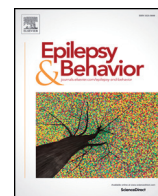


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The efficacy of an educational program for parents of children with epilepsy (FAMOSSES): Results of a controlled multicenter evaluation study



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ABSTRACT

Objective: The objective of this study was to evaluate the efficacy of the educational program FAMOSSES (modular service package epilepsy for families) for parents of children with epilepsy.

Method: Parents of children with epilepsy from Germany and Austria were included in a controlled prospective multicenter study using a pre–post design. Participants of the FAMOSSES program (FAMOSSES group, $n = 148$) completed a standardized questionnaire immediately before the program and six months later. The matched control group of parents not participating in the program ($n = 74$, matching ratio 2:1) also answered the questionnaire twice, at an interval of six months. The questionnaire comprised epilepsy-specific outcome measures (e.g., knowledge, coping, fears) and disease-related variables (e.g., seizure frequency). The generalized estimation equation approach was used for statistical analysis. In addition, parents' satisfaction with the FAMOSSES program was assessed six months after participation.

Results: Parents of the FAMOSSES group significantly improved in epilepsy-specific knowledge (group \times time interaction: $p < .001$), coping ($p < .01$), epilepsy-related fears ($p < .05$), and in speaking about epilepsy with their child ($p < .05$) compared with the control group. No effects were found on disease-related variables. Nearly all of the participants rated the FAMOSSES parents' program as “very good” (71%) or “good” (27%).

Conclusion: The efficacy of the FAMOSSES parents' program was confirmed. The results indicate that imparting knowledge and the interactive approach help parents in coping with their child's epilepsy and reduce epilepsy-related fears.

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1. Introduction

Epilepsy has a considerable impact not only on affected children but also on their families [1]. Besides having to cope with the unpredictable occurrence of seizures, children with epilepsy (CWE) show an increased risk of emotional and behavioral problems [2–4]. For parents, having a child with epilepsy is associated with stress, symptoms of depression and anxiety, and impaired quality of life, especially when the child has comorbid neurological deficits [5–7]. At the same time, parents' concern for their child with chronic illness often results in overprotective and restrictive parenting behavior, which may interfere with the development of their child's autonomy [8,9].

Thus, it is not surprising that, even when the child's epilepsy is well-controlled, the parents' need for psychosocial care and information is still high two years after diagnosis [10]. Although it was shown that a

higher level of knowledge is associated with fewer restrictions in family activities, less parental worries, and less perceived stigmatization and isolation [11], knowledge gaps are common in parents of CWE [12–16]. Parents of CWE are afraid of misinforming their child when talking about its disorder [17] and often feel apprehensive about letting their child participate in physical activities or school trips [14,18], so that improving parents' knowledge is an important means to help them cope with their child's epilepsy. Moreover, a higher parental need for psychosocial care is associated with less encouragement of their child's autonomy [10]. Therefore, a comprehensive epilepsy care plan that addresses not only medical aspects but also psychosocial needs of parents of CWE is recommended [19,20]. Educational programs for CWE and their parents are an important component in this context because they provide information and also cover coping strategies and emotional aspects of living with epilepsy [21].

The educational program FAMOSSES (*modulares Schulungsprogramm Epilepsie für Familien*, modular service package epilepsy for families) [22] is provided in outpatient settings in Germany, Austria, and Switzerland. It consists of a program for CWE aged 7–12 years and a

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matched parents' program. Participants of the children's program go on a virtual "cruise" with islands representing the modules (e.g., "treasure island" for the diagnostic process) and discover aspects of living with epilepsy playfully. The parents' program covers the same topics (basic knowledge, diagnostic procedures, therapeutic options, and informing others about epilepsy) but also includes additional modules on prognosis, development, and everyday life with CWE (e.g., reducing unnecessary restrictions). The course is interactive, and different techniques (e.g., mind maps, group discussions) are used to encourage parents to express emotions and to exchange experiences. More detailed information on the program is given elsewhere [22]. The two programs are independent insofar as parents can attend FAMOSES even if their child does not participate in the children's program (e.g., if the child is too young). Thus, FAMOSES is the only educational program for parents of very young or disabled CWE in Germany. All FAMOSES trainers have professional experience in working with CWE and are qualified as trainers. Besides improving participants' knowledge, FAMOSES aims at the reflection of attitudes and the development of strategies to facilitate coping and disease management.

During the development of FAMOSES, a pilot study was performed to evaluate the feasibility of the two programs which yielded some promising results [23,24]. Yet the sample sizes were relatively small, partly because of high dropout rates, and the follow-up period of three months was relatively short. Most of the questionnaires used were newly developed and could not be pretested, leading to ceiling effects and low reliability for some scales, thus limiting the validity of the results.

Meanwhile, the FAMOSES manual was revised, and structured "train-the-trainer" courses have been introduced as a qualification criterion. For these reasons, the aim of the current study was to investigate the efficacy of the FAMOSES parents' program with a modified research design, a larger sample size, a longer follow-up period, and improved instruments.

2. Methods

This report follows the CONSORT (Consolidated Standards of Reporting Trials) guidelines for nonpharmacologic treatments [25,26].

2.1. Study design and participants

We performed a prospective, open, multicenter study using a pretest–posttest–control group design to evaluate the efficacy of the FAMOSES parents' program. Two groups of parents of CWE were asked to answer questionnaires twice at an interval of six months (baseline and follow-up). The FAMOSES group participated in the FAMOSES parents' program directly after the baseline measurement, while the control group did not take part in FAMOSES during the study period.

A random allocation of parents to the study groups was not feasible for the following reasons: For one thing, we wanted to give all parents who were interested in FAMOSES the opportunity to participate (as soon as possible). Moreover, most cooperating epilepsy centers were not able to offer the program more than once a year whereas a randomized design with a waiting list control group would require a second FAMOSES course after six months. These centers would not have been able to participate in a randomized study, limiting both sample size and generalizability of the results. Therefore, the control group was recruited independently from the FAMOSES group (see Section 3.1). However, participants were matched based on demographic and disease-related variables at baseline to achieve structural equality of the two groups without randomization.

The participants were parents whose children were treated in one of the 19 cooperating epilepsy centers in Germany and Austria. Inclusion criteria comprised having a child with a verified epilepsy diagnosis. Additionally, the child was supposed to have had at least one seizure

in the previous year (active epilepsy). During the recruitment phase, it turned out that some participants of the FAMOSES parents' program had children with longer seizure-free periods. Since these children were still treated by a neurologist/neuropediatrician and were on anti-convulsant medication, the latter inclusion criterion was relaxed. Parents were excluded from the study if they had an insufficient knowledge of the German language or if their child had solely nonepileptic seizures. Only one baseline and one follow-up questionnaire per family were included in the statistical analysis.

Parents filled in the questionnaires anonymously and returned them to the Society for Epilepsy Research in a sealed envelope. Written informed consent was obtained from all parents, and the study was approved by the ethics committee of the Department of Psychology at Bielefeld University.

2.2. Intervention

Each FAMOSES course was conducted by two certified FAMOSES trainers (one of them being a pediatrician) and carried out according to the trainer's manual which provides information on course contents, materials and didactic techniques, and also allows some adjustment of contents based on the participating parents' needs (e.g., provided that all children are at school age, topics about kindergarten are skipped). All courses were delivered on weekends (2–3 days) with a total duration of 14 h and group sizes of 6–13 parents.

2.3. Objectives and hypotheses

Three primary outcomes that cover different aims of the FAMOSES parents' program were defined. We hypothesized that, compared with the control group, the participation in the FAMOSES parents' program would improve

- epilepsy-specific knowledge,
- parental coping with epilepsy, and
- encouragement of the child's autonomy.

Furthermore, we expected an improvement in the following secondary outcomes: epilepsy-related fears, information seeking and sharing of information, seizure management, speaking about epilepsy with the affected child, and impact of epilepsy on the family.

The effects of the FAMOSES parents' program on different disease-related variables (e.g., seizure frequency) were explored, and the satisfaction of parents with the FAMOSES program was assessed six months after participation.

2.4. Outcome measures and questionnaires

Epilepsy-specific questionnaires were developed based on the experience from the pilot study [23,24]. We selected items with good psychometric properties for our outcome measures and added some new items. To assess the impact of the child's epilepsy on the family, a short form of the German version of the Impact on Family Scale (IOFS) [27] was used. Except for the questionnaire on satisfaction with the FAMOSES program, all scales were presented both at baseline and at follow-up. Psychometric information and sample items are given in Table 1.

Additionally, parents were asked for age, marital status, education level, and employment status. Demographic and disease-related data of their children were also assessed (e.g., age, gender, type of schooling, development status, age of onset of epilepsy, and comorbidities).

2.4.1. Primary outcome measures

Epilepsy-specific knowledge. The scale developed to assess parents' knowledge about epilepsy consists of statements which the parents were asked to judge as true or false. A "don't know" category was

Table 1
Outcome measures.

Scales and questionnaires (<i>sample item</i>)	No of items	Answer categories	Cronbach's alpha ^a	ICC ^b	Explained variance (EFA) ^c
Epilepsy-specific knowledge ("Epileptic seizures always cause loss of brain cells.")	18	"True", "False", "Don't know"	.752	.834	26.88%
Coping with epilepsy ("I feel strong enough to cope with my child's epilepsy.")	7	4-point Likert ("strongly disagree" to "strongly agree")	.714	.506	33.92%
Encouragement of the child's autonomy ("I encourage my child to stand up to its epilepsy.")	7	4-point Likert ("strongly disagree" to "strongly agree")	.777	.832	44.76%
Epilepsy-related fears (2 subscales ^d):					53.58%
Fears about short-term consequences of the child's epilepsy ("I am afraid that my child can have a seizure at any time.")	8	5-point Likert ("not at all" to "very much")	.891	.765	
Fears about the future development of the child and its epilepsy ("I am afraid that my child's seizures will deteriorate.")	9	5-point Likert ("not at all" to "very much")	.913	.801	
Information seeking and sharing of information ("I seek contact to other parents whose child has epilepsy as well.")	5	7-point Likert ("never" to "always")	.737	.687	44.38%
Seizure management ("I move objects out of reach on which the child could hurt itself.")	5	7-point Likert ("never" to "always")	.720	.551	52.61%
Speaking about epilepsy with the affected child ("I speak to my child about the seizures.")	3	4-point Likert ("strongly disagree" to "strongly agree")	.820	.799	69.39%
Impact on Family Scale (IOFS) 11-item short form ("We see family and friends less often because of the illness.")	11	4-point Likert ("strongly disagree" to "strongly agree")	.936	.862	66.95%

^a Cronbach's α was computed for all parents included at baseline with complete data on the specific scale ($n = 245$ – 288).

^b ICC = intraclass correlation coefficient (for absolute agreement, single measures) computed for the participants of the control group at baseline and follow-up ($n = 61$ – 85).

^c EFA = exploratory factor analysis. For all scales except epilepsy-related fears, EFAs led to one-factor solutions.

^d Based on the exploratory factor analysis (correlation between factors $r = .651$).

added to reduce guessing, and the percentage of correct answers was analyzed.

Coping with epilepsy includes active dealing with the child's condition as well as the absence of feelings of resignation and hopelessness. An exploratory factor analysis (EFA) showed that both aspects belong to a single dimension. All items were taken from the scale "adaptation to epilepsy" that was used in the pilot study [23,24].

Encouragement of the child's autonomy: Parents were asked how far they encourage their child to be autonomous and to cope with its disorder actively, in contrast to an (over)protective behavior of the parents.

2.4.2. Secondary outcome measures

Epilepsy-related fears. The scale used in the pilot study [23,24] was divided into two subscales based on the results of an EFA: Fears about short-term consequences of the child's epilepsy (e.g., constant fear of potential seizures) and fears about the future development of the child and its epilepsy (e.g., negative long-term effects of anticonvulsant medication).

Information seeking and sharing of information: Parents were asked to what extent they talk with teachers or friends about their child's condition and how far they seek further information on epilepsy.

Seizure management refers to adequate emergency management of the parents during their child's seizure. Parents rated how often they show different kinds of possible reactions.

Speaking about epilepsy with the affected child includes conversations about seizures and medication.

Impact on Family Scale. This instrument assesses the impact that a child with chronic illness or disability has on the family. In this study, an 11-item short form of the German version that was validated for parents of CWE was used [27–30].

All scale scores were transformed into values between 0 and 100 except for the IOFS, for which the original value range (1–4) was retained. Higher scores indicate a higher manifestation of the respective construct, i.e., an increase in scores reflects a positive development for all scales except epilepsy-related fears and IOFS where higher scores represent more fears and more (negative) impact, respectively.

2.4.3. Disease-related variables

Seizure frequency was assessed with six categories (Table 2). Tolerability and efficacy of anticonvulsant drug therapy were rated by the parents using a scale with four categories (Table S2). We assessed the number of days the child was absent at school/kindergarten because of seizures in the last three months as well as consultation rates due to seizures.

2.4.4. Satisfaction with the FAMOSES parents' program

Participants of the FAMOSES group evaluated the material used during the course, their expectations, their acquisition of new knowledge and skills, and whether they would recommend the program to other parents.

2.5. Sample size calculation

The required sample size was determined a priori based on the following assumptions: As the family-wise error rate for testing the three primary outcome measures should not exceed $\alpha = .05$, we used a Bonferroni-corrected significance level of $\alpha' = \alpha / n$ (tests) = $.05 / 3 = .017$. The expected correlation of $r = .70$ between baseline and follow-up measurement was derived from the pilot study [23,24], and the power was set at $1 - \beta = .80$. To detect a small interaction effect ($f = 0.1$) [31] in a 2×2 (FAMOSES vs. control group \times baseline vs. follow-up) repeated measures analysis of variance (ANOVA) based on these assumptions, the required sample size was calculated to be $n = 80$ per group (using G*Power 3.1.3) [32]. Expecting a dropout rate of 20%, we planned to include 200 parents in the study ($n = 100$ per group).

2.6. Statistical methods

Although the overall percentage of missing values for the outcome measures was low (3.0%), missing item values were imputed using the fully conditional specification method [33,34]. Five imputed datasets were created, and the parameters and standard errors of the following analyses were combined to an overall estimate using Rubin's rules [35].

Generalized estimation equations (GEE) [36,37] were used to analyze the effects of the FAMOSES parents' program. In contrast to repeated measures ANOVA, time-varying covariates can be included in

Table 2
Demographic characteristics of matched parents' groups and demographic and clinical characteristics of their children with epilepsy.

	FAMOSEs group n = 148	Control group n = 74	p-Value ^a
<i>Demographic characteristics of the parents</i>			
Respondent, n (%)			
Mother/foster mother/grandmother	122 (82.4%)	65 (87.8%)	.264
Father	13 (8.8%)	7 (9.5%)	
Mother and father	13 (8.8%)	2 (2.7%)	
Age (years), mean ± SD (range)	39.07 ± 6.64 (22–63)	38.69 ± 7.14 (21–62)	.697
Marital status, n (%)			
Living with spouse/partner	123 (83.1%)	63 (85.1%)	.874
Single parent	25 (16.9%)	11 (14.9%)	
Education, n (%)			
Without school leaving certificate	1 (0.7%)	2 (2.7%)	.337
Secondary general school (9 or 10 years)	26 (17.6%)	9 (12.2%)	
Intermediate school leaving certificate (10 years)	65 (43.9%)	38 (51.4%)	
Higher education entrance qualification	56 (37.8%)	25 (33.8%)	
Employment, n (%)			
Employed	100 (67.6%)	47 (63.5%)	.488
Unemployed	3 (2.0%)	2 (2.7%)	
Housewife/man	34 (23.0%)	15 (20.3%)	
No information/other ^b	11 (7.4%)	10 (13.5%)	
<i>Demographic characteristics of parents' children with epilepsy</i>			
Gender, n (% female)	70 (47.3%)	33 (44.6%)	.776
Age (years), mean ± SD (range)	8.73 ± 4.19 (0.42–20.92)	8.72 ± 4.54 (0.75–17.75)	.991
Educational institution, n (%)			
Kindergarten	30 (20.3%)	22 (29.7%)	.287
School	97 (65.5%)	42 (56.8%)	
Other	8 (5.4%)	6 (8.1%)	
None	13 (8.8%)	4 (5.4%)	
<i>Clinical characteristics of parents' children with epilepsy</i>			
Age at onset of epilepsy (years), mean ± SD (range)	4.60 ± 3.81 (0.00–15.67)	4.19 ± 3.54 (0.00–13.75)	.442
Treatment with anticonvulsant medication, n (% yes)	145 (98.0%)	72 (97.3%)	>.999
Identity card for severely disabled persons, n (%)	65 (43.9%)	38 (51.4%)	.320
Comorbidity, n (% yes)	64 (43.2%)	38 (51.4%)	.318
<i>Baseline seizure frequency (in the past six months), n (%)</i>			
	n = 146	n = 74	
No seizures	29 (19.9%)	12 (16.2%)	.209
1–2 seizures	23 (15.8%)	14 (18.9%)	
3–5 seizures	37 (25.3%)	9 (12.2%)	
1–2 seizures per month	13 (8.9%)	9 (12.2%)	
≥ 1 seizure per week	13 (8.9%)	9 (12.2%)	
≥ 1 seizure per day	31 (21.2%)	21 (28.4%)	

^a t-Test for independent groups (metric variables) or Fisher's exact test (categorical variables).

^b Other: self-employed, vocational training, retired, student, parental leave, marginal employment.

GEE analyses. This was necessary because the children's seizure frequency was not only correlated with several outcome measures but also changed over time. We modeled the main effects of group (FAMOSEs vs. control group) and time (baseline vs. follow-up) and their interaction. The GEE parameters were tested for significance using Wald tests, and estimated marginal means were computed to obtain statistics adjusted for seizure frequency. Effect sizes for the interaction effects were calculated as d_{ppc2} which was proposed and tested by Morris [38] for pretest–posttest–control group designs.

Except for the primary outcome parameters (adjusted $\alpha' = .017$), the criterion for statistical significance was set at $\alpha = .05$. For all analyses, two-sided p -values are reported. For statistical and psychometric analyses, we used IBM SPSS Statistics for Windows (version 23.0).

3. Results

3.1. Participant flow and sample characteristics

During the planned 14-month recruitment period (October 2011–November 2012), 194 participants from 27 FAMOSEs courses were included in the FAMOSEs group (Fig. 1). Since parents eligible for the control group were consecutively asked for study participation during their regular visits in the cooperating epilepsy centers, the recruitment

of the planned sample size for the control group had to be extended (21 months, December 2011–August 2013), resulting in a baseline sample size of 104 control group parents.

Most of the parents included at baseline returned a study questionnaire at follow-up after six months and were principally eligible for analysis (FAMOSEs group: 158/194, control group: 86/104; Fig. 1). The two groups were comparable with respect to demographic variables of parents and their children (data not shown). However, children of control group participants had a significantly higher seizure frequency than those of the FAMOSEs group ($p = .025$, Fisher's exact test), thus stressing the importance of matching in order to achieve comparability of the groups.

Since the FAMOSEs group was considerably larger than the control group, the groups were matched in a 2:1 ratio. Primary matching variables were parents' ratings of their child's impairment due to epilepsy (very strong/strong, moderate, slight/none) and parental education (Table 2). Both were selected because of their correlations with the outcome criteria. Additionally, the child's seizure frequency and age were taken into account.

The matching procedure resulted in 74 triples of comparable parents so that the matched sample for the analysis of the effects of the FAMOSEs parents' program consisted of 148 parents in the FAMOSEs group and 74 parents in the control group (Fig. 1). After matching, the

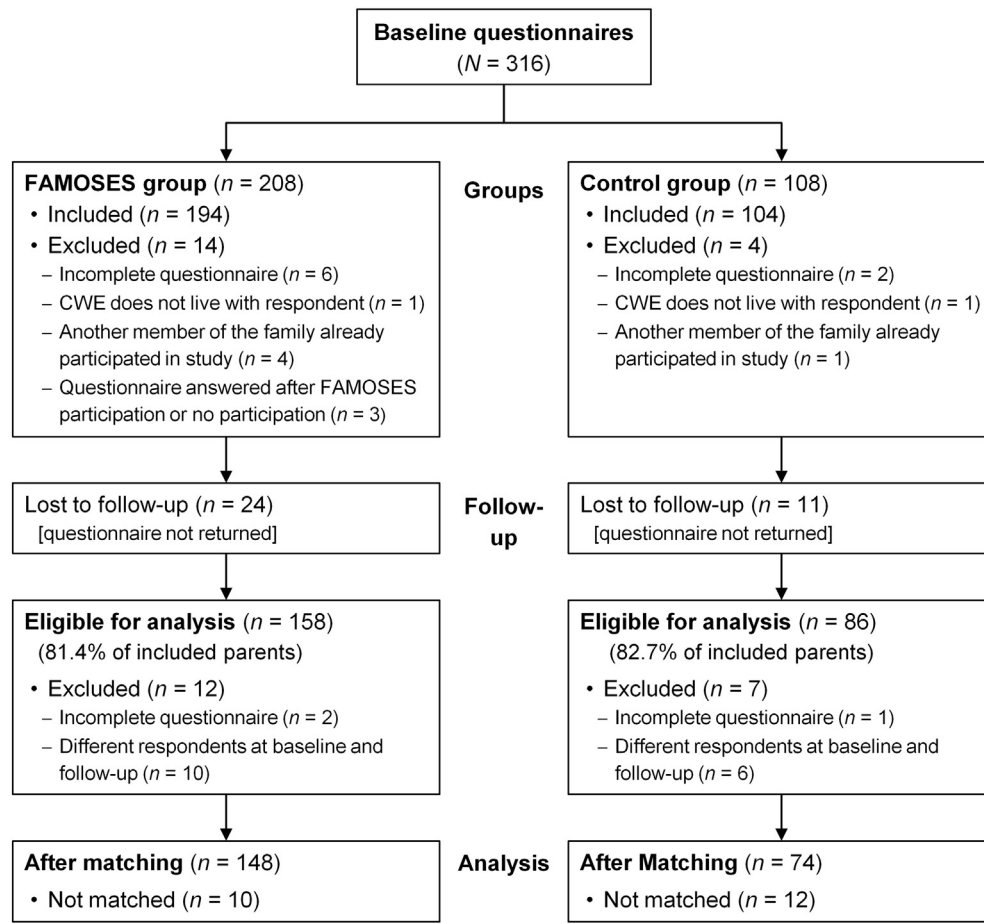


Fig. 1. Participant flow. Abbreviation: CWE = child with epilepsy.

groups did not differ significantly with regard to baseline demographic or clinical characteristics of the participants or their children with epilepsy, which are summarized in Table 2.

3.2. Effects of the FAMOSES parents' program on primary outcome criteria

3.2.1. Epilepsy-specific knowledge

Generalized estimation equation analysis revealed a significant group \times time interaction for parents' knowledge on epilepsy ($p < .001$) with a large effect size of $d_{ppc2} = .795$ (Table 3). This means that, six months after participation in the educational program, the increase in knowledge in the FAMOSES group was significantly higher compared with the control group in which the level of knowledge did not change significantly in the study period ($p = .819$; Fig. 2A). Baseline knowledge did not differ between the groups (main effect group: $p = .500$).

3.2.2. Coping with epilepsy

Since parents' coping with their child's epilepsy was correlated with the child's seizure frequency, this variable was included in the GEE analysis as a time-varying covariate. Results indicated a significant group \times time interaction ($p = .003$) with a small-to-medium effect size ($d_{ppc2} = .389$; Table 3). The improvement in coping was significantly higher in the FAMOSES group compared with the control group in which the level of coping even decreased slightly (although only marginally significant, $p = .086$; Table 3; Fig. 2B). Baseline level of coping did not differ between the groups (main effect group: $p = .155$).

3.2.3. Encouragement of the child's autonomy

Generalized estimation equation analysis (adjusted for seizure frequency) did not show an effect of FAMOSES on parents' encouragement of the child's autonomy (group \times time interaction: $p = .523$). Baseline levels of the groups were comparable, and the scores did not change significantly over time (main effects: $p = .997$ for group, $p = .767$ for time; Table 3; Fig. 2C).

3.3. Effects of the FAMOSES parents' program on secondary outcome criteria

3.3.1. Epilepsy-related fears

Six months after participation in the FAMOSES program, parents of the FAMOSES group reported less fears about short-term consequences of their child's epilepsy whereas the control group's level of fears remained stable (GEE, group \times time interaction: $p = .030$, $d_{ppc2} = .239$; Table 3). The effect of FAMOSES on parents' fears about the future development of the child and its epilepsy was slightly more pronounced with an effect size of $d_{ppc2} = .313$ (GEE adjusted for seizure frequency, group \times time interaction: $p = .003$; Table 3). Again, a decrease in the level of fears was observed after participation in the FAMOSES course, while the control group's fears remained unchanged (Fig. 2D).

3.3.2. Speaking about epilepsy with the affected child

Generalized estimation equation analysis (adjusted for seizure frequency) revealed a significant group \times time interaction ($p = .033$) with an effect size of $d_{ppc2} = .236$, indicating that the increase in speaking about epilepsy with the affected child was significantly higher compared with the control group. Neither the change in the control group nor the baseline difference between the groups was significant ($p = .524$ and $p = .173$, resp.).

Table 3
Outcome scores (estimated marginal mean \pm standard error [SE]) at baseline and at follow-up after six months.

	Control group <i>n</i> = 74		FAMOSEs group <i>n</i> = 148		GEE analysis ^a <i>B</i> (SE), <i>p</i> -value			Effect size (interaction)
	Baseline	Follow-up	Baseline	Follow-up	Group ^b	Time ^c	Group \times time ^d	<i>d</i> _{ppc2} ^e
<i>Primary outcome measures</i>								
Epilepsy-specific knowledge	48.36 \pm 2.42	47.99 \pm 2.28	46.40 \pm 1.61	61.92 \pm 1.37	−1.97 (2.92) <i>p</i> = .500	−0.38 (1.64) <i>p</i> = .819	15.90 (2.20) <i>p</i> < .001	0.795
Coping with epilepsy ^f	67.45 \pm 2.16	63.59 \pm 2.04	63.68 \pm 1.53	67.06 \pm 1.44	−3.77 (2.65) <i>p</i> = .155	−3.86 (2.24) <i>p</i> = .086	7.23 (2.45) <i>p</i> = .003	0.389
Encouragement of the child's autonomy ^f	58.42 \pm 2.44	57.84 \pm 2.66	58.43 \pm 1.76	59.26 \pm 1.85	0.01 (3.05) <i>p</i> = .997	−0.58 (1.94) <i>p</i> = .767	1.41 (2.20) <i>p</i> = .523	0.066
<i>Secondary outcome measures</i>								
<i>Epilepsy-related fears</i>								
Fears about short-term consequences	41.52 \pm 3.10	40.81 \pm 2.77	40.97 \pm 1.90	34.43 \pm 1.75	−0.55 (3.64) <i>p</i> = .879	−0.71 (2.19) <i>p</i> = .746	−5.83 (2.68) <i>p</i> = .030	0.239
Fears about the future development ^f	57.14 \pm 2.82	59.60 \pm 2.53	55.54 \pm 1.92	50.62 \pm 1.95	−1.60 (3.41) <i>p</i> = .640	2.46 (1.89) <i>p</i> = .193	−7.39 (2.34) <i>p</i> = .002	0.313
Information seeking and sharing of information	60.17 \pm 2.25	55.14 \pm 2.20	60.47 \pm 1.51	58.57 \pm 1.35	0.30 (2.70) <i>p</i> = .912	−5.03 (1.87) <i>p</i> = .007	3.13 (2.31) <i>p</i> = .176	0.167
Seizure management ^f	73.37 \pm 2.73	76.28 \pm 2.54	68.78 \pm 2.30	72.39 \pm 2.02	−4.58 (3.47) <i>p</i> = .186	2.91 (2.73) <i>p</i> = .287	0.69 (3.17) <i>p</i> = .827	0.026
Speaking about epilepsy with the affected child	57.24 \pm 3.80	53.00 \pm 3.97	54.28 \pm 2.82	57.99 \pm 2.51	−2.96 (4.64) <i>p</i> = .524	−4.23 (3.09) <i>p</i> = .173	7.94 (3.71) <i>p</i> = .033	0.236
IOFS 11-item short form ^f	2.34 \pm 0.09	2.29 \pm 0.09	2.30 \pm 0.06	2.35 \pm 0.06	−0.04 (0.11) <i>p</i> = .694	−0.05 (0.05) <i>p</i> = .371	0.10 (0.07) <i>p</i> = .126	−0.136

An increase in scores reflects a positive development for all scales except epilepsy-related fears and IOFS where higher scores represent more fears or more (negative) impact, respectively.

^a For the generalized estimation equations (GEE), parameters *B* and the corresponding standard errors (SE) are given together with *p*-values of Wald tests. (*p* < .05 in boldface)

^b The parameter "group" is the difference of estimated marginal means between the groups at baseline (e.g., for knowledge: 46.396 − 48.363 = −1.967).

^c The parameter "time" is the change in estimated marginal mean from baseline to follow-up in the control group (e.g., for knowledge: 47.988 − 48.363 = −0.375).

^d The parameter "group \times time" is the group difference in mean change over time (e.g., for knowledge: [61.922 − 46.396] − [47.988 − 48.363] = 15.901).

^e *d*_{ppc2} [38] standardizes the group difference in mean change using the pooled standard deviation at baseline.

^f Estimated marginal means adjusted for the child's seizure frequency (included in GEE analysis as time-varying covariate). For all other scales, no correlations with seizure frequency were found, or seizure frequency was not a significant covariate in GEE analyses.

For the other scales, analyses did not show significant effects of the FAMOSEs parents' program (group \times time interactions: all *p* > .10; Table 3). The amount of information seeking and sharing of information decreased in both groups, indicated by a significant main effect of time in the GEE analysis (*p* = .007; Table 3).

3.4. Effects of the FAMOSEs parents' program on disease-related variables

Both in the FAMOSEs and in the control group, children's seizure frequency significantly decreased from baseline to follow-up (main effect time: *p* < .001, GEE; Table S1), whereas the group \times time interaction was not significant (*p* = .756, GEE).

In line with this, parents reported an increase in the efficacy of their children's anticonvulsant drug therapy and a reduction of absenteeism at school or kindergarten due to seizures, which was irrespective of their group (main effect time: both *p* < .05, group \times time interaction: both *p* > .05, GEE; Table S2). Analyses of consultation rates and tolerability of anticonvulsant drug therapy did not show significant effects of group or time (*p* > .05 for main effects and interaction, GEE; Table S2).

3.5. Subgroup analyses and analysis of center effects

3.5.1. Children with severe chronic epilepsy

According to the guidelines of the German health insurances, not all participants' children could be classified as suffering from a "severe chronic condition"¹ [39]. For this reason, all analyses were repeated using only those parents whose children met the guideline criteria,

¹ These guidelines ("Chroniker-Richtlinie") specify criteria for the definition of severe chronic conditions according to the German Social Code, Book Five (SGB V, §62). A medical condition is chronic if it is treated by a physician for at least one year (at least once in every quarter of a year) and a) the patient has a need for long-term care (care level 2 or 3, German Social Code XI, Chapter 2) and/or b) the patient is severely disabled (degree of disability of at least 60 according to the German law) and/or c) continuous medical treatment (e.g. drug therapy) is necessary.

based on the information available from the study questionnaires (FAMOSEs group: *n* = 109, control group: *n* = 63). The results were comparable with those of the complete sample in terms of significant effects and effect sizes (Table S3).

3.5.2. Exclusion of parents of children without seizures in the last year

In contrast to the initially formulated inclusion criteria, parents of children without seizures in the last year participated in the study and were not excluded from the main analyses. In the sense of a per protocol analysis, the analyses for all outcome measures were repeated only for parents of children with active epilepsy (as defined in the inclusion criteria; FAMOSEs group: *n* = 131, control group: *n* = 67). Again, the results did not differ considerably compared with those of the main analyses (Table S4).

3.5.3. Center effects

A comparison of the effects of the FAMOSEs parents' program between the cooperating epilepsy centers did not indicate differences in efficacy (data not shown).

3.6. Satisfaction with the FAMOSEs parents' program

For the evaluation of the educational program by the participating parents, all questionnaires returned at follow-up were included (*n* = 169). Six months after participation, the parents' answers showed a high degree of satisfaction with FAMOSEs (Table 4).

4. Discussion

4.1. Methodological aspects

Our aim was to evaluate the efficacy of the FAMOSEs parents' program in the context of its regular implementation in various epilepsy centers in a methodologically sound study. Therefore, we used the

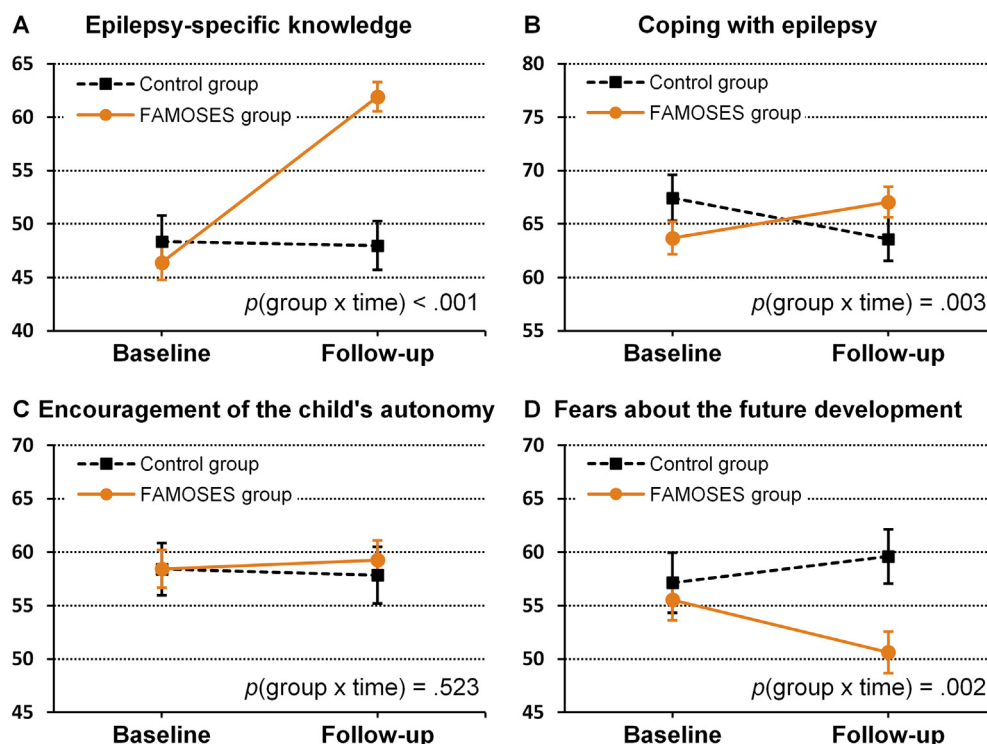


Fig. 2. Effects of FAMOSES parents' program on the three primary outcome measures (A–C, increase in scores = positive development) and on the parents' fears about the future development of the child and its epilepsy (D, decrease in scores = positive development). Data are expressed as estimated marginal means \pm standard error (adjusted for seizure frequency as covariate for all scales except epilepsy-specific knowledge). *p*-Values for the group \times time interaction are taken from the GEE analyses.

experience from a pilot study [23,24] and improved its design in several ways to ensure validity of the results:

We increased the number of study participants. A priori sample size calculations were executed, and the required number of parents was reached for the control group and exceeded for the FAMOSES group. The larger number of FAMOSES participants in the study is due to the fact that the number of FAMOSES courses within the recruitment period was larger than expected based on prior experience [23]. Both the dropout rate and the number of excluded questionnaires were low and within the expected range of 20%.

Moreover, the length of the follow-up period was extended from three to six months to give FAMOSES participants the opportunity to apply what they had learned during the course in everyday life. Outcome measures were developed based on the item pool of the pilot study, leading to internally consistent and reliable scales without bottom or ceiling effects.

In addition to that, we used a matching procedure. Although this led to the exclusion of some participants who could not be matched, it ensured structural equality of the two matched groups with respect to baseline variables. To further improve internal validity, data were

analyzed using the GEE approach allowing us to account for changes in the children's seizure frequency.

4.2. Efficacy of the FAMOSES parents' program

Imparting knowledge is one of the primary aims of psycho-educational programs [21,40], taking into account the need for information that many parents of CWE have [10,19]. In line with previous studies evaluating psychoeducational programs for families of CWE or adult patients with epilepsy [21,40,41], the FAMOSES parents' program successfully improved participants' knowledge about epilepsy.

Aside from providing information, FAMOSES trainers encourage participants to develop strategies to cope with their child's disorder in everyday life [23], and the interactive course setting gives various opportunities to express and discuss attitudes and opinions as well as fears. In accordance with these aims, our study has shown that the FAMOSES parents' program was successful in improving parental coping and in reducing epilepsy-related fears. The FAMOSES parents' program further emphasizes the importance of understanding the child's situation so that parents can help their child to cope with its

Table 4
Parents' satisfaction with the FAMOSES program.

	Completely	Rather yes	Partially	Rather not	Not at all
Expectations have been fulfilled	64.5%	29.0%	5.3%	0.0%	0.0%
Recommendation to other parents	86.9%	11.9%	1.2%	0.0%	0.0%
	Very much	Much	Somewhat	Little	Not at all
New skills, new knowledge acquired	38.7%	39.3%	18.5%	3.6%	0.0%
FAMOSES helped in everyday management	26.2%	38.7%	29.2%	4.8%	1.2%
Feeling safer with seizure management	23.7%	39.1%	28.4%	6.5%	2.4%
	Very good	Good	Fair	Poor	Very poor
Evaluation of FAMOSES material	66.1%	30.4%	3.6%	0.0%	0.0%
Rating of FAMOSES program as a whole	71.4%	27.4%	1.2%	0.0%	0.0%

disorder. In this context, the increase in speaking about epilepsy with the affected child is an important finding.

Parents' encouragement of the child's autonomy did not change after participation in the FAMOSES parents' program. This is possibly due to the fact that many participants' children were relatively young and/or disabled. Questions about their autonomy may not be appropriate because of their ongoing dependence on their parents.

Participation in the FAMOSES course did not improve parents' seizure management, but the scores on this scale were already relatively high at baseline, indicating mainly adequate reactions of parents to their children's seizures. Additionally, it has to be kept in mind that the appropriate reaction depends on the seizure type.

We did not find effects of the FAMOSES parents' program on disease-related variables. In line with the results of the pilot study [23] and of FLIP&FLAP [42], the only other evaluated psychoeducational program for families of CWE in Germany, the change in children's seizure frequency did not differ between treatment and control groups. However, seizure frequency significantly decreased in both groups, demonstrating the efficacy of the medical treatment that all participants' children received in the cooperating epilepsy centers.

4.3. Satisfaction with the FAMOSES parents' program

Since the educational program FAMOSES was implemented, parents' satisfaction with the course was and still is assessed shortly after participation. An analysis of these data revealed constantly high or increasing ratings over the years [23]. Our study contributes to these encouraging findings by showing that parents' ratings remain high even six months later.

4.4. Limitations and strengths

Our evaluation of the FAMOSES parents' program is the first study of a psychoeducational program for families of CWE replicating the results of a previous study [23,24]. This was possible because the FAMOSES program has been regularly implemented in a number of German, Austrian, and Swiss epilepsy centers for more than 10 years. The encouragingly high number of study participants, especially in the FAMOSES group, is due to this wide use of the program and increases the generalizability of our results. "Train-the-trainer" courses and the detailed trainer's manual further contribute to the external validity.

The lack of random assignment of participants to study groups is a weakness of our study. This possible threat to validity is common in studies evaluating psychosocial interventions for patients with epilepsy or families of CWE [21,40,41]. It may lead to differences between the groups so that effects are attributable to these differences rather than to program participation. However, results from randomized studies can be approached in quasi-experimental designs, given the correct choice and control of covariates [43]. In our study, we did so by matching FAMOSES and control group on important demographic and disease-related variables. This and the inclusion of children's seizure frequency in the analyses as a time-varying variable increased the internal validity of our results. Furthermore, additional analyses confirmed the effects for specific subgroups.

The instruments used in this study were mainly specifically developed to cover the aims of the FAMOSES parents' program. To increase comparability between studies, the use of standardized outcome measures is generally recommended [40], yet appropriate questionnaires are not available for most outcome epilepsy-specific criteria. However, we were able to develop reliable instruments based on the previous study. These instruments may be used in future studies, for instance in evaluations of other educational programs or psychosocial care.

The current study is limited to the evaluation of the FAMOSES parents' program. Although the FAMOSES parents' and children's programs have been developed at the same time and it is assumed

that the parallel participation of parents and children is advantageous [23], the two programs are independent. Since parents can take part in the parents' course even if their child is too young or too impaired for the children's program, the target groups of both programs only partly overlap. Future research should therefore aim at evaluating the efficacy of the FAMOSES children's program from both children's and parents' perspective and the combination of children's and parents' program.

5. Conclusion

This controlled multicenter study confirmed the efficacy of the FAMOSES program for parents of CWE. The successful improvement of knowledge about epilepsy and the interactive approach of the program help parents to cope with their child's disorder and reduce epilepsy-related fears. Together with the wide and regular implementation in many epilepsy centers and participants' high satisfaction with the course, this makes the FAMOSES parents' program a valuable component of comprehensive epilepsy care.

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Disclosures

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Appendix A. Supplementary data

Supplementary data to this article can be found online at <http://dx.doi.org/10.1016/j.yebeh.2016.09.027>.

References

- [1] Ellis N, Upton D, Thompson P. Epilepsy and the family: a review of current literature. *Seizure* 2000;9:22–30.
- [2] Austin JK, Dunn DW, Johnson CS, Perkins SM. Behavioral issues involving children and adolescents with epilepsy and the impact of their families: recent research data. *Epilepsy Behav* 2004;5:33–41.
- [3] Dunn DW. Neuropsychiatric aspects of epilepsy in children. *Epilepsy Behav* 2003;4:101–6.

- [4] Freilinger M, Reisel B, Reiter E, Zelenko M, Hauser E, Seidl R. Behavioral and emotional problems in children with epilepsy. *J Child Neurol* 2006;21:939–45.
- [5] Cushner-Weinstein S, Dassoulas K, Salpekar JA, Henderson SE, Pearl PL, Gaillard WD, et al. Parenting stress and childhood epilepsy: the impact of depression, learning, and seizure-related factors. *Epilepsy Behav* 2008;13:109–14.
- [6] Hoare P. The quality of life of children with chronic epilepsy and their families. *Seizure* 1993;2:269–75.
- [7] Taylor J, Jacoby A, Baker GA, Marson AG. Self-reported and parent-reported quality of life of children and adolescents with new-onset epilepsy. *Epilepsia* 2011;52:1489–98.
- [8] McEwan MJ, Espie CA, Metcalfe J, Brodie MJ, Wilson MT. Quality of life and psychosocial development in adolescents with epilepsy: a qualitative investigation using focus group methods. *Seizure* 2004;13(1):15–31.
- [9] Rodenburg R, Meijer AM, Scherphof C, Carpay JA, Augustijn P, Aldenkamp AP, et al. Parenting and restrictions in childhood epilepsy. *Epilepsy Behav* 2013;27(3):497–503.
- [10] Shore CP, Buelow JM, Austin JK, Johnson CS. Continuing psychosocial care needs in children with new-onset epilepsy and their parents. *J Neurosci Nurs* 2009;41:244–50.
- [11] Hirfanoglu T, Serdaroglu A, Cansu A, Soysal AS, Derle E, Gucuyener K. Do knowledge of, perception of, and attitudes toward epilepsy affect the quality of life of Turkish children with epilepsy and their parents? *Epilepsy Behav* 2009;14:71–7.
- [12] Asiri N, Bin Joubah M, Khan S, Jan M. Maternal knowledge of acute seizures. *Neurosciences* 2015;20:346–9.
- [13] Camfield C, Camfield P. Most adults with childhood-onset epilepsy and their parents have incorrect knowledge of the cause 20–30 years later: a population-based study. *Epilepsy Behav* 2014;37:100–3.
- [14] Gazibara T, Nikolovski J, Ladic A, Pekmezovic T, Kusic-Tepavcovic D. Parental knowledge, attitudes, and behaviors towards children with epilepsy in Belgrade (Serbia). *Epilepsy Behav* 2014;41:210–6.
- [15] Hall-Parkinson D, Tapper J, Melbourne-Chambers R. Parent and caregiver knowledge, beliefs, and responses to convulsive seizures in children in Kingston, Jamaica – a hospital-based survey. *Epilepsy Behav* 2015;51:306–11.
- [16] Tieffenberg JA, Wood EI, Alonso A, Tossutti MS, Vicente MF. A randomized field trial of acindes: a child-centered training model for children with chronic illnesses (asthma and epilepsy). *J Urban Health* 2000;77:280–97.
- [17] O'Toole S, Lambert V, Gallagher P, Shahwan A, Austin JK. Talking about epilepsy: challenges parents face when communicating with their child about epilepsy and epilepsy-related issues. *Epilepsy Behav* 2016;57:9–15.
- [18] Ali DB, Tomek M, Lisk DR. The effects of epilepsy on child education in Sierra Leone. *Epilepsy Behav* 2014;37:236–40.
- [19] Laybourn AN, Hill M. Children with epilepsy and their families: needs and services. *Child Care Health Dev* 1994;20:1–14.
- [20] Ronen G, Streiner D, Rosenbaum P. Health-related quality of life in childhood epilepsy: moving beyond 'seizure control with minimal adverse effects'. *Health Qual Life Outcomes* 2003;1:36.
- [21] May TW, Pfäfflin M. Psychoeducational programs for patients with epilepsy. *Dis Manag Health Outcomes* 2005;13:185–99.
- [22] Wohlrab GC, Rinnert S, Bettendorf U, Fischbach H, Heinen G, Klein P, et al. Famoses: a modular educational program for children with epilepsy and their parents. *Epilepsy Behav* 2007;10:44–8.
- [23] Pfäfflin M, Petermann F, Rau J, May TW. The psychoeducational program for children with epilepsy and their parents (FAMOSEs): results of a controlled pilot study and a survey of parent satisfaction over a five-year period. *Epilepsy Behav* 2012;25:11–6.
- [24] Rau J, May TW, Pfäfflin M, Heubrock D, Petermann F. Schulung von Kindern mit epilepsie und deren Eltern mit dem modularen Schulungsprogramm epilepsie für Familien (FAMOSEs) – ergebnisse einer evaluationsstudie. *Rehabilitation* 2006;45:27–39.
- [25] Boutron I, Moher D, Altman DG, Schulz KF, Ravaud P. Extending the CONSORT statement to randomized trials of nonpharmacologic treatment: explanation and elaboration. *Ann Intern Med* 2008;148:295–309.
- [26] Boutron I, Moher D, Altman DG, Schulz KF, Ravaud P. Methods and processes of the CONSORT group: example of an extension for trials assessing nonpharmacologic treatments. *Ann Intern Med* 2008;148:W60–6.
- [27] Ravens-Sieberer U, Morfeld M, Stein REK, Jessop DJ, Bullinger M, Thyen U. Der familien-belastungs-fragebogen (FaBel-fragebogen). *Psychother Psychosom Med Psychol* 2001;51:384–93.
- [28] Dehn LB, Korn-Merker E, Pfäfflin M, Fischbach H, Frantz M, Hauser A, et al. Erfassung der belastungen von Eltern anfallskranker Kinder. *Z Epileptol* 2013;26:214–22.
- [29] Dehn LB, Korn-Merker E, Pfäfflin M, Ravens-Sieberer U, May TW. The Impact on Family Scale: psychometric analysis of long and short forms in parents of children with epilepsy. *Epilepsy Behav* 2014;32:21–6.
- [30] Williams AR, Piamjariyakul U, Williams PD, Bruggeman SK, Cabanela RL. Validity of the revised impact on family (IOF) scale. *J Pediatr* 2006;149:257–61.
- [31] Cohen J. *Statistical power analysis for the behavioral sciences*. 2nd ed. Hillsdale, New Jersey: Lawrence Erlbaum Associates; 1988.
- [32] Faul F, Erdfelder E, Lang A, Buchner A. G*Power 3: a flexible statistical power analysis program for the social, behavioral, and biomedical sciences. *Behav Res Methods* 2007;39:175–91.
- [33] Raghunathan TE, Lepkowski JM, van Hoewyk J, Solenberger P. A multivariate technique for multiply imputing missing values using a sequence of regression models. *Surv Methodol* 2001;27:85–95.
- [34] van Buuren S. Multiple imputation of discrete and continuous data by fully conditional specification. *Stat Methods Med Res* 2007;16:219–42.
- [35] Rubin DB. *Multiple imputation for nonresponse in surveys*. Hoboken, NJ: Wiley-Interscience; 2004.
- [36] Liang K, Zeger SL. Longitudinal data analysis using generalized linear models. *Biometrika* 1986;73:13–22.
- [37] Zeger SL, Liang K. Longitudinal data analysis for discrete and continuous outcomes. *Biometrics* 1986;42:121–30.
- [38] Morris SB. Estimating effect sizes from pretest–posttest–control group designs. *Organ Res Methods* 2008;11:364–86.
- [39] Bundesausschuss G. Richtlinie des Gemeinsamen Bundesausschusses zur umsetzung der regelungen in § 62 für schwerwiegend chronisch erkrankte ("Chroniker-Richtlinie"). In der Fassung vom 22. Januar 2004, veröffentlicht im Bundesanzeiger; Nr. 18: S. 1 343; zuletzt geändert am 19. Juni 2008, veröffentlicht im Bundesanzeiger; Nr. 124: S. 3 017, in Kraft getreten am 20. August 2008. 2008 Available from: https://www.g-ba.de/downloads/62-492-278/Chr-RL_2008-06-19.pdf.
- [40] Mittan RJ. Psychosocial treatment programs in epilepsy: a review. *Epilepsy Behav* 2009;16:371–80.
- [41] Corrigan FM, Broome H, Dorris L. A systematic review of psychosocial interventions for children and young people with epilepsy. *Epilepsy Behav* 2016;56:99–112.
- [42] Jantzen S, Müller-Godeffroy E, Hallfahrt-Krisl T, Aksu F, Püst B, Kohl B, et al. FLIP&FLAP—a training programme for children and adolescents with epilepsy, and their parents. *Seizure* 2009;18:478–86.
- [43] Cook TD, Steiner PM. Case matching and the reduction of selection bias in quasi-experiments: the relative importance of pretest measures of outcome, of unreliable measurement, and of mode of data analysis. *Psychol Methods* 2010;15(1):56–68.