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Epilepsy Journey: A proof of concept trial of a web-based executive functioning intervention for adolescents with epilepsy

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Summary

Objectives: To examine the preliminary feasibility and acceptability of a web-based program, Epilepsy Journey, to improve executive function behaviors in adolescents with epilepsy.

Methods: We conducted a proof of concept single-arm pilot trial of Epilepsy Journey with 31 adolescents (average age 15.3 ± 1.3 years) who had an epilepsy diagnosis and executive function (EF) deficits on the caregiver-report version of the Behavior Rating Inventory of Executive Function (BRIEF). Epilepsy Journey coupled a gamified problem-solving website comprised of 10 learning modules targeting EF deficits (e.g., working memory, organization, problem-solving) with Skype sessions with a trained therapist. Outcomes included feasibility (attrition, sessions completed) and acceptability (satisfaction ratings). Exploratory analyses examined changes in caregiver, self- and teacher-reported BRIEF scores from baseline to post treatment and at 2- and 5-month follow-ups.

Results: Seventy-nine percent of participants completed the program. Satisfaction was high with 97% of caregivers and adolescents rating the program as helpful and indicating they would recommend it to others. Caregivers and adolescents reported global improvements on the BRIEF, with caregivers reporting significant improvements on all BRIEF subscales. EF symptoms rebounded slightly between the 2 and 5-month follow-ups for some of the self- and caregiver-reported BRIEF scales. Notably, clinically meaningful improvements (e.g. clinical/subclinical to normative levels) were reported for several caregiver-reported BRIEF subscales, including the Global Executive Composite (62% to 33–34%) and Metacognitive Index (74% to 41–42%) from baseline to 2 and 5-month follow-up.

Significance: Findings suggest that a web-based problem-solving intervention tailored to EF deficits for adolescents with epilepsy is both feasible and acceptable and may contribute to improvements in EF behaviors across domains.

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Disclosure of Conflicts of Interest

None of the authors has any conflict of interest to disclose.

Ethical Publication Statement

We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this report is consistent with those guidelines.

Keywords

working memory; clinical trial; behavioral regulation; metacognition; executive dysfunction

Introduction

Executive functioning (EF) comprises a constellation of skills required for complex, goal directed activities. Researchers have divided EF into hot and cold EF skills. Hot skills are affectively-driven abilities such as behavior regulation, emotion regulation, affective decision making, social skills, and theory of mind.^{1; 2} Cold EF skills include metacognition, cognitive flexibility, attention, inhibition, working memory planning, and problem-solving.³ Success in social and academic settings requires an interplay of both hot and cold EF abilities in the context of social problem solving, with deficits in the EF domains of behavior and emotion regulation contributing to depression in adulthood. Importantly, EF deficits have been documented in almost 50% of youth with epilepsy,⁴ which is 3 times the prevalence in healthy youth.^{5; 6} EF deficits predict academic underachievement, poor school performance,^{5; 7} neurobehavioral comorbidities (e.g., internalizing and externalizing problems),^{8; 9} and poor HRQOL^{9; 10} in pediatric epilepsy. Evidence-based interventions to improve EF could play a critical mediating role in preventing adverse outcomes (e.g., psychological comorbidities) and promoting optimal functioning in adolescents with epilepsy; however few interventions exist for this vulnerable population.

Although the effects of executive dysfunction are far-reaching, researchers have only tested existing tools with specific training around one EF skill (e.g., working memory and CogMed¹¹). While this intervention can certainly be beneficial to address working memory deficits in youth with epilepsy^{10; 11} that can generalize to other aspects of EF,^{12; 13} a more comprehensive approach that specifically targets multiple EF domains and generalizes to everyday functioning may provide a more comprehensive solution for EF deficits. More specifically, a comprehensive EF intervention must target the hot EF domains of behavior and emotion regulation to support successful application of the cold EF metacognitive skills in complex, everyday settings. Using the ORBIT model of behavioral intervention development,¹⁴ we conducted Phase 1a (Define the problem: Focus Groups¹⁵) and 1b (Refine: Formative User Centered Evaluation including usability testing and heuristic evaluation¹⁶) design elements. These two steps yielded an engaging web-based intervention, called Epilepsy Journey, in which participants took a journey to 10 different EF-themed lands (learning modules), providing training in both metacognitive (cool EF) and behavior regulation (hot EF) skills, over a 12-week timeframe. Participants learned about various EF skills with subsequent videoconferencing therapy sessions to review materials and problem-solve EF deficits specific to the individual.

The goal of the current study was to examine the preliminary feasibility and acceptability of Epilepsy Journey in a proof-of-concept (ORBIT Phase 2a) trial with adolescents with epilepsy. The primary outcome was caregiver-reported Behavior Rating Inventory for Executive Functioning (BRIEF). The BRIEF is a robust, reliable measure of EF that correlates highly with biological markers of EF,^{17–19} has concurrent and predictive

relationships with real-world functioning (e.g., academic skills),^{20; 21} and can serve to define intervention targets and outcomes.²² It was hypothesized that the program would be feasible as defined by high rates of treatment completion and acceptable based on ratings of satisfaction by caregivers and adolescents. In exploratory analyses, we examined changes on the global scales of the caregiver-reported BRIEF (i.e., Global Executive Composite, Metacognition Index, and Behavioral Regulation Index). Statistical and clinically meaningful improvements on specific caregiver BRIEF subscales (e.g., Inhibit, Emotional Control, Working Memory, Plan/Organize, and Monitor) were also explored. Additional exploratory outcomes included self-reported and teacher-reported BRIEF global and subscale scores.

Methods

Participants

Participants were recruited from the Comprehensive Epilepsy Center in a Midwestern children's hospital from October 2016 to January 2018. Inclusion/exclusion criteria for this proof-of-concept trial included: 1) epilepsy diagnosis, 2) adolescents ages 13–17 years, 3) no comorbid non-epilepsy medical disorders requiring daily medications, with the exception of asthma/allergies and attention deficit hyperactivity disorder, 4) no comorbid significant developmental disorders (e.g., autism), 5) family residence 100 miles of the hospital, 6) English-speaking, 7) no plan to wean from epilepsy treatment in the following six months, and 8) caregiver-reported EF deficits based on the BRIEF, defined as 1 clinically elevated BRIEF subscale (T-score ≥ 65)²³ or 2 sub-clinically elevated BRIEF subscales (e.g., 1 standard deviation; T-scores $60 < 65$) based on normative data for standardized tests. We chose to use the BRIEF parent-report as a single indicator of EF because it assesses the application of EF skills and is more likely to reflect the demands of daily tasks (e.g., completing homework, remembering to do chores) compared to performance-based tests. Rating scales are also easily administered in clinical practice, which allows for rapid identification of EF deficits.⁴

Procedures

A trained research coordinator identified eligible patients and obtained written caregiver consent and adolescent assent once enrolled. Caregivers then completed the caregiver-reported BRIEF to screen for EF deficits. If adolescents demonstrated EF deficits based on inclusion/exclusion criteria, they proceeded with the study. Caregivers and adolescent participants completed a battery of questionnaires. Additionally, caregivers were asked to have the adolescent's primary teacher (e.g., math, English) complete the teacher BRIEF. The teachers were not explicitly told about the child's participation in the study; however, it is possible the caregivers or adolescents provided study information when asking the teacher to complete the form. Teachers were also provided with self-addressed stamped envelopes to send the form back to study staff directly. Measures were completed at baseline, post-treatment (e.g., after completion of the 12 weeks of intervention), 2-month and 5-month follow-up. At the post-treatment and follow-up time points, caregivers and adolescents also completed a measure of acceptability/satisfaction. Families were compensated using a graduated incentive schedule for time and effort for completing questionnaires and

intervention sessions using reloadable gift cards (i.e., adolescents up to \$260 and caregivers up to \$80). This study was approved by the hospital's Institutional Review Board.

Epilepsy Journey Intervention—Epilepsy Journey is a gamified, web-based EF intervention, in which adolescents develop problem-solving skills to address EF deficits by navigating through various “lands” (10 learning modules) focused on core components of EF (e.g., working memory, problem-solving). The modules have interactive components (e.g., games), video content, and the ability to input responses. Participants independently completed online learning modules (~20 minutes) before meeting with a therapist via videoconferencing (~30 minutes) to review skills and problem-solve solutions to EF-related challenges in their everyday lives. Videoconferencing intervention sessions (e.g., Skype for Business) were conducted by trained interventionists (psychology doctoral student/post-doctoral fellow) and audio-recorded for treatment fidelity and weekly supervision. Prior to the videoconference, the interventionist ensured the module had been completed and reviewed any response input by the adolescent in the module (e.g., examples of the adolescent's ideas for problem-solving). If the module was not completed, videoconferencing was rescheduled. The goal was to complete the 10 modules with videoconferencing within a 12-week period.

At the time of enrollment, coordinators ensured that participants had functioning devices (e.g., computer, tablet, smartphone) and Internet connections for the intervention. If the participant did not have access (n=1 out of 39), a study tablet with a data plan was provided. One participant no longer had Internet access during the study and used the school/public library computer to review the remaining modules.

Each adolescent's Epilepsy Journey session order was tailored based on the adolescent's particular EF deficits. The first two sessions were the same for each participant, as they represented the core skills necessary for having a positive attitude, problem-solving for future sessions, and navigating the Epilepsy Journey website. The remaining modules were ordered based on the adolescent's and caregiver's BRIEF subscale scores, with sessions addressing areas in greatest need of intervention (e.g., highest scores) occurring first. Non-elevated sessions or similarly elevated sessions could be ordered based on the adolescent's preference. One reason to tailor the sessions based on EF deficits and preference was to engage the adolescent and show the potential benefit of Epilepsy Journey early in the treatment process. The problem-solving process provided adolescents with an opportunity to develop cross-cutting solutions drawing upon the skills and strategies learned across EF domains.

Treatment fidelity was measured via review of audio-recorded sessions by the first and senior authors. A treatment fidelity checklist was created for each module to ensure the primary goals and content were reviewed by the interventionist.

Description of the Epilepsy Journey Intervention

Session 1 (Peninsula of Positive Thoughts): The first session took place at the adolescent's home with an adult present to demonstrate how to access the online modules for that session and future sessions, as well as setting up videoconferencing capabilities. One

goal of this session was to build initial rapport between the adolescent and interventionist (~5 minutes) as all future sessions were done via videoconference. In addition, the interventionist reviewed the structure of Epilepsy Journey, limits to confidentiality, and session guidelines (~10 minutes). The adolescent developed individual goals for the intervention (~10 minutes), with help from the interventionist to ensure goals were specific, focused in areas of EF, and modifiable. The interventionist showed caregivers the technical aspects of the intervention in case the adolescent required help in the future. The interventionists then guided adolescents through the Peninsula of Positive Thoughts module. This session provided information about the importance of staying positive, skills for reframing problems, and introduced positive self-statements (~40 minutes). Finally, the interventionist concluded the session and scheduled the next teleconference with the adolescent (~5 minutes). Overall, this study visit took approximately 60–90 minutes.

Session 2 (Problem-solving Peak).: This module focused on teaching adolescents about problem-solving skills using the ABCDE's of problem-solving (Aiming/Identifying a goal, Brainstorming, Choosing a plan, Do(ing) or implementing the plan, and Evaluating what works and what does not). Adolescents also completed quizzes. During the videoconference, the interventionist reviewed the problem-solving skills and supported the adolescent in applying the skills to one of their three identified goals/problems from Session 1.

Sessions 3–9 (Mists of Memory, Orchards of Organization, Falls of Inhibition, Island of Initiation, Land of Nod, State of Emotional Control, Mesa of Monitoring).: As noted, the order of these sessions was based on the adolescent's individual EF deficits and preferences. For each of these modules, the adolescent reviewed content and strategies to improve the specific skill. The videoconference session with the interventionist then focused on reviewing the specific skills and problem-solving around a deficit in that area (e.g., messy backpack in Orchards of Organization or remembering to do chores in Mists of Memory). The Land of Nod addressed sleep and stress-related issues. The interventionist also reviewed problem-solving from the prior session and whether those solutions worked and if the solutions did not work, the adolescent and therapist identified new solutions.

Session 10 (Wrap-up).: This last session focused on reviewing the primary skills of each module and having the adolescent problem-solve around future issues that may arise.

Measures

Background Information Form.—The Background Information Form is a demographic questionnaire completed by caregivers that provides information about the adolescent's age, socio-economic status, history of seizures, comorbid illnesses (e.g., learning disorders), and school information.

Behavior Rating Inventory of Executive Function (BRIEF²³).—The BRIEF provides caregiver-, self- and teacher-reported items to assess EF in the home and school environments. The BRIEF is 86-items with eight scales (Inhibit, Shift, Emotional Control, Initiate/Task Completion, Working Memory, Plan/Organize, Organization of Materials, and Monitor) and two validity scales. The clinical scales form two broader indexes (Behavioral

Regulation; BRI and Metacognition; MI) and an overall score, the Global Executive Composite (GEC). The Behavioral Regulation Index consists of Inhibition, Shifting, and Emotional Control and would be considered “hot” EF skills while the Metacognition Index consists of Initiation, Working Memory, Plan/Organize, Organization of Materials, and Monitoring, which would be considered “cold” EF skills. For the self-report, additional indices are also calculated: Behavioral Shift (hot) and Cognitive Shift (cold). Cronbach’s alphas ranged from 0.80–0.98 for the scales.

Pediatric Epilepsy Side Effects Questionnaire (PESQ²⁴)—The PESQ is a 19-item validated measure assessing side effects of anti-seizure medications in youth ages 2–18. Items cover neurological, behavioral, gastrointestinal, skin, and motor side-effects and are rated on a Likert scale. Items are summed and transformed into a score ranging from 0 to 100, with higher scores representing more side effects. The PESQ has strong internal consistency (0.72–0.93) and test-retest reliability (0.74–0.97).²⁴

Epilepsy Journey Satisfaction Questionnaire.—Caregivers and adolescents completed a study-specific questionnaire assessing feasibility, satisfaction and acceptability of Epilepsy Journey, which was adapted from prior questionnaires.^{25; 26} Content included items related to satisfaction of the format, content, length, convenience, as well as use of technology and perceived impact of the intervention on outcomes.

Interventionist Ratings of Sessions.—After completion of each videoconference, the interventionist rated the participant’s engagement, openness, cooperation, hostility/friendliness, and therapeutic alliance, as well as time spent conducting the session.

Electronic Medical Chart Review.—Chart reviews were conducted to collect data on epilepsy type, prescribed anti-seizure medication(s), time since epilepsy diagnosis, and seizure frequency. Seizure frequency was dichotomized into the absence (coded=0) or presence (coded=1) of seizures in the past 3 months given the heterogeneity of seizure types. Chart reviews covered the entire study period.

Statistical Analyses

Means, standard deviations, and frequencies were used to examine demographic and medical variables, as well as measures of treatment fidelity, feasibility, and acceptability. To examine whether there were statistically significant improvements on the outcome measures, longitudinal mixed models were conducted in Stata version 15²⁷ using robust maximum likelihood estimation. Each of the BRIEF subscales were modeled as an outcome with time as the predictor variable and adolescent race, years since diagnosis, seizure type, presence of seizures in the last 3 months, and total side effects as covariates. Alpha was set to 0.05 for all analyses and no correction was made for multiple comparisons due to the pilot nature of the study. Each caregiver and adolescent-reported outcome was tested for the presence of a significant non-linear trajectory (i.e., quadratic function of time) and this non-linearity was modeled if applicable. Given that the teacher-reported outcomes were only collected at two time points, no nonlinear trajectories were modeled for these outcomes. An ANOVA was

also used to examine group-based changes in the number of BRIEF subscale elevations from baseline to post, 2-month, and 5-month post intervention periods.

Clinically meaningful improvements were defined as a participant moving from subclinical or clinically elevated (T scores ≥ 60) BRIEF subscale scores to normative levels (T scores < 60). Z-tests, accounting for the repeated measurements, were used to compare proportions of subclinical/clinical elevations from baseline to post-treatment, baseline to 2-month follow-up, and baseline to 5-month follow-up.

Results

Participants

Sixty-eight eligible patients were approached for study participation. Forty-seven agreed to participate (69% recruitment). Of the 47 that agreed to participate, $n=8$ did not meet the BRIEF screening criteria, resulting in 39 total participants. Of the 39 participants, 8 withdrew (3 prior to intervention), resulting in a final sample of 31 participants (retention=79%; Figure 1 Consort Diagram). Demographic and medical data are provided in Table 1. The primary antiseizure medications prescribed included levetiracetam (56%), lamotrigine (15%), valproate (15%), topiramate (10%). Additional antiseizure medications included carbamazepine, oxcarbazepine, lacosamide, ethosuximide, felbamate, zonisamide, and clobazam.

Feasibility

Eighty-three percent of total sessions ($n=325$ of 390) were completed by participants. Of the 39 participants, $n=31$ completed all 10 sessions. The other eight patients completed from 0 to 6 sessions. Nine percent of the sessions were reviewed for treatment fidelity (first/senior authors), resulting in a review of 30 total sessions. Treatment fidelity across the two interventionists was 95.8% (343 of the 358 tasks).

Adolescents spent an average of 33 minutes in the videoconference sessions (range 15–58 minutes) and 9 minutes completing the online modules (range 6–15 minutes), with the exception of the therapist-guided initial home visit focused on Peninsula of Positive Thoughts. Participants took an average of 13 weeks to complete all 10 sessions (67% within 12 weeks and 33% > 12 weeks). The therapists rated the adolescent as having a positive emotional response to sessions between 74% and 97% of the time, depending on the session. Therapists noted the highest positive response to the sessions focused on problem solving (94.3%) and memory (93.7%). Engagement in homework after each session was also very high, ranging from 87% for the final session to 100% for the sleep/stress session.

The most frequent technical issue encountered by adolescents was difficulty using a videoconferencing platform; the solution typically included another method of contact (e.g., Facetime or phone call). The only other technical issue encountered by participants included a hospital-level migration to a different online platform that briefly affected participant access to the website. This issue was identified and resolved within one week.

Acceptability/Satisfaction

An overwhelming majority of caregivers and adolescents found the program useful (caregivers=97%, adolescents=97%), and endorsed that they would recommend the program to others (caregivers=97%, adolescents=97%). Most caregivers identified improvements in all targeted areas, except for sleep (41%). The majority of adolescents identified improvements in all targeted areas, although the areas of stress (59%) and sleep (66%) were the least strongly endorsed. Ninety percent of caregivers and 87.5% of adolescents stated they/their adolescent continued to use the skills learned from the program five months after it ended. Further feasibility and acceptability details can be found in Supplementary Table 1.

Open-ended questions assessed qualitative responses to the program. Overall, caregivers identified interaction with the therapist, problem-solving skills, and specific targeted modules (e.g., organization, stress) as most helpful. Caregivers identified benefits beyond the specific targets of the Epilepsy Journey program, including helping the adolescent feel they were not alone (“understanding that he isn’t the only one who has epilepsy”) and a general sense of “responsibility” for the adolescent to be independent in completing sessions. A few caregivers (2–3 total) identified specific modules (e.g., sleep, organization, mood) as least helpful for their adolescent, although most characterized all of the content as helpful. Feedback at later time points revealed a desire for longer-term follow-up or review for ongoing goals/management for their adolescent.

Adolescents similarly identified interaction with the therapist, problem-solving, and specific modules (e.g., mood, memory, sleep) as helpful. However, adolescents generally identified stress-management as the most helpful aspect of the program, differing from their caregivers. They also identified increased knowledge of epilepsy as a helpful benefit from the program. The least helpful aspect of the intervention noted by adolescents was that specific modules were not applicable to them due to previous mastery of the material or the issue being not salient to them. When asked what they would change to improve the intervention, the most common feedback from adolescents was to make the intervention more personalized or relatable to the adolescent’s specific needs, less young/childlike looking, and to change the length of the intervention, including suggestions for both lengthening and shortening of the intervention. While 88% of adolescents identified the teleconferencing with therapists as beneficial, approximately half of the adolescents reported a desire to work with the therapist face to face.

Exploratory Analyses

Caregiver-Report BRIEF Scales.—As hypothesized, *statistically significant improvements* were found over time for the global scales of the caregiver-reported BRIEF (GEC $b=-7.26$; BRI $b=-8.01$, and MI $b=-2.11$). Caregiver-reported BRIEF subscales also decreased over time (Table 2). Notably, the GEC, BRI, and Inhibition subscale demonstrated significant quadratic effects, indicating that the decrease in these subscales rebounded slightly between 2 and 5-month follow-up (Figure 2). Further, the total number of BRIEF subscale elevations decreased significantly over time, $F(3, 123) = 3.3$, $p < 0.05$ (baseline $M=4.3+2.2$; post $M=3.4+2.3$; 2-month follow-up $M=2.5+2.3$; 5-month follow-up $M=2.9+2.7$).

Regarding *clinically meaningful changes*, the proportion of patients who experienced subclinical/clinical elevations at baseline was significantly reduced for: GEC at 2- and 5-month follow-ups (62% to 33% and 34%, respectively), MI at 2- and 5-month follow (74% to 42 and 41%, respectively), Shifting at 2-month follow-up (56% to 21%), Initiation at 2-month follow (62% to 33%), and Plan/Organize at 2-month follow-up (64% to 27%; Table 3).

Self-Report BRIEF Scales.—Statistically significant improvements over time were found for the GEC ($b=-6.10$) and MI ($b=-5.64$) for the self-reported BRIEF. Improvements were also seen on the Shift, Working Memory, Plan/Organize, and Cognitive Shift subscales, with betas ranging from -1.55 to -6.80 (Table 2). The GEC, MI, and Working memory subscale had significant quadratic effects for time, indicating a slight rebound in these scores from 2 to 5-month follow-up (Figure 3). The proportion of patients who experienced subclinical/clinical elevations at baseline was significantly reduced for Shifting at 2-month follow-up (43% to 16%) (Table 3), which demonstrates clinically meaningful improvements.

Teacher-Report BRIEF Scales.—No statistically significant or clinically meaningful improvements were noted for the teacher-reported BRIEF (Tables 2–3).

Discussion

The current study was a proof of concept trial of a novel, web-based executive functioning intervention, Epilepsy Journey, for adolescents with epilepsy. Relatively low rates of attrition and high rates of completion among those who remained in the program support the program's feasibility. These findings are consistent with prior studies involving online/web-based modules and teletherapy in adolescents with neurological impairments^{28; 29} and web- or telephone-based self-management interventions for adults with epilepsy^{30–32}. Combined, our data underscore the potential value of web-based interventions in these pediatric populations. Adolescents noted busy schedules as a barrier to completing intervention programs, and thus the relatively minimal weekly time commitment of < 10 minutes for module completion and 30 minutes for problem-solving with a therapist bodes well for uptake. Additionally, few technical problems were noted and therapists were able to identify alternate means of communication, including traditional telephone interaction or other video teleconferencing, when Skype for Business failed to work. Adolescents are proficient technology users, and being able to attend sessions independently (versus being driven by caregivers) may promote a sense of self-efficacy and autonomy and reduce stigma.³³ Interestingly, despite the high ratings regarding ease of use and comfort videoconferencing with the therapist, as many as 46% of adolescents indicated that in-person meetings might be preferable. Over a decade ago, Glueckauf and colleagues initiated the first videoconferencing-based interventions for adolescents with epilepsy and their families. Study results indicated no differences in efficacy or therapeutic alliance between the family and therapist for video-conferencing versus face-to-face.^{34; 35} Further research is needed to understand the disconnect between their positive experience with telehealth and desire to meet in-person.

In exploratory analyses, caregivers reported statistically significant and clinically meaningful improvements on global executive functioning, as well as in both the hot (e.g., behavioral regulation) and cold (e.g., metacognition) EF dimensions, with the greatest improvements occurring at 2-months post treatment. These findings suggest that Epilepsy Journey may help adolescents develop better metacognitive strategies as well as behavior regulation. Improvements in the latter may drive improvements in behavioral and emotional functioning (e.g., depressive symptoms) and quality of life, although we are unable to test these associations in the current proof of concept study.

Consistent with the conceptual framework of hot and cold EF skills, pediatric epilepsy studies show that executive functioning and emotional/behavioral difficulties co-occur in youth with epilepsy^{36; 37} and are strongly related.^{8; 9} Moreover, improvements in behavior regulation among participants in the current trial suggest that Epilepsy Journey may contribute to improvements in both hot and cold EFs. Epilepsy Journey was designed to target both hot and cold EF and problems identified for problem-solving cut across both cognitive and behavioral areas. For example, some adolescents identified reducing conflict with parents as a goal for the “State of Emotional Control” in Epilepsy Journey. One strategy taught in this module is the “STARRS” method, which involves stopping, thinking, accepting, relaxing, reframing, and solving. This strategy could simultaneously address emotional/behavioral skills that are closely aligned with the EF skill of self-regulation. An important area for future research is to understand if Epilepsy Journey has an impact on emotional and behavioral functioning, sleep behaviors, and quality of life, which are likely mediated by EF.

Although self-reported improvements were somewhat less robust across EF domains, adolescents also reported significant improvements in overall EF behaviors that peaked 2-months post treatment. This pattern of improvement indicates a consolidation of treatment effects over time suggesting that practice may contribute to maintenance of effects. Scores rebounded by 5-months but remained significantly improved from baseline. In general, agreement between caregiver and self-report on the BRIEF is modest, with adolescents rating themselves more favorably than caregivers.³⁸ In fact, self-reported BRIEF scores for our sample were in the average range at baseline. Compared to caregivers, it seems possible that adolescents under-reported problems initially, thus, creating less room for significant improvement. Although caregivers and adolescents noted that they continued to use the skills after treatment ended, a booster session or intermittent check-ins may prevent relapse.

Importantly, caregiver- and self-reported improvements were also clinically meaningful,³⁹ with reductions in the number of clinically significant subscales, as well as the proportion of adolescents falling into the clinically elevated range on the GEC. These clinically meaningful improvements would be anticipated to translate to observable improvements in everyday settings, such as school. However, teachers did not report significant improvements in EF behaviors at post-treatment, which is the only time teacher report was obtained following treatment. However, several limitations are noted for the teacher-reported data, including high schools students often having several teachers but only choosing one to complete the measure and inconsistency in who completed the measure at both time points. These limitations are compounded by some participant’s crossing over school years due to

summer break. It is possible that teachers who consistently worked with the adolescent would have noted improvements over time as caregiver- and adolescent-reported improvements peaked at the 2 month follow-up. Changes in school behaviors may take longer to consolidate or further intervention targeting the school may be necessary. Heterogeneity among the classroom content and subject area may also contribute to unwanted variability and our inability to detect teacher-reported effects.

Patient- and parent-reported improvements in EF behaviors are further supported by very strong satisfaction among adolescents and their caregivers. All but one participant rated the program as helpful and more than 90% noted that they enjoyed the program, made positive changes, and were better at solving problems. Satisfaction data corresponded to BRIEF data with a slight reduction in the proportion endorsing improvements at the final follow-up. Qualitative feedback suggests that a tailored program that assigned only the modules that were relevant to the adolescent may further improve satisfaction. However, potential lack of awareness regarding deficits (an EF skill itself) may require input from multiple informants to determine the optimal set of modules for each participant. Nonetheless, high caregiver and adolescent ratings of the program and associated endorsement of improvements in targeted skills provide further support for its potential utility for adolescents with epilepsy. Taken together, these findings support the potential utility of Epilepsy Journey and the need for further testing.

As a single-arm pilot, it was not possible to examine improvements relative to treatment as usual or active control (e.g., epilepsy education). Given that participants were aware of the goals of the study, they may have reported improvements in a desire to please the therapist and investigators. Additionally, a small and heterogeneous sample precluded examination of change by epilepsy subtypes or initial profile of EF deficits. Data from multiple informants was a strength, but other EF measures including neuropsychological tests and classroom observations may afford additional insights and obviate social desirability biases. Further, independent self-report measures of problem-solving or other EF constructs may have been beneficial. Notably, studies have shown that multi-method, multi-informant reporting is optimal for EF measurement given the strengths and weaknesses of various approaches.⁴ Finally, we were unable to examine the role of EF as it relates to emotional/behavioral functioning in Epilepsy Journey given the small sample size. A larger trial with sufficient power to test EF as a mediator of our Epilepsy Journey intervention and psychosocial outcomes would be beneficial.

Our findings support the feasibility and acceptability of Epilepsy Journey and despite the absence of a control group, exploratory analyses suggest Epilepsy Journey may be helpful in reducing EF deficits in adolescents with epilepsy. The current pilot sheds light on how the program can be optimized to further enhance benefits. Future randomized controlled trials are needed to establish efficacy relative to a comparison condition, evaluate the long-term effects of Epilepsy Journey, and identify whether EF is a moderator or mediator of treatment effects for emotional/behavioral functioning or quality of life. If validated through further research, Epilepsy Journey has the promise to reduce EF problems and associated morbidity in a cost-efficient and easily delivered fashion.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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Key Points

- Interventions to improve executive functioning in adolescents with epilepsy are lacking.
- Epilepsy Journey is a web-based intervention with telehealth sessions developed for adolescents with epilepsy.
- Epilepsy Journey was well-received and perceived as beneficial by adolescents with epilepsy and their caregivers.

Figure 1.
Epilepsy Journey CONSORT Diagram

Figure 2.
Caregiver-reported BRIEF scores over time

Figure 3.
Child-reported BRIEF scores over time

Table 1.

Demographic data (n = 39)

	N (%) or M±SD
Age	15.3 ± 1.3 years
Sex	
• Females	132 (56%)
• Males	105 (44%)
Race/Ethnicity	
• White: Non-Hispanic	34 (87%)
• Black: Non-Hispanic	1 (3%)
• Bi/multiracial or other	4 (10%)
Seizures in the Past Three Months	
• Yes	15 (38.5%)
• No	24 (61.5%)
Seizure Type and Etiology	
• Idiopathic Focal	5 (13%)
• Idiopathic Generalized	21 (54%)
• Idiopathic Unclassified	11 (28%)
• Symptomatic or Cryptogenic Focal	2 (5%)
Antiseizure medication	
• Monotherapy	32 (82%)
• Polytherapy	7 (18%)
Years Since Diagnosis	1.9 ± 2.0 years
PESQ Total Score	16.8 ± 17.0
Insurance Status	
• Private	28 (71.8%)
• Medicaid/Public	8 (20.5%)
• Uninsured	3 (7.7%)

Note:

* PESQ=Pediatric Epilepsy Side Effects Questionnaire

Table 2.

Longitudinal Mixed Model Results

Outcome	Baseline	Post	Fup1	Fup2	b*	std. error	p-value	95% CI
Caregiver-Report								
**Global Executive Composite	63.2 (9.1)	58.9 (9.1)	55.3 (9.2)	57.0 (10.4)	-7.26	2.22	.001	-11.60, -2.91
**Behavioral Regulation Index	60.8 (10.4)	56.7 (10.3)	52.4 (8.6)	55.0 (10.9)	-8.01	1.94	<.001	-11.82, -4.20
• **Inhibit	56.5 (13.6)	51.9 (12.4)	49.5 (8.9)	51.2 (11.9)	-8.72	2.44	<.001	-13.50, -3.93
• **Shift	60.5 (11.2)	57.0 (11.2)	53.5 (9.3)	55.3 (10.3)	-1.28	0.48	.008	-2.21, -0.34
• **Emotional Control	61.1 (9.8)	58.5 (10.3)	53.5 (9.4)	56.4 (11.6)	-1.74	0.58	.003	-2.87, -0.61
*Metacognition Index	63.6 (8.9)	59.5 (9.1)	56.5 (10.4)	57.6 (10.6)	-2.11	0.53	<.001	-3.14, -1.08
• *Initiate	63.9 (11.1)	60.4 (9.2)	56.3 (8.9)	58.3 (9.7)	-2.11	0.55	<.001	-3.19, -1.02
• **Working Memory	66.2 (10.5)	61.3 (12.1)	59.8 (12.6)	58.7 (12.3)	-2.01	0.55	<.001	-3.08, -0.94
• **Plan/Organize	62.0 (10.0)	57.6 (9.7)	56.2 (11.4)	57.7 (12.3)	-1.61	0.56	.004	-2.70, -0.52
• **Organization of Materials	55.5 (11.1)	54.0 (10.8)	51.7 (10.9)	51.8 (11.5)	-1.30	0.50	.009	-2.29, -0.32
• **Monitor	58.2 (9.9)	55.7 (10.0)	52.1 (9.6)	54.3 (12.0)	-1.92	0.62	.002	-3.15, -0.70
Adolescent-Report								
**Global Executive Composite	54.4 (11.1)	52.3 (12.5)	47.4 (10.6)	50.4 (12.2)	-6.10	2.42	.01	-10.86, -1.35
Behavioral Regulation Index	53.3 (12.1)	51.5 (11.4)	45.9 (11.2)	49.5 (13.4)	-1.04	0.57	.07	-2.16, 0.09
• Inhibit	48.9 (11.1)	48.5 (10.4)	45.1 (10.6)	47.0 (11.2)	-0.25	0.56	0.65	-1.33, 0.84
• **Shift	54.9 (12.8)	52.2 (13.4)	46.4 (11.9)	49.8 (13.3)	-1.68	0.65	.01	-2.95, -0.41
• Emotional Control	56.0 (11.7)	55.4 (11.1)	50.1 (10.1)	53.4 (12.0)	-0.74	0.57	.20	-1.86, 0.38
• Monitor	50.6 (10.5)	47.7 (11.1)	44.4 (10.6)	46.9 (11.6)	-1.09	0.67	.10	-2.40, 0.22
*Metacognition Index	54.5 (10.7)	52.5 (13.6)	48.9 (9.5)	51.4 (11.0)	-5.64	2.27	.01	-10.09, -1.18
• **Working Memory	56.9 (12.9)	54.6 (14.8)	51.3 (13.1)	53.3 (12.6)	-6.80	2.58	<.01	-11.87, -1.74
• **Plan/Organize	52.2 (9.9)	50.3 (12.9)	46.4 (8.6)	48.3 (10.1)	-1.55	0.63	.01	-2.78, -0.33
• Organization of Materials	50.7 (9.0)	49.8 (9.9)	48.4 (8.2)	49.9 (9.1)	-0.82	0.43	.06	-1.66, 0.02
• Task Completion	54.6 (12.2)	53.3 (12.4)	50.1 (10.3)	53.0 (11.5)	-0.36	0.51	.48	-1.37, 0.64
Behavioral Shift	54.1 (12.8)	53.5 (14.3)	47.7 (11.9)	52.1 (13.4)	-0.86	0.76	.26	-2.35, 0.63

Outcome	Baseline	Post	Fup1	Fup2	η^2	std. error	p-value	95% CI
*Cognitive Shift	54.5 (11.7)	50.2 (11.8)	40.1 (11.9)	47.5 (12.2)	-2.20	0.58	<.001	-3.32, -1.07
Teacher-Report								
Global Executive Composite	60.9 (16.5)	56.6 (9.1)			-1.08	2.10	.61	-5.19, 3.03
Behavioral Regulation Index	58.8 (16.5)	54.4 (11.0)			0.31	2.52	.90	-4.62, 5.24
• Inhibit	52.8 (11.7)	51.3 (10.3)			1.09	2.49	.66	-3.79, 5.98
• Shift	62.7 (20)	55.8 (10.1)			-0.33	3.53	.93	-7.25, 6.60
• Emotional Control	60.9 (21.0)	55.7 (14.1)			-1.14	2.33	.62	-5.71, 3.43
Metacognition Index	60.7 (15.5)	57.2 (9.9)			-1.53	2.35	.52	-6.14, 3.09
• Initiate	61.7 (16.5)	56.9 (12.4)			-2.20	3.76	.56	-9.57, 5.17
• Working Memory	64.2 (17.0)	61.8 (13.9)			0.69	3.01	.82	-5.21, 6.59
• Plan/Organize	59.1 (14.0)	55.7 (10.6)			-2.93	2.94	.32	-8.69, 2.83
• Organization of Materials	55.9 (16.0)	52.6 (12.5)			-3.40	3.48	.33	-10.22, 3.42
• Monitor	55.1 (13.1)	53.7 (8.2)			0.99	2.28	.66	-3.48, 5.46

* Note: coefficient for time; covariates for all models included child race, years since diagnosis, seizure type, presence of seizures in the last 3 months, and total side effects;

** represents significant changes over time.

Table 3.

Proportion of patients with subclinical/clinical elevations on the BRIEF

	Baseline	Post-treatment	2-month follow-up	5-month follow-up
Parent BRIEF Subscales				
<i>Global Executive Composite</i>	62%	44%	33% *	34% *
<i>Behavioral Regulation Index</i>	46%	31%	25%	28%
• Inhibit	31%	16%	17%	17%
• Shifting	56%	44%	21% **	38%
• Emotional Control	54%	50%	33%	38%
<i>Metacognition Index</i>	74%	53%	42% **	41% **
• Initiate	62%	53%	33% *	52%
• Working Memory	72%	53%	54%	45% *
• Plan/Organize	64%	44%	27% *	45%
• Org. of Materials	41%	44%	29%	24%
• Monitor	46%	41%	25%	31%
Adolescent BRIEF Subscales				
<i>Global Executive Composite</i>	35%	27%	16%	24%
<i>Behavioral Regulation Index</i>	27%	21%	20%	20%
• Inhibit	11%	18%	16%	16%
• Shifting	43%	30%	16% *	32%
• Emotional Control	30%	24%	12%	24%
• Monitor	22%	18%	8%	16%
<i>Metacognition Index</i>	32%	27%	12%	28%
• Working Memory	41%	30%	20%	36%
• Plan/Organize	22%	24%	8%	12%
• Org. of Materials	16%	9%	8%	12%
• Task Completion	30%	30%	16%	28%
<i>Behavioral Shift</i>	35%	30%	24%	36%
<i>Cognitive Shift</i>	32%	24%	8% *	20%
Teacher BRIEF Subscales				
<i>Global Executive Composite</i>	39%	39%		
<i>Behavioral Regulation Index</i>	30%	22%		
• Inhibit	17%	17%		
• Shift	43%	33%		
• Emotional Control	30%	28%		
<i>Metacognition Index</i>	39%	39%		
• Initiate	52%	44%		
• Working Memory	48%	50%		

	Baseline	Post-treatment	2-month follow-up	5-month follow-up
• Plan/Organize	39%	33%		
• Organization of Materials	30%	17%		
• Monitor	30%	27%		

* Note: $p < 0.05$,

** $p < 0.01$,

*** $p < 0.001$