てんかんをめぐって (1996.02) XVI巻:94～98.

日本人少年の読書てんかん例の発作中HMPAO-SPECT所見

Miyamoto Akie, Takahashi Satoru, Tokumitsu Aya, Oki Junichi

Ictal HMPAO-SPECT Findings in Reading Epilepsy in a Japanese Boy.

Akie Miyamoto Satoru Takahashi Aya Tokumitsu and Junichi Oki

Department of Pediatrics, Asahikawa Medical College

Key words ; reading epilepsy, Ictal, SPECT, EEG

Reading epilepsy was first described by Bickford et al. in 1956 [1]. It is included as an idiopathic localization-related epilepsy in the International Classification of Epilepsies and Epileptic syndromes [2].

Wolf performed a detailed review of 111 cases of reading epilepsy, but the physiopathology remains unknown [3]. Ictal EEG studies of reading epilepsy have shown maximal involvement of the temporoparietal region of the language-dominant hemisphere. We know of no reports describing findings on regarding reading epilepsy on single photon emission computed tomography (SPECT). We studied ictal and interictal SPECT using [^{99m}Tc] hexamethylpropylene amine oxime (HMPAO) in a Japanese boy with reading epilepsy to clarify the region of brain involvement.

Case report

The patient was a 14-year-old right-handed Japanese boy. There was no family history of epilepsy. The patient's medical history was remarkable only for mild bronchial asthma since age 10 years. At age 12 years, when he was in the first year of junior high school, he experienced jaw jerking while silently reading a textbook. This occurred every time he read an English textbook and sometimes during prolonged reading of a Japanese textbook. However, seizures never occurred while reading Japanese language textbooks of mathematics or science. He learned to control the jerking by ceasing reading. He said that he realized meanings of sentences by looking rather than reading. At the age of 13, the jaw jerking evolved to generalized tonic-clonic seizures (GTCS) on two occasions during prolonged reading aloud of a Japanese textbook. Baseline EEGs were normal,

including during hyperventilation, photic stimulation and sleep. After reading an English textbook for 1 minute, he began to have typical jaw jerking associated with bilateral 2-Hz, 150-μV spike-wave complexes with left frontotemporal accentuation on the EEG (Fig 1). Paroxysmal discharges disappeared as soon as he ceased reading. WISC-R I. Q. was 107 (verbal I.Q. 111, performance I.Q. 102). Magnetic resonance images (MRI) was normal. Ictal SPECT with HMPAO was perfomed. He began reading an English textbook silently, monitored by EEG. Five minutes after jaw jerking began, HMPAO 370 MBq was administered intravenously and he continued reading for 2 min more. The corresponding SPECT scan showed focal hyperperfusion in both frontal lobes and in the left temporal lobe (Fig 2). The hyperperfusion was especially notable in the left frontal area. Interictal HMPAO SPECT was normal. In a normal volunteer, a 25-year-old right-handed woman, HMPAO SPECT during reading of an English textbook for 7 min showed slight hyperperfusion in the left frontotemporal area and no abnormality at rest.

Fig. 1 Ictal EEG after 1 min of reading an English textbook : 2-Hz, 150-μV spike-wave complexes with left frontoal accentuation (arrowhead) are accompanied by jaw jerking at the beginning (arrow). EMG recorded jaw muscle activity.

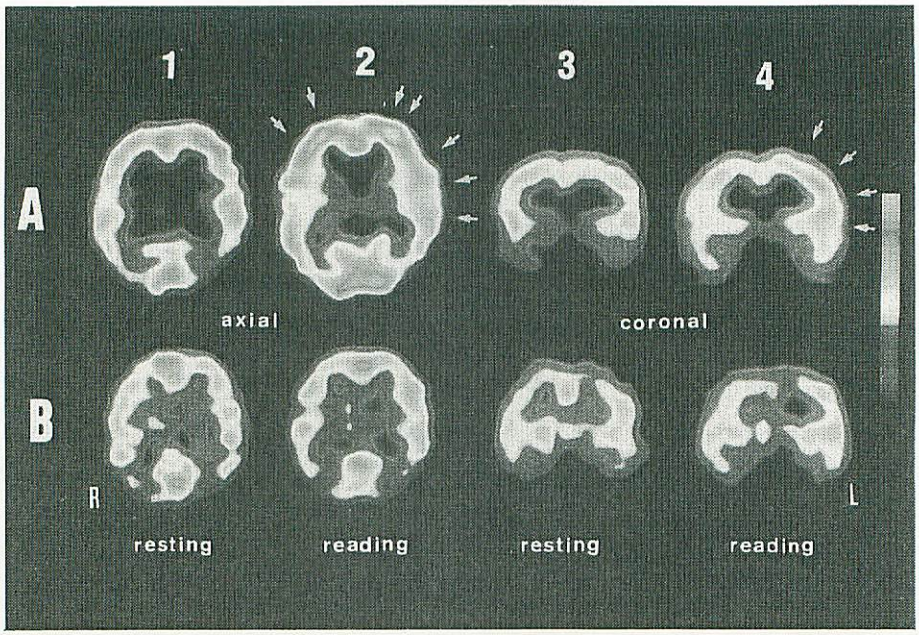

Fig. 2 [^{99m}Tc] HMPAO SPECT findings. The patient A : Axial view at rest (A1) and ictal (A2) ; coronal view at rest (A3) and ictal (A4). SPECT findings at rest showed normal. Ictal SPECT showed focal hyperperfusion in both frontal lobes and in the left temporal area (arrows).

A normal volunteer B : Axial view at rest (B1) and during reading (B2), coronal view at rest (B3) and during reading (B4). SPECT at rest showed normal and SPECT during 7 min of reading showed slight hyperperfusion in the left frontotemporal area.

After the ictal SPECT and EEG examination, we treated our patient with a combination of valproate (VPA) and clonazepam (CZP), which almost eliminated the frequency of jaw jerking episodes. There were no further generalized tonic-clonic seizures.

Discussion

Wolf reviewed the ictal EEGs of 73 cases of reading epilepsy and reported the most common type of discharge was bilateral with unilateral temporoparietal accentuation on the side of the language-dominant hemisphere [3]. The ictal EEG in our patient showed bilateral spike-wave complexes with left frontotemporal predominance. Furthermore, ictal SPECT in our patient demonstrated hyperperfusion in the same region. As compared with SPECT findings in the control, the area of hyperperfusion in the patient was more intense and larger (Fig 2). SPECT studies with [^{99m}Tc] HMPAO or

HIPDM in the ictal or immediate postictal state can be a practical and reliable method of seizure localization in partial epilepsies [4,5,6]. In our patient, the focus of reading epilepsy appeared to be in the frontotemporal region within the language-dominant hemisphere.

In Wolf's review of 111 patients with reading epilepsy, only 8 were Japanese [3]. Our patient had onset of reflex seizures when he began studying English in the first year of junior high school in Japan, and reading English provoked his seizures more than reading Japanese. The difficulty for most Japanese of reading the English material is believed to have a greater provocative effect [8]. However, reading a difficult textbook of mathematics or science in Japanese did not evoke seizures in our patient. Therefore, another factor may cause Japanese language processing to differ greatly from that of English [6]. Wolf suggested that the Japanese language, which is written in a combination of ideographic (Kanji) and phonic (Kana) systems, could aid in understanding the basis of reading epilepsy [3]. In 1 Japanese patient, Masaoka et al. [9] showed that nonsense Kana were more provocative than meaningful Kana, just as is an alphabetical nonsense text. We speculate that reading epilepsy is triggered by neuronal processes involved in the elaboration of language.

Acknowledgements

We thank Professor Akimasa Okuno for review of the manuscript and Yukio Ishikawa and Junichi Satou for technical assistant with SPECT work.

References

1. Bickford RG, Whelan JL, Klass DW, Corbin KB. Reading epilepsy : clinical and electroencephalographic studies of a new syndrome. Trans Am Neurol Ass 1956 ; 81 : 100 – 2.
2. Commission on Classification and Terminology of the International League against Epilepsy : Proposal for revised classification of epilepsies and epileptic syndromes. Epilepsia 1989 ; 30 : 389 – 99.
3. Wolf P. Reading epilepsy. In : Roger J, Bureau M, Dravet Ch, Dreifuss FE, Perret A, Wolf P eds. Epileptic syndromes in infancy, childhood and adolescence (2nd ed). London : John Libbey and Company, 1992 : 281 – 98.

4. Adams C, Hwang PA, Gilday DL, et al. Comparison of SPECT, EEG, CT, MRI, and pathology in partial epilepsy. Pediatr Neurol 1992 ; 8 : 97 – 103.
5. Lee BI, Markand ON, Wellman HN, et al. HIPDM-SPECT in patients with medically intractable complex partial seizures : ictal study. Arch Neurol 1988 ; 45 : 397 – 402.
6. Jones EA, Aoki C. The processing of Japanese Kana and Kanji Characters.In : de Kerckhove D, Lumsden ChJ, eds. The alphabet and the brain, Berlin, Heidergerg : Springer, 1988 : 301 – 20.
7. Rowe CC, Berkovic SF, Sia STB, et al. Localization of epileptic foci with postictal single photon emission computed tomography.Ann Neurol 1989 ; 26 : 660 – 8.
8. Stella L, Fels A, Pillo S, Fragassi N, Buscaino GA, Striano S. Primary reading epilepsy : clinical and EEG study of a case and characteristics of the effective stimulus. Acta Neurol Napoli 1983 ; 5 : 426 – 31.
9. Masaoka S, Suguita T, Nanno H, et al. Reading epilepsy : epileptic seizures induced by mental activity. Clin Electro-Enceph (Jpn) 1975 ; 17 : 595 – 605.

Abstract

Reading epilepsy is rare. We report a 14-year-old right-handed Japanese boy who had jaw jerking only while reading since age 12 years. The episodes occurred every time he read an English textbook and sometimes with prolonged reading of a Japanese textbook. The jaw jerking evolved to generalized tonic-clonic seizures (GTCS) on only two occasions during prolonged reading aloud. Routine EEGs showed no abnormality. After a few minutes of reading, however, the EEG showed bilateral 2-Hz, 150-μV spike-wave complexes with left frontotemporal accentuation, accompanied by jaw jerking. Ictal single photon emission computed tomography (SPECT) using [^{99m}Tc] hexamethylpropylene amine oxime (HMPAO) showed focal hyperperfusion of the frontal lobes bilaterally and the left temporal area. Interictal SPECT and MRI were normal. The combination of valproate and clonazepam almost reduced his symptoms. Ictal SPECT is a useful technique for seizure localization in reading epilepsy.