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Epilepsy & Behavior 6 (2005) 463–466
Epilepsy & Behavior
www.elsevier.com/locate/yebeh

Case Report

Writing epilepsy: A neurophysiological, neuropsychological and neuroimaging study

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Received 1 December 2004; revised 6 February 2005; accepted 8 February 2005

Abstract

Writing epilepsy is a rare reflex syndrome in which seizures are triggered by writing. We describe a 33-year-old, right-handed man, with a history of juvenile absence epilepsy in remission and a family history of epilepsy, in whom myoclonic jerks precipitated exclusively by writing started at the age of 30. Intensive video/EEG monitoring during neuropsychological tests revealed, at about 1 minute after starting to write, a dystonic posture, followed by myoclonic jerks involving the right hand that shortly after became generalized. Concomitantly, the ictal EEG documented generalized hypersynchronous polyspike–wave discharges, maximal over the right parietocentral area. SPECT revealed an ictal hyperperfusion and interictal hypoperfusion over right parietofrontal regions, and fMRI showed extensive and intense left frontal, supplementary motor area activation, induced by writing. This case study provides some evidence supporting the hypothesis that the mechanism underlying writing-triggered seizures may be a generalized seizure process, with a focal cortical trigger zone, presumed to be the left frontal lobe as suggested by clinical and fMRI data. A relevant role played by the right hemisphere (right parietofrontal region) is postulated in the full-blown expression of reflex epileptogenesis, as supported by EEG and SPECT findings.

Keywords: Writing-induced epilepsy; Graphogenic epilepsy; Language epilepsy; Reflex epilepsy

1. Introduction

In language-induced epilepsy, a well-known syndrome proposed by Geschwind and Sherwin [1], different aspects of language function, namely, reading, writing, and speaking, may precipitate seizures. In the majority of the reported cases [2–4] reading is the major triggering factor and writing is only an additional precipitating stimulus. Furthermore, seizures induced exclusively by writing are very rarely described in the literature [5], and in the reported cases of graphogenic epilepsy, the reflex seizures were related to juvenile myoclonic epilepsy [4–8]. To our knowledge, a pure graphogenic epilepsy syndrome associated with a past history of juvenile absence epilepsy has not yet been described.

This case is a good experimental paradigm for studying writing-induced epilepsy with neuropsychological tests under video/EEG monitoring and functional neuroimaging (SPECT and fMRI).

2. Case report

The 33-year-old, right-handed man has been attending our epilepsy clinic in S. João Hospital in northern

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doi:10.1016/j.yebeh.2005.02.004

Portugal since he was 12 years old. At that age, he was electroclinically diagnosed with juvenile absence epilepsy and, shortly after onset, became seizure-free.

At the age of 30, he began to complain about jerks involving initially the right hand and immediately spreading to the whole body. This ictal behavior was reproducibly precipitated by handwriting, with a regular latency of 1 minute. His family history included an aunt with epilepsy. His neurological exam was normal (WAIS: 34). Interictal EEGs and cerebral MRI scans were normal.

We obtained video/EEG recordings during neuropsychological tests (NPT) (Table 1) using the same methodology as described by Matsuoka et al. [6] and Pegna et al. [9]. Ictal and interictal HMPAO-SPECT and functional MRI using semantic decisions, reading, finger tapping, and writing paradigms were also performed.

For the ictal SPECT study, 740 mBq of $^{99m}T_C$-HMPAO was injected intravenously at the onset of a clinical seizure (right-hand myoclonus), which occurred 1 minute after the patient started writing.

During NPTs and only when writing, EEGs showed generalized hypersynchronous polyspike–wave discharges, maximal over the right parietocentral area (Fig. 1), concomitant with a dystonic posture of the writing hand, followed by a brief myoclonic jerk (few seconds) that affected the arm and the axial muscles and, finally, the entire body. The patient was fully alert during the seizure and remembered all the actions requested. He had clinical and subclinical seizures precipitated by all forms of writing (spontaneous writing, dictation, and copying), even when he was writing blindfolded. In comparison to dictation and copying, spontaneous writing induced the shortest latency to discharges, longest duration of discharges, and largest number of seizures. No clinical seizure or EEG discharges were induced by other NPTs such as written and mental calcu-

Table 1
Neuropsychological tests

Reading silently and aloud
Regular text
Noncomprehensible text
Writing
Spontaneous writing, dictation, copying
Spontaneous writing and dictation blindfolded
Speaking
Spontaneous speaking and repeating
Other verbal activities
Visualizing numbers and letters
Visual labyrinth
Constructing sentences in the mind
Calculation
Written and mental calculation
Spatial construction
Copying figures (Rey figure)
Spontaneous drawing
Block Design test of Wais-R
Mental construction of block design

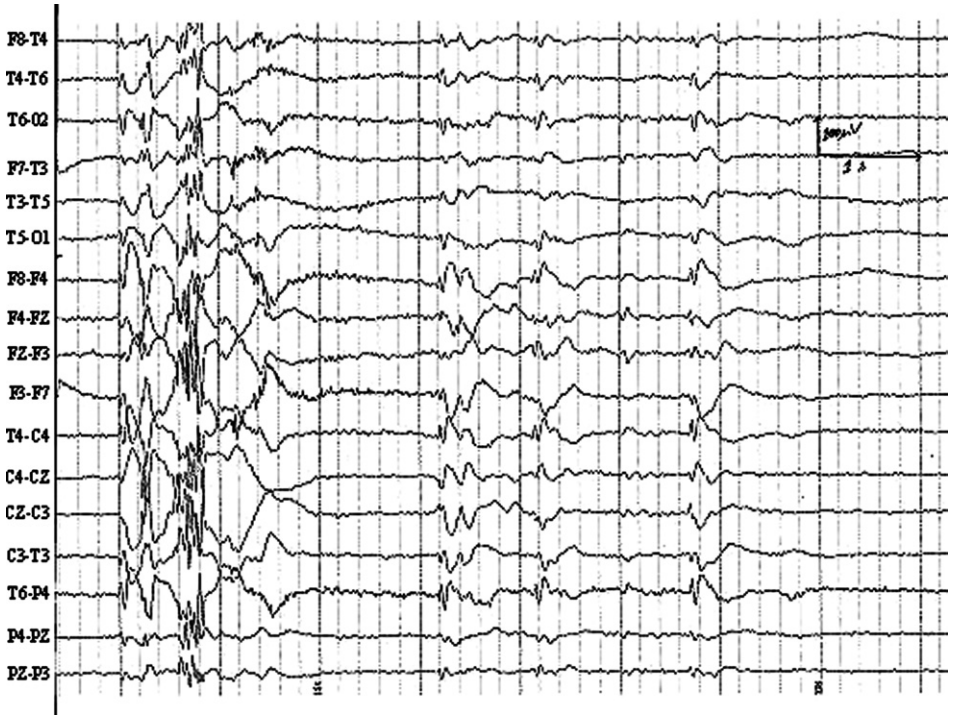

Fig. 1. Spontaneous asymmetrical discharges (maximal over right parietocentral area) with clinical seizures, observed when our patient was writing.