Seizure 57 (2018) 27–31

Contents lists available at ScienceDirect

Seizure

journal homepage: www.elsevier.com/locate/yseiz

Benign rolandic epilepsy and generalized paroxysms: A study of 13 patients

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ARTICLE INFO

Article history:
Received 13 December 2017
Received in revised form 20 February 2018
Accepted 5 March 2018
Available online xxx

Keywords:
Benign
Centrotemporal spikes
Rolandic epilepsy
Generalized paroxysms
Spike and waves

ABSTRACT

Purpose: To present a retrospective study of 13 children with benign epilepsy with centrotemporal spikes (BECTS), also known as benign rolandic epilepsy (BRE), associated with generalized spikes and waves as the only EEG manifestation at onset.
Method: Charts of children with typical clinical criteria of BRE electroclinically followed-up between February 2000 and February 2015 were reviewed.
Results: Among 309 patients who met the electroclinical criteria of BRE, we identified 13 children who presented with the typical clinical manifestations but who, on the EEG, only had generalized paroxysms at onset that continued along the course of the syndrome. Generalized spike-and-wave discharges were observed in all patients when awake and during sleep (100%). During the evolution no particular electroclinical pattern was observed. The patients responded well to antiepileptic drugs, such as valproic acid and levetiracetam. Outcome was good in all patients.
Conclusions: We found evidence that patients with BRE may have generalized EEG discharges at onset as the sole manifestation lasting throughout the course of the syndrome. In some, focal paroxysms developed later. The course was benign. In our group of patients, clinical features and evolution were similar to those of typical cases of BRE. Response to valproic acid and levetiracetam was found to be particularly good.

1. Introduction

Benign rolandic epilepsy (BRE) also known as benign epilepsy with centro-temporal spikes (BECTS) is a focal epilepsy of childhood characterized by motor focal seizures and particular EEG abnormalities in the absence of neurological deficits [1]. BRE is the most common form of idiopathic epilepsy in children [2]. In spite of the concept that the qualification "benign" should be avoided in the milder epilepsy syndromes such as BRE, in this study we have kept this term. [3]. In the recent ILAE Classification proposal the term "idiopathic" has been replaced by "genetic" [3]; however, as the genetic marker in patients with BRE is not well known, it could be considered as probably genetic. Here, we have also maintained the terms "idiopathic localization-related epilepsy" and "idiopathic generalized epilepsy (IGE)".

In BRE the seizures are usually infrequent, brief, and partial, paradoxically showing abundant interictal EEG abnormalities, and remit spontaneously before the end of adolescence [4]. The seizures are somatosensory and motor focal, mainly affecting the face and oropharynx, with speech arrest and hypersalivation and in some cases involving the upper limbs [5]. The typical EEG shows high-voltage spikes or spikes and waves in the centrotemporal region that may shift from side to side with a normal background.

In BRE patients, generalized spike-wave discharges are rather common and brief. Generalized 3–5-Hz slow-wave bursts intermixed with small spikes, with or without clinical seizures, were reported to occur in 4% of the cases [6,7].

BRE is easily diagnosed based on EEG features. Generalized spike discharges, also observed in childhood absence epilepsy (CAE) or IGE, were found to be appearing with or without seizures during follow-up periods. Considering the benign, age-related, and age-limited features of idiopathic localization-related epilepsies and IGEs, this finding suggests a neurobiological continuum between the two epilepsies [8].

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https://doi.org/10.1016/j.seizure.2018.03.004
1059-1311/

The aim of this study was to describe BRE patients in whom EEG findings were generalized spikes and waves as the only manifestation at onset.

2. Methods

Between February 2000 and February 2015, we evaluated 309 patients with BRE at the department of neurology of our hospital; 13 of these patients only had generalized spike-and-wave paroxysms at onset.

The following inclusion criteria for BRE were used:

a Typical sensorimotor seizures affecting the orofacial district with or without generalization
b Normal pregnancy and delivery
c Uneventful past medical history
d Normal psychomotor development at diagnosis
e Normal brain imaging
f Generalized spike-and-wave paroxysms as the sole manifestation at onset
g Patients with typical electroclinical features of BRE who during follow-up presented with electroclinical features of other idiopathic focal epilepsies of childhood were also included

Patients with centro-temporal spikes alone or combined with generalized paroxysms at onset, or those with a normal EEG, with known mental or neurological deficits, or with typical electroclinical features of Panayiotopoulos (PS) or Gastaut-type childhood epilepsy with occipital paroxysms (CEOP) at onset were excluded.

We analyzed sex, age at onset, personal and family history of epilepsy, seizure duration and manifestations, circadian distribution, seizure frequency, response to therapy, and outcome.

EEGs were performed while awake and asleep. Electrodes were placed according to the international 10–20 system. Brain CT scan and MRI were performed in all patients.

Between February 2000 and February 2015, 13 patients who met the inclusion criteria of BRE were identified and have been followed up to the present time. All patients were evaluated longitudinally, clinically, and with EEGs for 2–8 years (mean: 4.5 years). A mean of 8 ± 3 EEGs were obtained for each patient. We evaluated the charts reviewing clinical and EEG details of all patients that were unanimously agreed upon by all authors.

3. Results

3.1. Number of patients and gender

Overall, 13 children, 8 boys and 5 girls, met the inclusion criteria of idiopathic BRE over a 15-year period between February 2000 and February 2015.

3.2. Age at onset

Age at first afebrile seizure ranged from 5 to 11.5 years, with a mean age of 6.5 and a median of 7 years.

3.3. Personal and family history of febrile seizures and epilepsy

A family history of epilepsy was found in five cases (38%). Febrile seizures were reported in two (31%) and migraine in one (11%).

3.4. Ictal manifestations

Oropharyngeal manifestations were observed in seven children (54%), unilateral facial sensorimotor seizures in four (31%), speech arrest in six (46%), and hypersalivation in seven (54%). Hemiclonic focal seizures were observed in three patients (23%). Six children had focal seizures to generalized seizures (46%). Consciousness

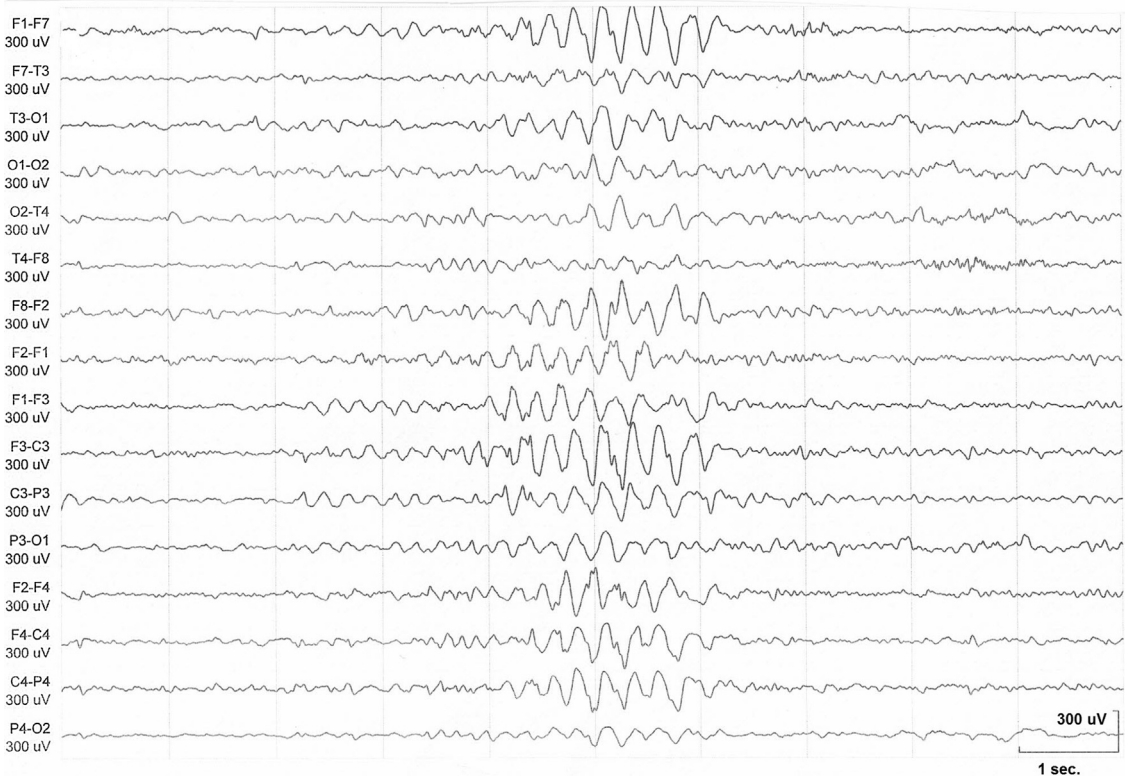

Fig. 1. The interictal EEG recording during sleep shows generalized spike-and-wave paroxysms predominantly in the anterior regions in a 7-year-old boy. Focal abnormality in the left frontal region preceding the generalized paroxysms is evident.