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LETTER TO THE EDITOR

Consecutive occurrence of benign epilepsy with centro-temporal spike and childhood absence epilepsy: true coexistence or atypical evolution?

Eun Hye Lee[1] · **Hoi Soo Yoon**[1]

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We read with great interest the recent review of benign epilepsy with centro-temporal spikes (BECTS) and childhood absence epilepsy (CAE) by Verrotti et al. [1]. BECTS and CAE are the most common epilepsy syndromes in childhood and share certain clinical features: similar age at onset, overall good prognosis, and genetic predisposition. Several studies have reported the coexistence of the two syndromes, and recently, Verrotti et al. analyzed 19 cases reported to have clinical features of CAE and BECTS. Among the total 19 cases, three cases had features of both syndromes contemporarily; three cases experienced absence seizures initially followed by BECTS; and the other 13 cases experienced BECTS at onset and had absence seizures later.

Here, we would like to consider the patients who presented with BECTS at onset and absence seizures later. Of the 13 patients, seven had typical or simple absence seizures and the other six had atypical absence seizures, atonic absences, absence seizure with fluttering, or eyelid myoclonia. According to inclusion and exclusion criteria for CAE proposed by Panayiotopoulos, these feature are not compatible with CAE [2]. Atypical absence seizures are differentiated from absence seizures in the frequency of spikes and wave discharges, refractoriness to medical therapy, and severely abnormal cognitive and neurodevelopmental outcome. Both typical and atypical absence epilepsy are thought to be generated by a rhythmogenic interplay between the cortex and the thalamus. However, it is hypothesized that atypical absence seizures engage different neuronal networks within the thalamo-cortical circuitry, as compared to typical absence seizures [3].

In some patients with BECTS, atypical evolution occur during the natural course or medical treatment. Atypical features at onset, such as frequent spikes or spike-wave discharges, or early age at onset, are considered risk factors for, and atypical evolution of, BECTS [4].

We suggest that some of the 13 patients with BECTS who later developed CAE might be examples of atypical evolution of BECTS rather than a true coincidence of two syndromes. Notably, three of 13 patients have experienced atypical absence seizures after the administration of antiepileptic drugs, which improved after drug withdrawal. Several authors have reported children with BECTS who developed CAE after admiration of antiepileptic drugs such as oxcarbazepine, phenobarbital, and lamotrigine [5–7]. This implies that the evolution to atypical BECTS was triggered by certain antiepileptic drugs in prone subjects. Similarly, five of the six patients in Dimova et al. were treated with carbamazepine, and they also suggested the possibility of aggravation of BECTS by the drugs [8].

Obviously, it is possible CAE and BECTS can simply coexist. The three patients with contemporary onset of CAE and BECTS in this review are such cases, in whom neurocognitive function is normal and they have good responses to drugs. However, when CAE and BECTS occur consecutively in one patient, they should be distinguished from atypical evolution of BECTS. Especially in these cases, the effect of antiepileptic drug should be considered.

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✉ Eun Hye Lee
leeeh80@khmc.or.kr

[1] Department of Pediatrics, College of Medicine, Kyung Hee University, Seoul, Korea

Compliance with ethical standards

Ethical approval Not needed.

Conflict of interest The authors have no conflicts of interest relevant to this article.

References

1. Verrotti A, D'Alonzo R, Rinaldi VE, Casciato S, D'Aniello A, Di Gennaro G. Childhood absence epilepsy and benign epilepsy with centro-temporal spikes: a narrative review analysis. World J Pediatr. 2017;13:106–11.
2. Panayiotopoulos CP. Absence epilepsies. In: Engel J Jr, Pedley TA, editors. Epilepsy: a comprehensive textbook. Philadelphia: Lippincott-Raven; 1997. p. 2327–46.
3. Onat FY, van Luijtelaar G, Nehlig A, Snead OC 3rd. The involvement of limbic structures in typical and atypical absence epilepsy. Epilepsy Res. 2013;103:111–23.
4. Fejerman N. Atypical rolandic epilepsy. Epilepsia. 2009;50:9–12.
5. Grosso S, Balestri M, Di Bartolo RM, Corbini L, Vatti G, Curatolo P, et al. Oxcarbazepine and atypical evolution of benign idiopathic focal epilepsy of childhood. Eur J Neurol. 2006;13:1142–5.
6. Hamano S, Mochizuki M, Morikawa T. Phenobarbital-induced atypical absence seizure in benign childhood epilepsy with centrotemporal spikes. Seizure. 2002;11:201–4.
7. Catania S, Cross H, de Sousa C, Boyd S. Paradoxic reaction to lamotrigine in a child with benign focal epilepsy of childhood with centrotemporal spikes. Epilepsia. 1999;40:1657–60.
8. Dimova PS, Daskalov DS. Coincidence of rolandic and absence features: rare, but not impossible. J Child Neurol. 2002;17:838–46.