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Impact of benign childhood epilepsy with centrotemporal spikes (BECTS) on school performance

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ABSTRACT

BECTS represents the vast majority of childhood focal epilepsy. Owing to the age peculiarity of children who suffer from this disease, i.e., school-going age of between 6 and 9 years, the condition is often referred to as a school disorder by parents and teachers.

Objective: The aim of this study was to evaluate the academic performance of children with BECTS, according to the clinical and electroencephalographic ILAE criteria, and compare the results of neuropsychological tests of language and attention to the frequency of epileptic discharges.

Methods: The performances of 40 school children with BECTS were evaluated by applying a school performance test (SBT), neuropsychological tests (WISC and Trail-Making), and language tests (Illinois Test Psycholinguistic Abilities – ITPA – and Staggered Spondaic Word – SSW). The same tests were applied in the control group.

Results: Children with BECTS, when compared to those in the control group, showed lower scores in academic performance (SPT), digits and similarities subtests of WISC, auditory processing subtest of SSW, and ITPA – representational and automatic level. The study showed that epileptic discharges did not influence the results.

Conclusion: Children with BECTS scored significantly lower scores in tests on academic performance, when compared with those in the control group probably due to executive dysfunction.

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1. Introduction

Benign epilepsy with centrotemporal spikes (BECTS) is the most common focal epilepsy of childhood, representing between 15 and 20% of epilepsies in children between 5 and 14 years of age.¹ The prevalence of BECTS is estimated to be about 2% in children, and it is four times more common than typical absence of epilepsy.² According to the ILAE, BECTS is classified as idiopathic focal epilepsy, and its underlying structural causes and possible genetic aetiology are unknown.³ Studies of families with children with BECTS suggest that the characteristic electroencephalography is

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transmitted in an autosomal dominant fashion with age-dependent penetrance.^{4–6} BECTS is characterized by seizures involving one side of the face with motor and/or sensitive components, of short duration, without impairment of consciousness, and sometimes with secondary generalization. In most cases, the seizures are related to sleep. Three-quarters of the insults occur during non-REM sleep, i.e., at the beginning of sleep or near waking.⁷ The onset of epileptic events occurs between 3 and 13 years of age (peaking between 6 and 9 years), and spontaneous remission occurs in adolescence. It is much more common in males. The characteristic electroencephalographic pattern features a spike or sharp waves of large amplitude, followed by a slow wave, projecting from the centrotemporal regions. These discharges are activated by drowsiness or sleep and spread through the brain. According to some authors, the origin of this epileptiform activity is in the lower Rolandic cortex, and BECTS is sometimes referred to as benign Rolandic epilepsy. The somatotopic distribution of the discharges extends along the central sulcus. The cerebral cortex,

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which is responsible for the motor and sensory function of the arms, face and tongue, is located sequentially from rostro-medially to ventro-laterally along the central sulcus.^{8–10}

Patients with epilepsy are at risk for learning disorders, and most require special educational support. Multiple factors are associated with the low academic performance of children with epilepsy, including some characteristics of seizures that act as major generators of cognitive disorders. These factors are as follows:

- (a) seizure frequency and duration;
- (b) duration of the disease;
- (c) age at the onset of seizures;
- (d) aetiology of the seizure;
- (e) seizure type(s);
- (f) use of anti-epileptic drugs (AEDs);
- (g) psychosocial problems (stigma); and
- (h) genetic factors.^{11–17}

BECTS is a good experimental model because it represents a homogeneous group of patients with age-dependent focal seizures and cortical excitability that is not associated with structural damage.¹⁸ This disease profile allows researchers to isolate the effect of focal epileptiform activity on cognitive functions, which is important because neuropsychological abnormalities may be associated with the quantity [Gobbi Metz^{19,20}] and location of epileptic discharge. Another important consideration is that patients do not always receive AEDs [Shields²¹], which could confound the results of cognitive tests. The aim of this study was to assess the academic skills of patients with BECTS and to compare their results with those of a control group of children matched with respect to sex, age, education, and socio-economic status.

2. Methods

Forty children were recruited from our institution's outpatient service for child neurology. All patients were diagnosed with typical BECTS according to the clinical and electroencephalographic criteria adopted by the ILAE (Commission on Classification and Terminology of the International League against Epilepsy 1989).³ The patients were both male and female and were between 7 and 13 years of age. These children were observed to have had at least two seizures, with normal neurological examination results, no maternal problems during pregnancy or labour, and normal brain magnetic resonance imaging (MRI) or computed tomography (CT) scans. Twenty-two of the children had been medicated with anti-epileptic drugs during the assessments; six of them received sulthiame, 14 received carbamazepine/oxcarbazepine, two received sodium valproate, and one received phenobarbital. Children who had atypical EEG (electroencephalogram) manifestations and thus did not match the ILAE classification were excluded from the test group. A matched control group (sex, age, and school level) was recruited from children in governmental and private schools in Sao Paulo; controls had no neurological abnormalities and no personal or family history of epilepsy. The numbers of children in the control group varied with the test applied and were guided by statistical criteria. The school questionnaire, EEG, and neuropsychological evaluation assessment were all applied within a maximum interval of thirty days. The time between the EEG and neuropsychological tests did not exceed seven days.

2.1. Academic achievement

The school questionnaire evaluated the following areas of academic achievement: motor coordination, manual motor skills, concentration and memory, speech and learning, writing, reading, calculation, and behaviour. This information was obtained through interviews with the children's parents and through the SPT, which is a psychometric instrument for assessing ability essential for academic performance. The SPT consists of three subtests:

- (a) Writing subtest: This test begins with the writing of the child's name, followed by the writing of 34 isolated words dictated by the examiner. Each correctly written word receives one point, and the best possible gross score is a 35.
- (b) Arithmetic subtest: This test comprises both oral and written sections. The oral section includes three questions, and the written section includes 35 mathematical calculations that gradually increase in difficulty. Each correct answer receives one point, and the highest possible score is 38 gross points.
- (c) Reading subtest: This test is comprised of 70 words in Portuguese, which the children are asked to read aloud. The maximum score in this subtest is 70 points. The purpose of this subtest is to evaluate the ability of children to decode words on their own, independent of context.

The total score is obtained by adding the scores of the three subtests and is then classified according to the age and educational level of the child. The same battery of tests was administered to the 40 children in the control group, who were matched according to age, sex, level of education, and level of maternal education.

2.2. Neuropsychological variables

Several metrics were used to score neuropsychological variables. The Trail-Making test was used to assess divided attention and concentration. Several WISC subtest were used (comprehension, similarities, digit span block design m, mazes and coding). The assessment of language by the Illinois Test of Psycholinguistic Abilities (ITPA). The SSW dichotic listening test subtests on auditory processing were administered to evaluate the recognition of overlapped sounds presented as dichotic, according to Katz.²² The scores represent the number of errors in non-competitive and competitive conditions, and were categorized as ear effect (high-low, low-high), order effect (high-low, low-high), reversal, or type-A pattern.

2.3. EEG

All patients underwent forward regular electroencephalography. Records were obtained after sleep deprivation during drowsiness, sleeping, and wakefulness. In cases where it was not possible to count the number of epileptic discharges or where the EEG taken at the time of the application of the tests was normal, previously recorded EEGs were used to determine the location of the discharge. The EEGs were recorded using Berger[®] appliances, with a system of 8 (TP model 119 and ED 121), 16 (Polygraph TW-102), or 21 channels, in the EEG unit of the Department of Pediatric Neurology at the Hospital das Clínicas Faculty of Medicine, University of São Paulo, Brazil. The electrodes were distributed according to the 10-20 international system, with additional electrodes in the centrotemporal region distributed according to the 10-10 international system, mounted in a bipolar arrangement. Benchmarks were taken in the inter-critic period during the stages of drowsiness, sleep, and wakefulness, with hyperphoea and intermittent photic stimulation. The EEG signal was evaluated using longitudinal bipolar and A1 + A2 referential montages. The location of the epileptic discharge was determined as the region with the most electronegativity in the average mounting. In the second EGG, discharges were counted for 5 min, and patients were divided into two groups based on the number of discharges (more than or fewer than 10 discharges/5 min).

2.4. Statistical approach and significance test

The collected data were analysed using SPSS version 10.0 for Windows (Prentice Hall, Chicago, USA). We used Student's *t*-test and χ^2 to compare the patient groups. The level of significance adopted was 95% (p < 0.05).

3. Results

We evaluated 40 children (16 females and 24 males) between 7 and 13 years of age (average age = 8 years). A total of 353 crises were analysed. The average number of crises per child was 9, with a range from two (minimum) to 30 seizures (maximum). The principal characteristics of the children in this study and their clinical manifestations are presented in Tables 1 and 2. The average interval between the first and last seizure was four years (ranging from 7 days to 8.5 years). Seizures were simple partial in 77% of the children.

3.1. Academic achievement

The SPT was used to measure academic achievement. Ten children were unable to complete the tests due to significant difficulty in school learning. On the writing subtest, nine children were scored classified as inferior, six as superior, and 15 as average. On the arithmetic subtest, 14 children were classified as inferior, ten as average, and six as superior. On the reading subtest, 13 children were classified as inferior, nine as average, and eight as superior. On the overall SPT test score, i.e., the total of the three subtests, three children were classified as superior, 11 as average, and 16 as inferior. The BECTS group had a significantly lower mean SPT score than that of the control group (p = 0.008) (Table 3). Reports from parents and teachers about learning disorders in

Table 1

Characteristics of patients according to age and total number of seizures.

	Number	Average
Age at the first seizure (year)	2.5-11.83	6.66
Age during study	8.33-15	11.68
Total seizures analyzed (353)	2-30 ^a	8.83ª

^a Average attacks per child from the beginning of epilepsy.

Table 2

Main characteristics of epileptic seizures.

Clinical manifestation	%
Arrest speech	65.39
Drooling	59.68
Lip lateralization	33.65
Ocular fixation	21.89
Throat noises	17.76
Clonic movement of lips	17.77
Stiffness of the tongue	15.23
Todd paralysis	8.57
Clonic movements of the tongue	8.25

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Table 3

Results of SPT.

	Study group		Control group	
	Female (%)	Male (%)	Female (%)	Male (%)
Higher	03 (18.75)	0	06 (25)	09 (52.94)
Average	04 (25)	7 (29.17)	10 (41.67)	02 (11.76)
Lower	09 (56.25)	17 (70.84)	08 (33.33)	06 (35.30)
Total	16 (100)	24 (100)	24 (100)	17 (100)

SPT, school performance test.

school were consistent with the low SPT scores (p = 0.004 and p = 0.008, respectively). However, there was no significant correlation between SPT score and

- (a) number of epileptic discharges (more or fewer than 10) recorded in a 5-min EEG (p = 0.484); i.e., the students with the greatest number of epileptic discharges did not have the worst test scores;
- (b) laterality of epileptic discharge (p = 0.756);
- (c) Sex of the child (p = 0.345);
- (d) AED use (*p* = 0.547);
- (e) timing of seizure occurrence (during sleep, p = 0.768; awake, p = 0.942);
- (f) number of seizures (p = 0.484); and
- (g) child's age at the time of first seizure (p = 0.256).

The grade level of the children of the study group ranged from elementary school to secondary school. Parents and teachers recognized academic underachievement in 55% (22 children) and 70% (28 children) of the children, respectively. The level of maternal education ranged from 1 to 15 years of study; 12 mothers (30%) had studied for fewer than 4 years, 15 (37.5%) had studied for 4–8 years, and 4 (10%) had studied for over 11 years. Only three mothers had a university level education. The children with BECTS whose mothers had higher levels of education demonstrated better outcomes than those whose mothers had lower levels of education (p = 0.004). We could not demonstrate a relationship between maternal education and school performance for children in the control group.

3.2. Neuropsychological variables

The Trail-Making test was administered to the 31 children who were able to read and write; 20 (64.5%) of them returned normal results with no statistical correlation with school performance.

The aim of the neuropsychological evaluation was to estimate IQ and rule out mental retardation. None of the children included in the study were found to be mentally retarded. The children with BECTS showed the worst results in the digit span test, followed by the similarities test. We observed a statistically significant correlation between low scores (digit span and similarities) and poor results on the digits subtest of the SPT (p = 0.001). The children with BECTS also had difficulties on the similarities test, with a possibly significant correlation with the SPT (p = 0.043).

3.3. Language assessment

The results of the language assessment tests suggested greater impairment of the auditory–vocal representational level (47.5%) than the auditory–vocal automatic (42.5%) level, with preservation of the visuomotor representational and visuomotor automatic channels in the vast majority of the children. There was a statistically significant correlation between low scores on ITPA subtests and unsatisfactory SPT results (p < 0.001), as shown in Table 4.

3.4. SSW in Portuguese

The SSW subtest was carried out on 40 children with BECTS, and 29 children (72.5%) had abnormal results. Abnormal results on the SSW test were linked with low SPT scores (p = 0.013). There was a statistically significant difference between the scores of children with BECTS and children in the control group (p = 0.0013); however, SSW results were not correlated with the number of epileptic discharges (p = 0.412). No differences between the two

Table 4

Comparison between SPT vs. ITPA.

ITPA subtests	Significance (p)
Scalar score average	<0.001
Auditory association	0.001
Visual reception	0.039
Visual association	0.006
Auditory sequential memory	0.017
Grammatical closure	0.008
Visual sequential memory	0.042

SPT, school performance test.

groups (BECTS vs. control group) were observed when applying the data measured by Katz or those obtained from the Brazilian study. The values obtained in the study were compared to the Katz pattern, which is identical to the Brazilian standard.

3.5. EEG

All EEGs showed normal background activity. The first EEG was conducted using an 8-channelEEG in one child and a 16-channel EEG in the other children; a 21-channel EEG was used for the second EEG in all children. In the first EEG, the spike and wave discharges were bilateral in 16 children, on the right in 14 children and on the left in 10 children. At the time of neuropsychological evaluation (second EEG), 7 EEGs were normal (17.5%), 12 had bilateral discharges, 11 had discharges on the right, and 10 had discharges on the left. In 17 children, the number of discharges exceeded 10 per 5 min. The children with high-frequency epileptic discharges (>10 discharges/5 min) showed no statistically significant differences from children with low-frequency discharges on the SPT, WISC, or SSW tests. However, a significant between-group difference was observed in the auditory association subtest of the ITPA (p = 0.008). The laterality of the discharges did not affect the results of any neuropsychological test.

4. Discussion

Although BECTS has a good prognosis,²³ as noted by Gündüz et al.,²⁴ many children exhibit symptoms of cognitive disorders during the course of epilepsy and are very frequently reported by parents and teachers as unable to learn appropriately. Our study confirmed a higher incidence of learning disorders in children with BECTS than in a matched control group. These results are similar to those reported by Fonseca,²⁵ also in Brazil. We also found that the level of mother's education was a determining factor of low SPT performance. Children with BECTS and learning disabilities had mothers with lower education levels, suggesting that there may be a close relationship between low stimulation and greater learning difficulties due to BECTS.

Some studies^{26–29} have linked the laterality of the epileptic focus with different cognitive disorders. In our study, the laterality of the epileptic discharge was not correlated with any neuropsy-chological test results. Our results also did not support a relationship between the number of epileptic discharges and neuropsychological scores, although this effect has been observed in other studies.^{30,31}

We found no statistically relevant relationships between low SPT scores and the type of seizure or the frequency or timing of the seizures. Treatment also did not seem to influence the results of the SPT. These observations can be explained by the many cognitive changes that are frequently associated with seizure characteristics and treatment.

Another relevant finding is the frequency of hearing disabilities with the concomitant preservation of visual attention in our patients, as demonstrated by the ITPA. Many studies cite attentional disorders in seizure patients, but do not emphasize the predominance of auditory disorders.^{32–35} Our results suggest that central visual functions are mostly preserved while auditory function is impaired. These findings may underlie the learning disabilities in children with epilepsy. Weglage et al.,³⁶ after evaluating psycholinguistic development through a German version of the ITPA, detected no significant difference between children with BECTS and controls. Meanwhile, Staden et al. studied 20 children with BECTS and found that 13 of them had problems in reading, auditory-verbal learning, hearing discrimination with background noise (words), and grammar.³⁰ Our results confirm the presence of attention and language disorders in children with BECTS.

The patients with BECTS in this study had poor results on the digit span subtest, which reflects several different mental activities (memory, comprehension, and associative ability) that may involve the right frontal region, as described by Metz-Lutz and Filippini.²⁰ BECTS patients also exhibited low scores on the similarities subtest, indicating impairment in verbal abstract reasoning, verbal categories, and conceptual understanding.²⁰

Our group of patients with epilepsy also had poor scores on the SSW subtest. The SSW test is a dichotic listening test that is frequently used to evaluate auditory perceptual abilities in children with language and learning problems.²² The abnormalities observed in the group were related to auditory memory, particularly the low/ high-ear effect and high-/low-order effect disorders. These disorders could affect the production of verbal expression. Many of these children also had difficulty in the analysis and synthesis of hearing and phonemic decoding, i.e., receptive language dysfunction. Other studies of children with epilepsy have shown a high prevalence of central auditory processing disorders using the staggered spondaic word (SSW) test.²²

The main finding of our study is that children with BECTS are more likely to have learning disorders than are children without epilepsy, principally as a consequence of auditory attention disorder. Diminished auditory skills were observed in children with BECTS, while visual ability was largely normal. These results suggest that central visual functions are preserved, and auditory function is impaired. These findings may underlie the prevalence of learning disabilities in children with epilepsy. The interdependence of listening, language, and learning should be the basis for the diagnosis of auditory processing deficits.³⁷

Various auditory and language tasks are used to measure children's auditory processing abilities. However, it has been suggested that these measures may actually assess language, rather than auditory processing. In addition, recent studies have suggested that the tasks used to assess auditory processing may, in fact, reflect attention instead. According to Metz-Lutz and Filippini,²⁰ recovery from epilepsy was accompanied by improvement in executive functions, particularly those related to sustained attention and behavioural control. The circadian variation of intensity of epileptic activity should also be analysed.³⁸ We cannot be sure that the patients in our study will show similar improvement, as this would require a reassessment of the children after the cessation of epileptic discharges. However, all of the observed abnormalities could be consequences of an underlying disorder of cerebral maturation, probably of genetic origin,³⁹ and mainly involving the perisylvian region.

5. Conclusions

We conclude that children with BECTS have educational impairment when compared with their peers (matched with respect to sex, age, and level of education) who do not have epilepsy. These children may have a greater chance of developing language organization and auditory attention disorders. This study demonstrated cognitive impairment (e.g., intelligence, memory, language, and attention) in BECTS patients with language disorders being most severe. These analyses suggest that the concept of BECTS as "benign" refers only to the resolution of epileptic seizures and changes in the electroencephalographic pattern; the possibility of cognitive abnormalities should always be considered.

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