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Brief Behavioral Intervention for Disruptive Behavior in a Child with a Hypothalamic Hamartoma: A Case Report

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Abstract
Most children with hypothalamic hamartoma (HH) manifest symptoms of epilepsy and associated cognitive deficits and behavioral difficulties as well as central precocious puberty (CPP). However, there is little to no research examining behavioral difficulties in children with HH *without* epilepsy, nor is there research examining treatments to address the behavioral difficulties of patients with HH *without* epilepsy. In the current case report, the authors implemented a validated parent management training program [the Brief Behavioral Intervention (BBI)], to treat symptoms of ADHD and disruptive behavior in a 6-year-old female patient with HH and CPP. The family participated in six BBI sessions over a period of 8 weeks. Parent behavioral ratings suggested significant reductions of symptoms of ADHD and disruptive behaviors to the normal range. The current case report demonstrates the effectiveness of the BBI program in the treatment of behavioral difficulties in a patient with HH and CPP. Further, the present study explores behavioral manifestations rarely explored in patients with HH *without* epilepsy.

Keywords Hypothalamic hamartoma · Central precocious puberty · Parent management training · Attention-deficit/hyperactivity disorder · Brief behavioral intervention

Hypothalamic hamartoma (HH) is a rare, congenital, benign tumor within the hypothalamus, affecting approximately 1 in 50,000–100,000 individuals (Weissenberger et al., 2001). Some researchers describe the clinical features of HH as a spectrum (De La Mota et al., 2012), whereas other researchers suggest that the clinical features associated with HH usually consist of epilepsy, intellectual disability, and central precocious puberty (CPP) (Prigatano, 2007). However, the size and location of the HH often determines the manifestation of symptoms. For instance, intrahypothalamic (also called sessile) hamartomas are posteriorly located on the hypothalamus and are highly associated with treatment-resistant epilepsy [usually beginning with gelastic (laughing) seizures], subsequent intellectual disability, and behavioral difficulties, as well as CPP (Chan et al., 2010; Killeen, Bunch, & Kerrigan, 2017). In contrast, parahypothalamic (also called pedunculated) hamartomas are located on the anterior hypothalamus and are typically associated with CPP only (Chan et al., 2010; Killeen et al., 2017).

The majority of research related to HH has generally focused on patients with gelastic seizures and the associated behavioral difficulties related to those seizures. Weissenberger et al. (2001) found that 83% of families with children with HH and epilepsy reported significant behavioral problems including poor frustration tolerance, physical aggression, temper tantrums, and emotional dysregulation. Other researchers have found that 49% of patients with HH and gelastic seizures manifest cognitive deficits and 31% of patients exhibited behavioral difficulties (Nguyen et al., 2003).

Although there is ample research related to behavioral difficulties associated with HH and gelastic seizures, research related to the behavioral manifestations of patients with HH *without* gelastic seizures is scarce. To date, there are only two known studies that have examined patients with HH, *without* seizures, and behavioral difficulties. Nagaki et al. (2010) conducted a case report describing two patients with HH and CPP: one with and one without gelastic seizures. The patient *without* gelastic seizures was a 4-year 10-month-old male who began manifesting symptoms of CPP at 7 months of age

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and behavioral difficulties (e.g., hyperactivity and aggression) around 18 months of age. Following surgical removal of the HH as well as participation in gonadotropin-releasing hormone (GnRH) analog treatment, the patient's behavioral difficulties reportedly improved, which supports previous research reporting improvements of behavior following HH resection (Mullatti et al., 2003) and GnRH analog treatment (Xhrouet-Heinrichs et al., 1997). In the second study, Katayama, Yamashita, Yatsuga, Koga, & Matsuishi, (2016) conducted a case report describing a 9-year-old male patient with HH, *without* gelastic seizures, who manifested CPP, ADHD-like behavior, and conduct disorder. Despite successfully managing the patient's ADHD symptoms with methylphenidate as well as the patient's symptoms of CPP with GnRH analog treatment, the authors did not report any success addressing the patient's behavioral difficulties (e.g., emotional dysregulation, physical aggression). These two case reports offer some insight into the behavioral manifestations of patients with HH and CPP *without* gelastic seizures. Further, given the patients' mixed behavioral responses to medical treatment, these studies illustrate that the patients' associated behavioral difficulties are likely multifactorial in origin (i.e., related to the HH itself, associated with CPP, and/or a response to psychosocial factors) (Katayama et al., 2016; Nagaki et al., 2010) and would benefit from behavioral intervention.

In addition to limited research related to the behavioral manifestations of patients with HH *without* gelastic seizures, there have been no studies to date regarding behavioral intervention targeting disruptive behaviors in children with HH. An evidence-based treatment to address behavioral difficulties in children is parent management training (PMT), which involves teaching parents necessary behavior management strategies to increase prosocial skills and decrease disruptive behaviors (Eyberg, Nelson, & Boggs, 2008). Although not yet studied with HH patients, PMT has been implemented with a wide variety of pediatric populations with co-occurring disruptive behaviors including epilepsy (Treble-Barna, Chapman, Schwartz, & Axelrad, 2013), traumatic brain injury (Cohen, Heaton, Ginn, & Eyberg, 2012; Garcia, Barroso, Kuluz, & Bagner, 2016), autism spectrum disorder (Zlomke, Jeter, & Murphy, 2017), and disorders of sex development (Jensen, Palacios, & Drury, 2011). Although there are many forms of PMT available to families (e.g., Parent–Child Interaction Therapy and Kazdin's PMT), the majority of PMT programs are time intensive (e.g., lasting between 13 and 27 sessions) (Axelrad, Garland, & Love, 2009) resulting in high rates of parental attrition (Fernandez & Eyberg, 2005) as well as an increase in barriers to treatment (e.g., cost of services, absences from school and work) (Axelrad et al., 2009).

In the current case report, the authors implemented a validated PMT program, the Brief Behavioral Intervention program (BBI; Axelrad et al., 2009, Axelrad, Butler, Dempsey, & Chapman, 2013; Axelrad & Chapman, 2016) to treat symptoms of ADHD and disruptive behavior in a 6-year-old female patient with parahypothalamic hamartoma. The objectives of this study were two-fold: (1) demonstrate the effectiveness of the BBI program to treat a patient with HH and co-occurring symptoms of ADHD and disruptive behaviors and (2) add to the literature by describing a patient with HH *without* gelastic seizures and the associated behavioral manifestations.

Methods

Patient Presentation

The patient is a 6-year-old Asian American female with a history of early onset breast development and a diagnosis of premature thelarche (e.g., early development of breast tissue) at 2 years of age. At 5 years of age, the patient was diagnosed with CPP and began receiving Lupron injections (sex hormone suppression therapy). An MRI revealed a parahypothalamic hamartoma when the patient was 5 years of age. Although the patient did not experience any seizure activity, she displayed difficulty with reading development as well as symptoms of inattention, hyperactivity, and other disruptive behaviors, warranting a referral for a neuropsychological evaluation to assess the possible impact of her medical conditions on her neurodevelopment.

As part of the neuropsychological evaluation, relevant history was obtained through clinical interview with the patient's mother. Perinatal history was reportedly unremarkable, and achievement of early developmental milestones was within age expectations. Both English and Arabic were reported to be spoken in the home, but English was described as the patient's primary language since infancy. Behavioral difficulties first emerged between 24 and 28 months of age, which involved screaming that was difficult to control. Behavioral difficulties such as non-compliance, occasional hitting of an elder sibling, and screaming in the home setting remained present at the time of the neuropsychological evaluation, but no such concerns were present at school. The home-based behaviors were described as disruptive to completion of homework involving language arts activities. Despite no behavioral difficulties at school, the patient was consistently observed to become anxious and withdrawn during language arts tasks in the school setting.

Her academic history was remarkable for difficulty learning letters and developing early reading skills despite participation in a private preschool followed by completion of 1 year of pre-Kindergarten and 1 year of Kindergarten in an English as a Second Language program within the public school setting. Approximately 6 months prior to her referral

for a neuropsychological evaluation, a psychoeducational evaluation was conducted through her school. The patient's Cognitive Academic Language Proficiency scores revealed greater proficiency in English than Arabic at that time. The results of her school-based evaluation were determined to be consistent with dyslexia, and both dyslexia intervention services as well as educational accommodations (preferential seating, note-taking assistance, oral reading assistance, opportunities to correct spelling errors, and extended time) were recommended at that time.

With regard to psychosocial history, the patient lived with her biological parents and two older siblings. The patient's father successfully completed high school, and her mother held a Master's degree and was enrolled in a PhD program at the time of referral. The patient's family denied any significant mental health history within the immediate or extended family.

As part of her outpatient neuropsychological evaluation, several measures were administered to assess the patient's cognitive functioning (Wechsler Intelligence Scales for Children, 5th Edition, prorated [WISC-V]; Wechsler, 2014), memory (select subtests from the Wide Range Assessment of Memory and Learning-II [WRAML-II]; Sheslow & Adams, 2003), visual motor skills (The Beery–Buktenica Developmental Test of Visual Motor Integration; Beery, Buktenica, & Beery, 2010), fine motor skills (The Purdue Pegboard; Tiffin & Asher, 1948), academic achievement (Kaufman Test of Educational Achievement-3rd Edition [KTEA-3]; Kaufman & Kaufman, 2014), attention regulation (Conners Kiddie Continuous Performance Test-II [K-CPT-2]; Conners, 2015), and everyday behavioral, executive, and emotional functioning (Behavior Assessment System for Children, Third Edition—Parent Informant [BASC-3]; Reynolds, Kamphaus, & Vannest, 2015; Behavior Rating Inventory of Executive Function [BRIEF]—parent informant; Gioia, Isquith, Guy, & Kenworthy, 2000; NICHQ Vanderbilt Assessment Scale—Parent Informant; available at https://www.nichq.org/resource/nichq-vanderbilt-assessment-scales; Revised Children's Manifest Anxiety Scale, Second Edition; Reynolds & Richmond, 2008). Results of the WISC-V revealed average performance across scales assessing her verbal reasoning (Verbal Comprehension Composite = 92), working memory (Working Memory Composite = 100), and psychomotor speed (Processing Speed Composite = 92), and she demonstrated a clinically significant strength with Superior range performance in visual reasoning (Visual–Spatial Composite = 122). Similarly, she demonstrated high average visual–spatial matching skills (Standard Score = 117). Her performance across measures of memory and visual motor integration was also average and commensurate with her general cognitive abilities. In terms of academic achievement, the patient demonstrated average to low average math computation abilities relative to other children her age (Standard Score = 89), but her language-based academic skills were relatively weaker and ranged from low average for both letter/word recognition skills and phonological processing skills (Standard Scores = 80 and 85, respectively) to below average for her reading comprehension (Standard Score = 70), spelling (Standard Score = 65), and written expression skills (Standard Score = 66). Consistent with her existing educational classification of dyslexia, these scores and her overall pattern of academic performance was conceptualized as dyslexia.

Performance-based and parent report measures of attention and executive functioning identified at-risk to clinically significant levels of inattention, hyperactivity/impulsivity, and disruptive behaviors, whereas examiner observation and clinical interview identified symptoms of anxiety, parent, and child self-report ratings were not significant. Due to the neuropsychological evaluation taking place in between academic calendar years, behavioral rating forms were unable to be obtained from the patient's teacher. Therefore, the patient was given a diagnosis of Unspecified Disruptive Impulse-Control and Conduct Disorder. The patient and her family were subsequently referred for PMT to address her behavioral difficulties. School-based interventions and accommodations were also recommended to address the impact of her language-based academic deficits and parent-reported deficits with attention in the school setting.

Upon starting school and enrolling in PMT, the patient's mother and teacher completed a set of behavioral measures (e.g., BASC-3, Disruptive Behavior Rating Scale, Eyberg Child Behavior Inventory), which were reviewed by the clinicians providing PMT. Both parent and teacher measures identified at-risk to clinically significant levels of inattention, hyperactivity/impulsivity, and disruptive behaviors. Taken together, the patient's overall behavioral presentation was conceptualized as Attention-Deficit/Hyperactivity Disorder and Unspecified Disruptive Impulse-Control and Conduct Disorder.

Procedure

The patient's mother provided written, informed consent to participate in the Institutional Review Board approved study examining the effectiveness of the BBI program (Axelrad & Chapman, 2016) on treating disruptive behaviors in pediatric patients. The patient and her mother attended six 50-min family therapy sessions as a part of the BBI program (Axelrad & Chapman, 2016). The program was designed to facilitate short-term PMT in tertiary care centers and has demonstrated significant improvement in child behavior at home and school immediately following and 1 year post intervention (Axelrad et al., 2013). BBI is designed to be administered in weekly sessions conducted by a BBI-trained

clinician with credentials ranging from advanced graduate student to doctoral-level practitioner.

BBI consists of five core sessions focused on specific behavior management skills (Table 1). The first session focuses on exploring antecedents and consequences of problem behavior and can occur within the context of an initial diagnostic interview. Core sessions two through five focus on skill building and begin with introducing child-directed play and planning for implementation within the context of family's routine, cultural practices, and family structure. The third session introduces differential attention including frequent positive attending to desired behavior (e.g., labeled praise) and active ignoring of attention-seeking behavior (e.g., tantrums, screaming, invasion of personal space). This session also includes soliciting information from parents regarding how the practice of positive attention and active ignoring fit within their cultural and parenting practices. The fourth session focuses on delivering effective commands in a concise developmentally appropriate manner, that directly describes the desired behavior as well as outlining consistent consequences for non-compliance that are specifically tailored to the family. The last of the core sessions introduces a time-out consequence for aggressive behavior as an elevated form of differential attention. After core sessions have been completed, additional sessions are provided as needed to address common difficulties at this age range (e.g., sleep, toileting). A follow-up session is scheduled 1 month post-treatment, as indicated by parent preference and clinician judgment. Each session is accompanied with written handouts reviewing the skills introduced and a behavior log to track behaviors over the following week. Completion of the weekly behavior log involves parents identifying problematic behaviors that occur as well as the antecedents that directly precede the behavior and the consequences that directly follow the behavior. Review of the behavior log allows clinicians to evaluate parents' implementation of newly learned behavioral strategies as well as monitor progress in the patient's behavior outside of session. Modifications to the BBI protocol based on cultural considerations are implemented after discussions with the family and therapists to insure modifications that are culturally responsive and in keeping with the BBI protocol.

Following a diagnostic interview with a licensed clinical psychologist (MEA) and post-doctoral fellow (RF), the patient and her mother participated in six total sessions over the course of 8 weeks. Therapy duration was prolonged to 8 weeks given family's preference. Sessions were conducted in English, by a post-doctoral fellow (RF) and an advanced graduate student trained in the protocol (GB), who participated in sessions as a co-therapist and learner. Both clinicians were supervised by MEA throughout treatment in an outpatient psychology clinic within a children's hospital/academic medical setting.

The patient and her mother attended each session together to allow for in vivo modeling and coaching of each BBI skill. Each session began with a review of the previously learned skill and behavior log, which the patient's mother returned each session. Clinicians invited information regarding success and challenges implementing previously learned skills, problem solved to family satisfaction, and proceeded to introduce the next skill. The patient was encouraged to play with toys in the session room as her mother spoke with clinicians. Notably, aside from the patient engaging in mild non-compliance in response to her mother's instructions in session (e.g., dawdling rather than immediately following her mother's instructions), the patient was not observed to engage in any disruptive behaviors in session. As skills were introduced and acquired, clinicians highlighted opportunities for the patient's mother to practice skills in session throughout treatment (e.g., clinicians prompted the patient's mother to engage in child-directed play, offer labeled praise or use active ignoring, and provide the effective command to clean up at the end of session).

Although the patient's mother did not report specific cultural components that required a modification to the BBI protocol, she shared information regarding family dynamics

Table 1 Summary of the Brief Behavioral Intervention (BBI) Program (Table adapted from Axelrad & Chapman, (2016))

Session 1: Daily routines	• Provide overview of treatment and how it relates to target behaviors • Review child's daily routine/behaviors/problem times with specific emphasis on when tantrums occur, when child sleeps, how they get to sleep, etc. • Collection of pre-treatment behavioral measures
Session 2: Special time	• Introduce child-directed interaction • Model and allow parents to practice special time
Session 3: Differential attention	• Introduce practice of praising desired behavior and ignoring undesired behavior • Problem solve and prepare for differential attention in public
Session 4: Effective commands	• Introduce effective command language, timing, and behavior contingencies
Session 5: Time-out	• Introduce principles of time-out as a disciplinary strategy for aggression only • Problem solve use of time-out
Follow-up session	• Problem solve additional concerns such as toileting, school problems, sleep • Collection of post-treatment behavioral measures

and values that were notable during treatment. Due to the patient's parents being separated and living apart throughout treatment as well as her mother being enrolled as a full-time student, discussions related to barriers of treatment implementation (e.g., division of responsibilities between caregivers and limited time) occurred on a weekly basis. Therefore, the clinicians assisted the patient's mother with finding times in the evening and weekends to engage in child-directed play with the patient as well as activities that required minimal effort to set up and clean up (e.g., coloring, doll house). Further, the patient's mother reported that her family placed high value on respect and the patient's disobedient behavior was particularly remarkable to the family and distressing to her mother. Specifically, the patient's mother noted that it was common for her to host family members and family friends in their home on the weekends with the expectation that the children play with one another while the adults socialize in a separate room. Per parent report, the patient frequently interrupted adult conversations and refused to play with other children, which often resulted in conflict among the patient's mother and her guests. She also noted praise for desired behavior was not a common practice for her or her extended family. Therefore, the clinicians spent time facilitating discussions about which effective commands to use (e.g., "I need you to play with the other children for 15 min."), appropriate consequences (e.g., "If you play with the other children until the timer goes off, I will talk with you for 5 min."), how to use preventative praise (e.g., intermittently checking on the patient prior to interruptions and saying, "Thank you for playing with the other children while I talk with my guests."), and how to respond to comments or concerns from guests in respectful manner (e.g., "Thank you for your concern. We are trying something new to address her behavior."). Additionally, the patient's mother preferred to conduct her descriptions during child-directed interactions in Arabic. Clinicians encouraged this practice at home and asked the patient's mother to speak in English during her in-session practices to allow for clinician coaching. However, given the patient's mother's general preference for speaking to her children in Arabic, the clinicians spent time eliciting feedback from the patient's mother regarding how the wording of questions or commands may differ in Arabic versus English.

Measures

The patient's mother completed rating scale measures pre- and post-treatment. The measures were a part of the standard BBI research protocol battery.

The *Eyberg Child Behavior Inventory* (ECBI; Eyberg, 1999) is a measure of disruptive behavior in the home. Parents report on the frequency of specific disruptive behaviors on a 7 point scale ranging from never to always as well as answer yes or no question asking if each behavior presents a problem in the home environment. Sample items include "dawdles in getting dressed" and "whines." Raw scores are added and converted to *T* scores to yield two subscales, and Intensity scale (i.e., frequency of behavior) and a Problem scale (i.e., severity of behavior). *T* scores of 60 or higher are in the clinical significant range. Test–retest reliability for the ECBI Intensity scale is 0.94 (Eyberg & Pincus, 1999).

The *Disruptive Behavior Rating Scale-Parent Report* (DBRS; Barkley & Murphy, 1998) is a parent report measure of inattention, hyperactivity–impulsivity, and oppositional defiant behavior among school-aged children. Parents rate the frequency of behaviors over the past 6 months on a 4-point Likert-type scale. The DBRS yields four subscales that correspond with symptom criteria for ADHD Primarily Inattentive type, ADHD Primarily Hyperactive type, ODD, and Conduct Disorder. Ratings of 2 or 3 are considered positive indicators of symptoms. Ratings of 6 and above on the ADHD scales and 4 and above on the ODD scale indicate clinical significance. Reliability or validity information is not available for this measure.

The *Behavior Assessment Scale for Children-3 Parent Report Scale* (BASC-3-PRS) is a broadband measure of behavioral, social, and emotional functioning in children. Parents rate the frequency of specific behaviors. Scores are reported as *T* scores using a norm-referenced sample relative to age and sex. *T* scores of 70 and above are within the "clinically significant" range and *T* scores between 60 and 69 are considered "at-risk." *T* scores under 60 are classified as within the normative range for age expectations. The test–retest reliability of the scale ranges from 0.91 to 0.93 (Reynolds et al., 2015). The patient's mother completed the Child version of this scale.

The *Parenting Stress Index/Short Form* (PSI-SF; Abidin, 1990) is parent self-report measure of stress in the parent–child relationship. The PSI-SF yields three subscales (Parent Distress, Parent–Child Dysfunctional Interactions, and Difficult Child) and a total score. Raw scores are tallied and converted to *T* scores. *T* scores above 70 are considered elevated in the clinically significant range. Reliability on subscales and total score range from 0.75 to 0.98.

Results

Reliable Change Indices (RCI; Jacobson & Truax, 1991) were calculated in order to assess clinically significant change across treatment. Reliable improvement in rating measures (RCI scores > 1.96) was indicated for three of the four rating scale measures (Table 2). RCI scores could not be calculated for the DBRS given the lack of published reliability and validity indices.

Table 2 Reliable change statistics for Parent Rating Scales

Rating scales	Reliability	SD	SEM	Total Score		Δ	SΔ	RCI
				Pre-treatment	Post-treatment			
ECBI	0.94	13.43	3.29	75	56	19	4.65	4.08
BASC-3 PRS								
Externalizing problems	0.93	16.26	4.30	79	56	23	6.08	3.78
Internalizing problems	0.91	24.04	7.21	82	48	34	10.20	3.33
Behavior Symptoms Index	0.92	17.68	5.00	76	51	25	7.07	3.54
PSI-SF	0.98	21.92	3.10	87	56	31	4.38	7.07

SD, standard deviation; *SEM*, standard error of measurement; Δ, difference value between pre- and post-treatment; *S*Δ, standard deviation of the error of measurement of the difference score; *RCI*, Reliable Change Index

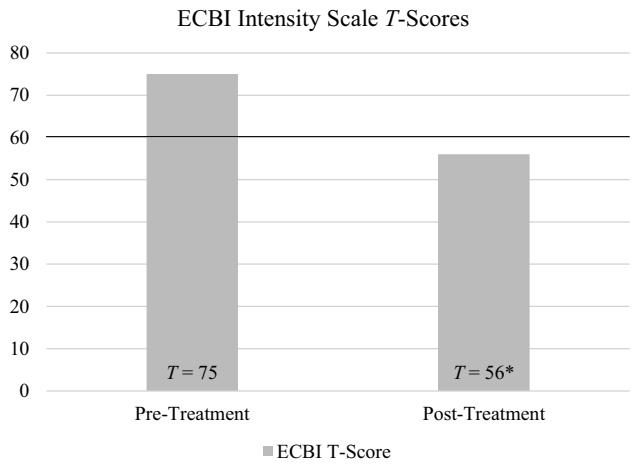

Fig. 1 Eyberg Child Behavior Index Intensity Scale Scores at pre- and post-treatment. Line indicates a clinically significant *T* score of 60. A * indicates clinically significant change and reliable decrease in scores

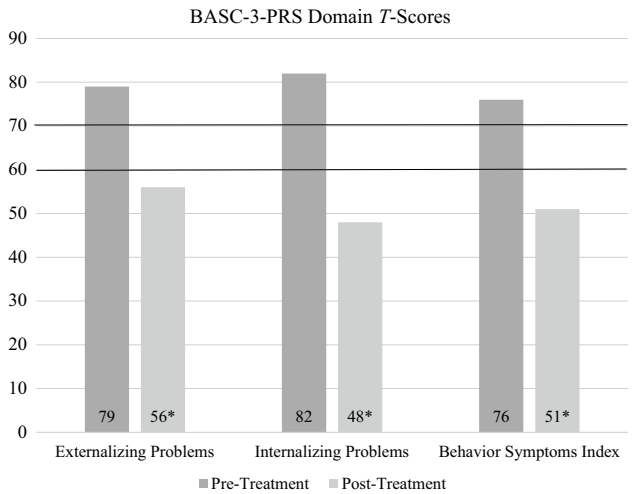

Fig. 2 BASC-3 Domain Scores at pre- and post-treatment. *T* scores between 60 and 69 indicate scores within the at-risk range. *T* scores at or above 70 indicate scores within the clinically significant range. A * indicates clinically significant change and reliable decrease in scores

Prior to the beginning of treatment, the patient's mother reported a clinically significant frequency of disruptive behaviors on the ECBI Intensity Scale ($T=75$). At the end of treatment, the patient's mother's post-treatment scores on the ECBI Intensity Scale fell within normal range ($T=56$), representing a significant decrease in frequency of disruptive behaviors from pre- to post- treatment (RCI = 4.08, $p<0.5$) (Fig. 1). Similar to her pre-treatment ratings on the ECBI Intensity Scale, the patient's mother reported clinically significant ratings on the BASC-3 across the following domains: Externalizing Problems ($T=79$), Internalizing Problems ($T=82$), and the Behavior Symptom Index ($T=76$). Post-treatment scores on the BASC-3 yielded reliable improvement across all three domains: Externalizing Problems ($T=56$; RCI = 3.78, $p<0.5$), Internalizing Problems ($T=48$; RCI = 3.33, $p<0.5$), and Behavior Symptom Index ($T=51$; RCI = 3.54, $p<0.5$) (Fig. 2). Of note, on each domain, pre-treatment *T* scores were within the clinically significant range and post-treatment *T* scores were within the normal range.

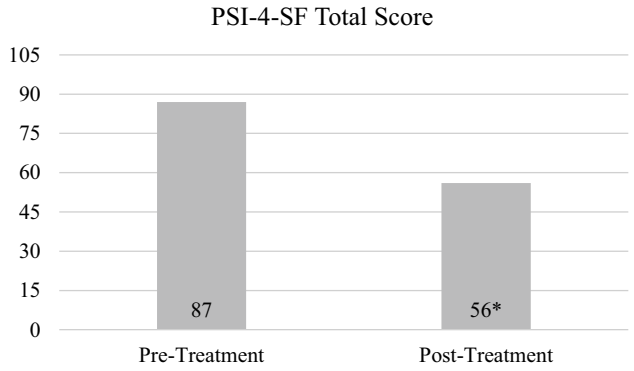

Fig. 3 PSI-SF Total Scores at pre- and post-treatment. A * indicates clinically significant change and reliable decrease in scores

With regard to the patient's mother's ratings on the PSI-SF, pre- to post-treatment scores represented improvement in Total Stress from the clinically significant range to the normal range (RCI of 7.07, $p < 0.5$) (Fig. 3). Lastly, subscale scores on the DBRS scales revealed a decrease in symptom report from scores in the clinically significant range to scores in the developmentally typical range (Fig. 4).

Discussion

This case report illustrates the successful implementation of the BBI program in treating symptoms of ADHD and disruptive behaviors in a 6-year-old female patient with parahypothalamic hamartoma and CPP. Treatment was associated with clinically significant reductions in parent ratings of symptoms of ADHD and disruptive behaviors, decreased attention-seeking and physically aggressive behaviors in the home setting reported by parent, and increased compliance with effective commands reported by parent and observed in session.

In addition to demonstrating clinically significant change in behavior in a medical presentation not previously reported, this study offers a number of additional strengths. First, it adds to the literature by demonstrating the effectiveness of the BBI program (i.e., PMT) with patients with parahypothalamic hamartoma and co-occurring disruptive behaviors. Research has demonstrated that PMT is effective in addressing disruptive behaviors in various pediatric populations (Garcia et al., 2016; Jensen et al., 2011; Treble-Barna et al., 2013; Zlomke et al., 2017); therefore, the current study further reinforces the expansion of the BBI program to additional pediatric populations (e.g., HH). Second, this study adds to the development of an evidence base that offers insights into the behavioral manifestations of patients with HH with disruptive behavior

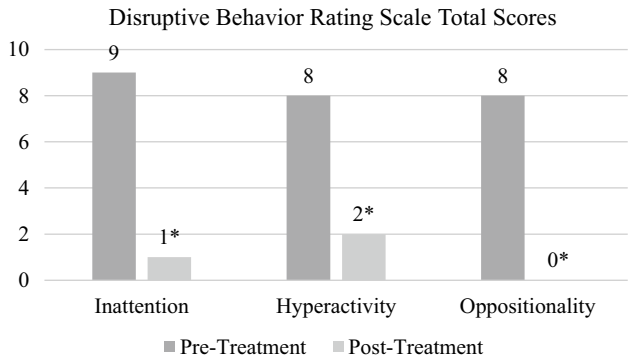

Fig. 4 DBRS Subscale Scores at pre- and post-treatment. Scores above 6 on the Inattention and Hyperactivity scales indicated clinically significant elevations. Scores above 4 on the Oppositionality scale indicate clinically significant elevations. A * indicates a clinically significant change in scores

and *without* gelastic seizures (Katayama et al., 2016; Nagaki et al., 2010). Third, the current study offered a unique opportunity to discuss and address cultural factors within the context of treating a pediatric patient. Specifically, questions regarding the family's dynamics, values, and language were actively assessed and discussed throughout treatment in order to provide culturally responsive treatment. Lastly, this study illustrates the value of working within an academic medical setting by being able to consult with other specialties including neuropsychology, endocrinology, and oncology in order to provide true interdisciplinary care.

Although results from this case study are quite promising, it is not without its limitations. The present study is limited by those elements that are characteristic of case study designs. Specifically, the study involves a single case without a control group offering limited generalizability and making it difficult to distinguish the direct effects of treatment on changes in behavior from those of time. In consideration of these limitations, future research should focus on implementing the treatment with a larger sample size of patients with HH (both parahypothalamic and intrahypothalamic hamartomas). However, given the low base rates of this condition, continued publication of case studies related to these conditions and other rare pediatric conditions may be most appropriate.

Further, despite parent-reported improvements in behavior via weekly behavior logs and pre- and post-test behavioral measures (e.g., BASC-3, ECBI, and DBRS), the patient's behavioral progress was measured by parent report only. The clinicians did not collect in-session frequency data related to parent or child behaviors, nor did they collect treatment integrity data related to parent implementation of the strategies. Having this in-session data would have strengthened the current study by allowing clinicians to collect inter-rater reliability data, measure progress across sessions in an objective manner rather than relying on parent report only, compare data collected by clinicians to that of the pre- and post-test behavioral measures as well as potentially allow the clinicians to assess whether a specific strategy led to the greatest behavioral change. Future studies related to this population and the BBI program should consider the inclusion of in-session data collection.

Although the BBI program has shown to be effective with neurotypical children (Axelrad et al., 2009, 2013), it has not yet been validated across pediatric populations. This study offers a natural stepping-stone for future research to demonstrate the effectiveness and validity of the BBI program across pediatric populations. Further, when compared to other PMT programs available, BBI is a feasible option for clinicians working with pediatric populations given that the program is manualized, inexpensive, and easy to disseminate. Although the program is manualized, it offers clinicians the flexibility to provide individualization of the core

components of the program as needed in both inpatient and outpatient settings with cultural and developmental modifications as needed.

Compliance with Ethical Standards

Conflict of interest The authors Rachel H. Fein, Gabrielle G. Banks, Marsha N. Gragert, and Marni E. Axelrad declare that they have no conflict of interest.

Ethical Approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed Consent Informed consent was obtained from all individual participants included in the study.

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