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Functional imaging in reading epilepsy: A case report

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Reading epilepsy is an uncommon epileptic syndrome preferentially related to the temporoparietal region of the language dominant hemisphere. We report ictal and interictal brain perfusion SPECT images in a 28-year-old woman who was reading epilepsy.

Key words: brain SPECT, reading epilepsy

READING EPILEPSY (RE) is an uncommon epileptic syndrome firstly described by Bickford et al. in 1956.[1] Wolf performed a detailed review of 111 reported cases of RE, and pointed out controversies regarding classification and basic mechanisms of precipitation of the seizures.[2] In the International Classification of Epilepsies and Epileptic Syndromes (ICES), RE is recognized as an idiopathic localization-related epilepsy.[3] But this categorization was debated by Radhakrishnan et al., who proposed that RE would be an idiopathic generalized epilepsy, with some resemblance to juvenile myoclonic epilepsy.[4] Ramani suggested a more complex division of the RE subtypes, according to the broad categories of the ICES.[5]

Functional imaging by positron emission tomography (PET) or single photon emission computed tomography (SPECT) was rarely performed in RE. We studied ictal and interictal SPECT in a woman with RE to contribute to the understanding of brain perfusion abnormality of this disorder.

CASE REPORT

The patient was a 28-year-old woman with no family history of epilepsy. Since the age of 17, she had focal tonic contractions of the left leg, sometimes evolving to a generalized convulsion. She was seizure-free for 5 years while on primidone 750 mg daily, tapered in the last year to 250 mg daily. At the age of 24, she began to have jaw jerking and groaning when reading regardless of the design of the letters, that progressed to a generalized convulsion on two occasions. She did not have any history of epilepsy with any other stimulant such as music or pictures. Neurological examination, cranial computed tomography and magnetic resonance imaging (MRI) were normal. The baseline electroencephalogram (EEG) was unremarkable, whereas bilateral paroxysms of short spike and wave complexes appeared in association with typical jaw jerking, after about 30 min of reading a text. The patient was assessed for handedness by using a battery of neuropsychological tests, including the Ankara University Hemispheric Dominance Test, the Dominance Inventory by Kimura and Vanderwolf, the Handedness Questionnaire modified from Annette and the Grooved Pegboard Test; she was right-handed. An ictal SPECT was performed during a seizure provoked by silent reading. Jaw jerking began after 15 min of reading, and HMPAO 740 MBq was administered intravenously.

The ictal and interictal Tc-99m HMPAO brain SPECT were obtained (GE Starcam 4000I, 128 × 128 matrix, 30 sec/frame, 64 projections and 360° tomographic study). There was hyperperfusion of the right superior temporal region in the ictal phase whereas normal perfusion was seen in the same region in the interictal phase (Fig. 1).

DISCUSSION

Our patient satisfies the original diagnostic criteria of primary reading epilepsy, the only exception being the antecedent non reading-related seizures. This feature seems not sufficient to evoke a "secondary" etiology, since she has normal neurological examination, resting EEG and MRI. We suggest to categorize her case as idiopathic localization-related epilepsy, according to

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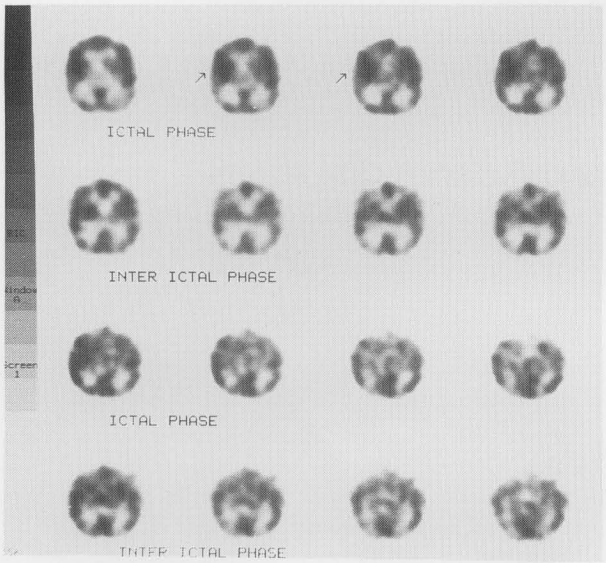

Fig. 1 The ictal and interictal Tc-99m HMPAO brain SPECT transaxial slices of a reading epileptic patient. The hyperperfusion of right superior temporal region in ictal phase (arrow) whereas the normal perfusion was shown at the same region in the interictal phase.

Ramani's classification.[5]

Wolf, in his review of the EEG paroxysmal findings with reading provocation, noted that discharges were lateralized or unilateral in about two-thirds of the cases, lateralization to the language-dominant hemisphere being more common than to the non-dominant side.[2] Koepp et al. also observed lateralization in RE.[6] In our patient, however, ictal EEG activity is not lateralized. Koepp et al. reported the first study of functional imaging in a patient with RE and demonstrated by PET a significant release of endogenous opioids during epileptic activity in the thalamus and left and right temporoparietal regions.[7,8] Their findings do not support a presumed lateralization of seizure onset. Miyamoto et al. studied a Japanese boy with RE by ictal HMPAO-SPECT, that showed focal hyperperfusion of the frontal lobes bilaterally and of the left temporal area, and they speculated that reading epilepsy would be triggered by neuronal processes involved in the elaboration of language, that is, in the frontotemporal region of the language-dominant hemisphere.[9] In our patient, the brain region involved appears on the right side, strongly suggested to be the non language-dominant hemisphere by the neuropsychological assessment. Her antecedent non reading-related seizures affecting the left leg could be another argument for this right lateralization.

The clinical, EEG and functional imaging data provided by this case study militate against a simplistic understanding of the RE syndrome, especially with respect to neuroanatomical substrate and basic mechanisms of seizure precipitation. We propose, in agreement with Ramani, that multiple mechanisms would be involved in RE and SPECT images would have an additive effect in evaluating RE.

REFERENCES

1. Bickford RG, Whelan JL, Klass DW, Corbin KB. Reading epilepsy: clinical and electroencephalographic studies of a new syndrome. *Trans Am Neurol Assoc* 81: 100–102, 1956.
2. Wolf P. Reading epilepsy. In: Roger J, Bureau M, Dravet C, Dreifuss FE, Perret A, Wolf P, eds. *Epileptic Syndromes in Infancy, Childhood and Adolescence*. 2nd ed. London: John libbey, pp. 281–298, 1992.
3. Commission on Classification and Terminology of the International League Against Epilepsy. Proposal for revised classification of epilepsies and epileptic syndromes. *Epilepsia* 30: 389–399, 1989.
4. Radhakrishnan K, Silbert PL, Klass DW. Reading epilepsy: An appraisal of 20 patients diagnosed at the Mayo Clinic, Rochester, Minnesota, between 1949 and 1989, and delineation of the epileptic syndrome. *Brain* 118: 75–89, 1995.
5. Ramani V. Reading epilepsy. In: Zifkin BG, Andermann F, Beaumanoir A, Rowan AJ, eds. *Reflex Epilepsies and Reflex Seizures: Advances in Neurology*, Vol. 75. Philadelphia: Lippincontt-Ravan Publishers, pp. 241–262, 1998.
6. Koutroumanidis M, Koepp MJ, Richardson MP, Camfield C, Agathonikou A, Riğed S, et al. The variants of reading epilepsy. A clinical and video-EEG study of 17 patients with reading-induced seizures. *Brain* 121 (Pt 8): 1409–1427, 1998.
7. Koepp MJ, Hansen ML, Guildin B, Pressier RM, Sheuler W, Ried S. Functional imaging in reading epilepsy. *Epilepsia* 35 (Suppl 7): 38, 1994.
8. Koepp MJ, Hansen ML, Pressler RM, Brooks DJ, Brandl U, Guldin B, et al. Comparison of EEG, MRI and PET in reading epilepsy: a case report. *Epilepsy-Res* 29 (3): 251–257, 1998.
9. Miyamoto A, Saturo T, Tokumitsu A, Oki J. Ictal HMPAO-single photon emission computed tomography findings in reading epilepsy in a Japanese boy. *Epilepsia* 36: 161–163, 1995.