Naples, Italy

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ABSTRACT

Objective: The objective of the study was to explore stress levels in the parents of children with idiopathic epilepsy at different time points of the disease, specifically, at the time of diagnosis, during follow-up, and 1 and 2 years after discontinuation of antiepileptic drugs.
Methods: Our study included 50 patients between 5 and 14 years of age, who were diagnosed with childhood absence epilepsy or idiopathic focal epilepsy with Rolandic paroxysms. Parents of the participants independently completed the Parenting Stress Index-Short Form at the time of initial diagnosis, and when the children started antiepileptic drugs (Time 0), and at 1 year (Time 1) and 2 years (Time 2) after discontinuation of therapy.
Results: At Time 0, parental stress levels were increased, both in mothers and fathers, with average scores in the "clinical range" of the parental distress (PD), dysfunctional parent–child interaction (P-CDI), and total stress (TS) scales. At Time 1, the scores on these scales remained high. At Time 2, a mild reduction in the stress scores was observed in both parents, despite values remaining in the "clinical range" for all the scales.
Conclusions: Results suggested that parents of children with epilepsy were not reassured about the child's condition, even after clinical improvement. Parental stress levels remained higher than expected, even 2 years after the discontinuation of therapy and freedom from seizures. This was probably due to concerns with the reappearance of new seizures or a more severe type of epilepsy with the discontinuation of drug(s), and feelings of inadequacy with their parental role(s).

1. Introduction

It is widely known that parents of children with chronic conditions experience high levels of stress [1,2]. For example, an increased level of stress has been detected in the parents of children with severe autism spectrum disorders [3], diabetes [4–7], and asthma [8]. In all these conditions, parents must daily cope with the burden of the necessary care of the child owing to the illness and with the constant adjustment to the changing demands of the chronic disease [1].

This is especially true for epilepsy, one of the most common neurological disorders of childhood [9,10]. Diagnosis of epilepsy in a child generates significant stress in the parents, which remarkably impacts the daily routine of the entire family [11,12]. Parents of children with epilepsy experience several significant worries regarding the epilepsy diagnosis, future seizures, the effects of seizures on the brain, possible adverse effects of antiepileptic drugs, comorbidities, and requisite lifestyle changes [13]. Stress may even be more troublesome in parents of children with intractable epilepsy. Although, most children with epilepsy achieve satisfactory control of the disease, approximately 20% continue to experience seizures despite medical therapy [9]. Moreover, children with severe epilepsy are at high risk of developing behavioral, mood, and sleep disorders, which in turn can increase the stress in a parent–child relationship [9]. Studies have shown that parents of children with epilepsy—particularly intractable epilepsy—also experience restriction in their social and recreational activities [13]. Moreover, they become more prone to sleep disturbances [14].

Abbreviations: DC, difficult child; MRI, magnetic resonance imaging; P-CDI, dysfunctional parent–child interaction; PD, parental distress; PSI, parental stress index; TS, total stress; VNS, vagus nerve stimulation.

* Corresponding author: Child Neuropsychiatry Unit, Department of Basic Medical Sciences, Neuroscience and Sense Organs, University of Bari "Aldo Moro", Piazza Giulio Cesare 11, Italy.
E-mail address: opertofrancesca@gmail.com (F.F. Operto).

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Table 1
Sample characteristics.

Sample size Patients = 50	Sex 20 males 30 females	Age 5–14 years (mean = 9.58 years; standard deviation = 2.14)	Diagnosis – Childhood absence epilepsy = 34 – Idiopathic focal epilepsy with Rolandic paroxysms = 16
Neurological objective examination – Normal in all patients	Psychomotor development – 1 language delay	Other pathologies – None	Nuclear magnetic resonance scan – Normal in all patients
Seizure frequency (at Time 0) – Monthly = 6 – Weekly = 7 – Multiple in week = 2 – Daily = 23 – Multiple in day = 12	Therapy – Monotherapy = 43 – Polytherapy = 7	Therapy response – All patient controlled	Adverse effects – 1 nausea – 1 stomach ache – 1 irritability

In the present study, we explored stress levels in the parents of children with idiopathic epilepsy at different time points of the disease course, specifically, at the time of diagnosis, during follow-up, and 1 and 2 years after the discontinuation of antiepileptic drugs.

2. Methods

2.1. Participants

The study cohort included 50 patients between 5 and 14 years of age (mean: 9.58 ± 2.14 years), who were recruited from the Child Neuropsychiatry Unit of the University of Salerno (Fisciano, Italy). Patients were brought for diagnostic assessment and therapeutic follow-up. Thirty-four patients were diagnosed with childhood absence epilepsy, and 16 were diagnosed with idiopathic focal epilepsy with Rolandic paroxysms.

A detailed history of the study participants was taken to assess psychomotor development, number of seizures, and information from the brain magnetic resonance imaging (MRI). In addition, neurological examination was performed in all the patients.

No subject showed abnormalities in the neurological examination or in the brain MRI; normal psychomotor development was reported for all patients, except for one who had a history of language delay.

Forty-three of the 50 patients were treated with only one antiepileptic drug while 7 were administered 2 antiepileptic drugs during the follow-up period. All the patients were seizure-free at 1 year and at 2 years after discontinuation of pharmacological therapy. Other factors related to family members were considered, including maternal age, level of maternal education, mother's occupation, and number of siblings (Table 1).

Parents of participants independently completed the Parent Stress Index (PSI)–Short Form at Time 0 (the time of diagnosis and commencement of antiepileptic drug therapy), at Time 1 (1 year after discontinuation of therapy), and at Time 2 (2 years after discontinuation of therapy), within the normal follow-up period.

After an interview to provide information on the study procedure, written informed consent was obtained from all the participants. The study was approved by the local ethics committee.

2.2. Parental stress index (PSI)

The PSI is a standardized tool that measures the stress level in a parent–child interaction. It is a 36-item scale, in which each item is graded on a 5-point Likert scale. The PSI yields scores of parenting stress across the following four domains:

- Parental distress (PD): defines the level of PD derived from factors related to parental role;
- Dysfunctional parent–child interaction (P-CDI): assessed the parental perception of a child who does not respond to the family expectations and of an interaction neither reinforcing nor rewarding the child;
- Difficult child (DC): defines parental stress based on some characteristics of the child's behavior;
- Total stress (TS): obtained from the sum of other scores, but can be interpreted as a stress index related only to the parental role.

Scores ≥85 are considered in the "clinical range", those <85 are in the "normal range" [15].

2.3. Statistical analyses

All clinical variables were subjected to statistical analyses. The neuropsychometric results are expressed as mean ± standard deviation. To evaluate significant differences in mean scores, between-group comparisons were performed using the two-tailed independent sample t-test. The two-tailed bivariate Pearson correlation test was performed to evaluate the correlation between scores of the two parents. Analysis of variance (ANOVA) was used to evaluate the impact of other variables on the obtained scores in the sample; $p < 0.05$ was considered to be statistically significant. Statistical Package for Social Science (SPSS) version 23.0 (IBM Corporation, Armonk, NY, USA) was used for statistical processing.

3. Results

3.1. Stress levels at Time 0, Time 1, and Time 2

From the analysis of the PSI, administered separately to the parents at Time 0, the levels of parental stress were found to be increased both in mothers and fathers compared with the reference population. The mean scores fell in the "clinical range" of the PD, P-CDI, and TS scales. However, the mean scores of the DC scale were in the "normal range" (Table 2).

Table 2
Mean scores of parental stress in mothers and fathers at Time 0, Time 1, and Time 2 (the values >85 are in the pathological range). PSI = parental stress index; PD = parental distress; P-CDI = dysfunctional parent–child interaction; DC = difficult child; TS = total stress.

PSI subscales	Time 0		Time 1		Time 2	
	Mothers	Fathers	Mothers	Fathers	Mothers	Fathers
PD	**90.4** ± 7.88	**88.8** ± 9.1	**89.1** ± 8.6	**88.3** ± 7.7	**85.8** ± 7.6	**85.2** ± 7.8
P-CDI	**90.0** ± 9.6	**88.1** ± 10.4	**88.9** ± 8.8	**88.4** ± 7.5	**85.5** ± 9.8	**85.3** ± 7.7
DC	71.9 ± 12.3	73.8 ± 17	72.5 ± 13	73.6 ± 14.6	72.7 ± 11.7	73.1 ± 12.8
TS	**90.8** ± 8.1	**89.1** ± 8.6	**89.6** ± 8.4	**88.8** ± 7.7	**85.9** ± 8.4	**85.1** ± 7.5

Table 3
a) Two-tailed Student's t-test for unpaired data reveals that the mean values between parents do not significantly differ for any of the analyzed scales at Time 0, Time 1, and Time 2; b) Pearson correlation test reveals a positive and statistically significant correlation between different scores of mothers and fathers at Time 0, Time 1, and Time 2. PSI = parental stress index; PD = parental distress; P-CDI = dysfunctional parent–child interaction; DC = difficult child; TS = total stress.

PSI subscales	a) Student's t-test			b) Pearson correlation test		
	Time 0	Time 1	Time 2	Time 0	Time 1	Time 2
PD	$p = 0.350$	$p = 0.625$	$p = 0.697$	**$p < 0.0001$**	**$p = 0.001$**	**$p = 0.002$**
P-CDI	$p = 0.343$	$p = 0.760$	$p = 0.909$	**$p < 0.0001$**	**$p = 0.005$**	**$p = 0.004$**
DC	$p = 0.523$	$p = 0.691$	$p = 0.870$	**$p < 0.0001$**	$p = 0.057$	$p = 0.055$
TS	$p = 0.310$	$p = 0.620$	$p = 0.616$	**$p < 0.0001$**	**$p < 0.0001$**	**$p = 0.001$**

Bold values statistically significant at $p < 0.05$.

Both at Time 1 and at Time 2, the mean scores of parental stress (PD, P-CDI, TS) remained in the "clinical range" (Table 2).

Analysis of variance revealed that at Time 0, stress levels in both parents were not influenced by factors, such as the age and sex of the patients, diagnosis, number of seizures, or type of therapy ($p > 0.05$ for the PD, P-CDI, and TS scales in mothers and fathers).

The differences in the average scores between the two parents were not statistically significant at Time 0, Time 1, and Time 2 (two-tailed Student's t-test for unpaired data) (Table 3a). In addition, the Pearson correlation test revealed a positive and statistically significant correlation between different scores of mothers and fathers at Time 0, Time 1, and Time 2 (Table 3b).

3.2. Stress level variation over time

3.2.1. Stress levels at Time 1 compared with Time 0 in mothers

The Student's t-test for paired samples revealed that the differences between the scores pertaining to maternal stress at Time 1 compared with those at Time 0 were not statistically significant for any of the analyzed scales (PD, $p = 0.108$; P-CDI, $p = 0.195$; DC, $p = 0.686$; and TS, $p = 0.103$) (Fig. 1).

3.2.2. Stress levels at Time 2 compared with Time 0 in mothers

The analysis of maternal stress levels at Time 2 compared with that at Time 0, revealed a statistically significant reduction in mean levels of the PD, P-CDI, and TS scales (PD, $p < 0.0001$; P-CDI, $p < 0.0001$; TS, $p < 0.0001$); however, this difference was not statistically significant in the DC scale ($p = 0.569$) (Fig. 1).

3.2.3. Stress levels at Time 1 compared with Time 0 in fathers

The Student's t-test for paired samples revealed that the differences between paternal stress levels at Time 1 compared with that at Time 0 were not statistically significant for any of the analyzed scales (PD, $p = 0.462$; P-CDI, $p = 0.773$; DC, $p = 0.895$; and TS, $p = 0.627$) (Fig. 2).

3.2.4. Stress levels at Time 2 compared with Time 0 in fathers

The analysis of stress levels in fathers at Time 2 compared with that at Time 0 revealed that there was a statistically significant reduction in mean levels of the PD, P-CDI, and TS scales (PD, $p < 0.0001$; P-CDI, $p = 0.018$; TS, $p < 0.0001$); however, this difference was not statistically significant in the DC scale (DC $p = 0.654$) (Fig. 2).

4. Discussion

The aim of this study was to explore the stress levels of parents of children with idiopathic epilepsy at the time of diagnosis (Time 0), and at 1 year (Time 1) and 2 years (Time 2) after the discontinuation of antiepileptic drugs.

Parental stress levels at Time 0 were high in both mothers and fathers, compared with normative reference values (normal values < 85; clinical range > 85) (Table 2). Stress values were not dependent on factors, such as age, sex, diagnosis, number of seizures before start of therapy, type of antiepileptic drug, maternal age, level of maternal education, mother's occupation, or presence of other siblings in the family. These data suggest that, even in conditions considered "benign", such as childhood absence epilepsy or idiopathic focal epilepsy with Rolandic paroxysms, parental stress levels can be high.

A detailed analysis of the individual PSI scales revealed that there was a "clinical" increase in the mean scores of the PD, P-CDI, and TS scales while the mean values of the DC scale were in the normal range (Table 2). This suggests that parental stress is mainly linked to factors concerning parental role and disease management, but not to those related to the child's behavior. Furthermore, the stress levels of mothers and fathers were positively correlated (Table 3). This suggests that the perception of stress and the strategies to cope with it might be dependent on factors related to the parental couple rather than a single parent.

The evaluation of parental stress levels after 1 year (Time 1) and 2 years (Time 2) from the discontinuation of antiepileptic drugs yielded unexpected results.

In fact, contrary to what was expected, the average levels of parental stress at Time 1 were still high. Moreover, the mean scores of the PD, P-CDI, and TS scales remained in the "clinical range" for both mothers and fathers (Figs. 1 and 2). On the contrary, at Time 2, there was a mild reduction in stress levels in both mothers and fathers, with a statistically

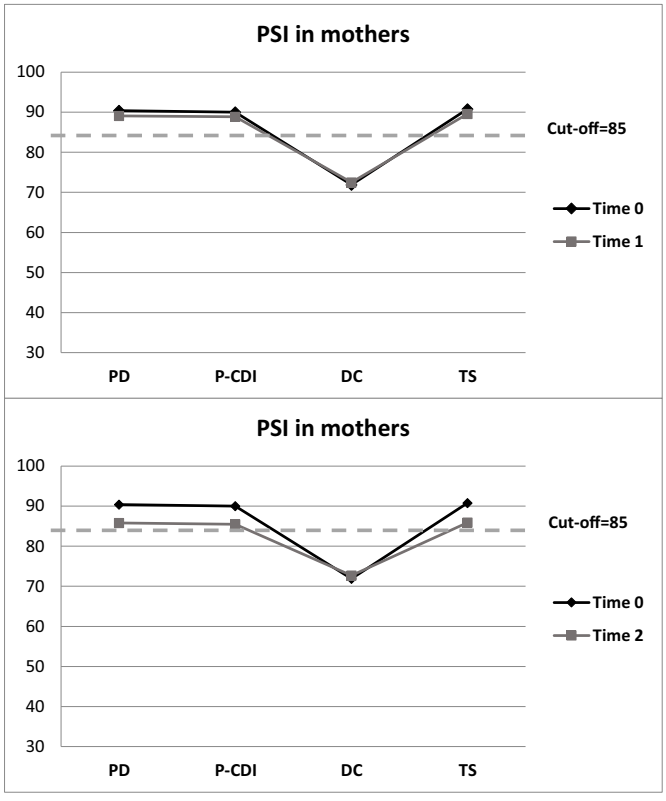

Fig. 1. Mean scores of stress in mothers at Time 2 and Time 1 compared with Time 0. PSI = parental stress index; the dotted line corresponds to cutoff = 85, scores >85 are in the "clinical range". PD = parental distress; P-CDI = dysfunctional parent–child interaction; DC = difficult child; TS = total stress.

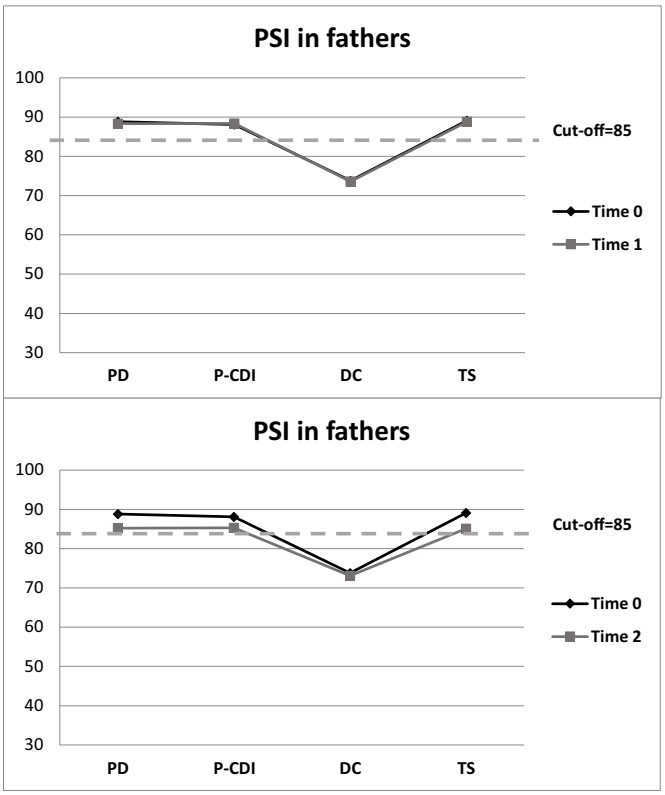

Fig. 2. Mean scores of stress in fathers at Time 2 and Time 1 compared with Time 0. PSI = parental stress index; the dotted line corresponds to cutoff = 85, scores >85 are in the "clinical range". PD = parental distress; P-CDI = dysfunctional parent–child interaction; DC = difficult child; TS = total stress.

significant difference compared with stress levels at Time 0. Nevertheless, mean PD, P-CDI, and TS values did not normalize and persisted in the "clinical range" compared with the normative reference population (Figs. 1 and 2).

To the best of our knowledge, no other study has explored parental stress in pediatric epilepsy after therapy withdrawal. Braams et al. evaluated stress experienced by parents before and 2 years after their children's epilepsy surgery and found that levels of stress decreased; however, it did not normalize in the first 2 years after surgery [16]. Li et al. evaluated stress in parents of children with refractory epilepsy before and after their children underwent vagus nerve stimulation (VNS) [17]; in this case, the authors found that parental stress significantly decreased. This is consistent with a study by Fan et al. [18], in which the authors compared PSI scores before and after VNS implantation in children with refractory epilepsy. They reported that VNS not only reduced the seizure frequency, but also the psychological burden on both parents and children.

It is difficult to draw definitive conclusions from the present study because of the inherent limitations owing to the small sample size, and to the fact that the participants were recruited from the same clinical site. Furthermore, other factors that may influence parental stress of a child diagnosed with epilepsy have not been analyzed. Thus, further studies with larger sample sizes are needed to confirm our results.

5. Conclusions

Our data suggest that parents were not fully reassured about their child's clinical condition, even after two years of drug withdrawal. Consequently, their stress levels remained higher than expected. We hypothesize that the fear of seizure recurrence or unexpected events and the end of a medical follow-up may, at least to some extent, explain these data. Hence, our study highlights the importance of adequate medical information to support and encourage coping behaviors in parents of children with epilepsy. A difficult parent–child relationship in a family with high parental stress levels may represent a risk factor for the further development of behavioral disorders in children [19].

Conflict of interest

The authors declare no conflict of interest.

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