Original article

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# Stuttering or reflex seizure? A case report

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**ABSTRACT –** Stuttering is characterized by involuntary syllabic repetitions and interruption in the smooth flow of speech. The exact cause of primary stuttering remains a matter of debate but a frontal dysfunction has been evoked. On the other hand, acquired stuttering is uncommon. We report a case of reflex epilepsy in which seizures were triggered by reading aloud or stressful conversation. Each paroxysmal event in left frontal region was associated clinically with a language disorder mimicking stuttering. Our observation suggests that reflex frontal focal epilepsy could be a putative etiology for acquired stuttering. [published with videosequences]

***KEYWORDS:*** stuttering, reflex epilepsy, partial frontal seizures

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Stuttering has been defined as a disruption of the fluency of verbal expression characterized by the involuntary repetition or prolongation in the utterance of sounds and syllables. Two forms of stuttering are distinguished. Developmental stuttering appears in children between the age of three and eight years and often disappears before puberty. Acquired stuttering is usually considered to be of neurogenic origin [1], but it is only recently that its biological substratum has been investigated. Incomplete left lateralization of speech motor areas has been inferred from EEG studies using dichotic word listening and word viewing [2]. This finding is supported by functional brain imaging, which reveals incomplete left lateralization of the speech-motor system and overactivity of premotor areas in stuttering [3].

However, in most subjects, stuttering is not a permanent condition and can be favored by the emotional context or some aspects of language production. In view of the dysfunctioning of the speech area reported in stuttering, an intriguing question is whether this paroxystic dysfunction could correspond to some form of focal epileptic activity. The diagnosis of simple partial seizure is sometimes difficult to establish because the clinical features of the seizure may mimic certain voluntary activity. In some cases, the seizures themselves are triggered by ordinary behavior and, if of short duration, can easily be misdiagnosed. In reflex epilepsy, clinical and electrical manifestations are also induced by a specific stimulus such as speech or reading [4]. These forms of epilepsy are relatively uncommon and can be classified according to the triggering factors and their etiology.

We report the original observation of a partial frontal seizure accounting for a brief interruption of language, with a

jerking jaw. This symptomatology was triggered by reading aloud or by stressful conversation. For five years, the disorder was considered as stuttering, the jerking jaw being interpreted as a facial hemispasm. We discuss the putative link between acquired stuttering and partial reflex epilepsy induced by language and reading.

## Case report

A 25-year-old, right-handed woman suffered perinatal hypoxia but had no psychomotor development problems. Her family history was unremarkable.

At the age of 20, she developed clonic jerking of the right jaw while reading aloud or speaking. Emotional stress tended to aggravate the symptomatology. The intensity of the jerking was variable and mimicked a tonic hemispasm. The clonic spasms were associated with difficulty in talking, and her language disorder during these episodes was very similar to stuttering. She never exhibited any typical generalized seizure.

Physical and neurological examinations were normal. No structural lesion was found on MRI, either with classical T1-T2 or inversion/recuperation flair sequences. Interictal SPECT with HMPAO was normal. Neuropsychological testing was considered to be normal for memory, attention and reasoning.

A video-electroencephalogram (EEG) with electromyogram (EMG) recordings was performed on 14 channels. The 12-channel EEG was recorded from a standard bipolar montage of electrodes situated bilaterally in the frontal and prefrontal, temporal, parietal and occipital regions. The EMG was recorded by bipolar disk electrodes over the right orbiculis oris (EMG 2) and the right deltoid (EMG 1). Recordings were performed at rest, while reading aloud and silently, and during writing. The standard EEG with the patient relaxed and awake, eyes closed, was normal (*figure 1A*), as well as during silent reading and writing (*figure 1B*). During reading aloud, these paroxysmal events in the left frontal region were consistently associated with a brief interruption of language (*figure 1C*). The clinical manifestations were accentuated, and speech became incomprehensible when the discharges were clustered for five to 10 seconds. The paroxysmal discharges were usually but not always associated with a brief EMG burst in the jaw muscles. Back-averaging of right facial myoclonia showed reliable EMG-EEG correlates (not shown). When the number of spikes, language suspensions and myoclonic jerks was studied for each minute of recording, there was a clear correlation between the number of spikes and either the number of language suspensions (*figure 1D*) or myoclonic jerks (*figure 1E*). EEG cartography showed low frequencies and high amplitude events in the left frontal internal cortex (*figure 2*).

Language suspensions and spikes were more numerous when reading was prolonged. Although the language progressively became more dysfluent, no dysphasia was observed. From time to time, the patient stopped for several seconds before recovering her speech. The symptoms resolved with normalization of EEG after administration of sodium valproate, 500 mg twice daily.

## Discussion

This case of stuttering associated with reflex frontal seizure induced by speech in a patient with no detectable MRI lesion challenges the putative link between acquired stuttering and partial reflex seizures induced by language. In complex reflex epilepsy, seizures are triggered by cognitive activities. Primary reading epilepsy (RE) [5] is characterized by the occurrence of myoclonic movements of the jaw during reading. The continuation of reading usually induces a generalized tonicoclonic seizure, each myoclonic jaw movement being associated with a brief, bilaterally synchronous paroxysmal discharge on the EEG. Since the first description, several reports have concerned myoclonic seizures induced either by reading or other language tasks such as speaking or writing. The pathophysiological mechanism and neuroanatomical substrate of RE are unknown. The putative mechanisms include proprioceptive inputs from the peripheral muscles (jaw, larynx, eyes) during phonation, cortical reflex myoclonus or a central language disorder. Such symptoms do not occur uniformly in every patient and no single factor can be incriminated as the critical precipitating stimulus. Indeed, the cumulative effect of different factors has been postulated [6].

In our patient, the seizures were characterized by a brief suspension of language, mimicking stuttering. Focal seizures on the left frontal cortex were induced by reading aloud and were associated with facial myoclonus. Since reading silently did not induce any epileptic discharge, articulatory movements during phonation could have been the main triggering factor. Although the limited routine EEG channels used in this patient could not rule out the primary involvement of the motor cortex, it is likely that the language disorder resulted from the diffusion of the epileptic discharge to the speech areas. Indeed, the speech disorder was too complex, especially during prolonged epileptic discharges, to be explained by the facial myoclonus. Interestingly, the phenomenon occurred in particular conditions of stress (social situations or stressful conversation), as did the stuttering. This suggests that a combination of events might be involved in triggering the seizures. The heterogeneity of the clinical aspect and the EEG characteristics in reading epilepsy might be due to a network hyperexcitability involving multiple, non-contiguous, anatomical cortical areas in both hemispheres [7]. This hypothesis is strengthened by studies investigating brain activity during normal reading during PET [8] and reading epilepsy during SPECT [9].

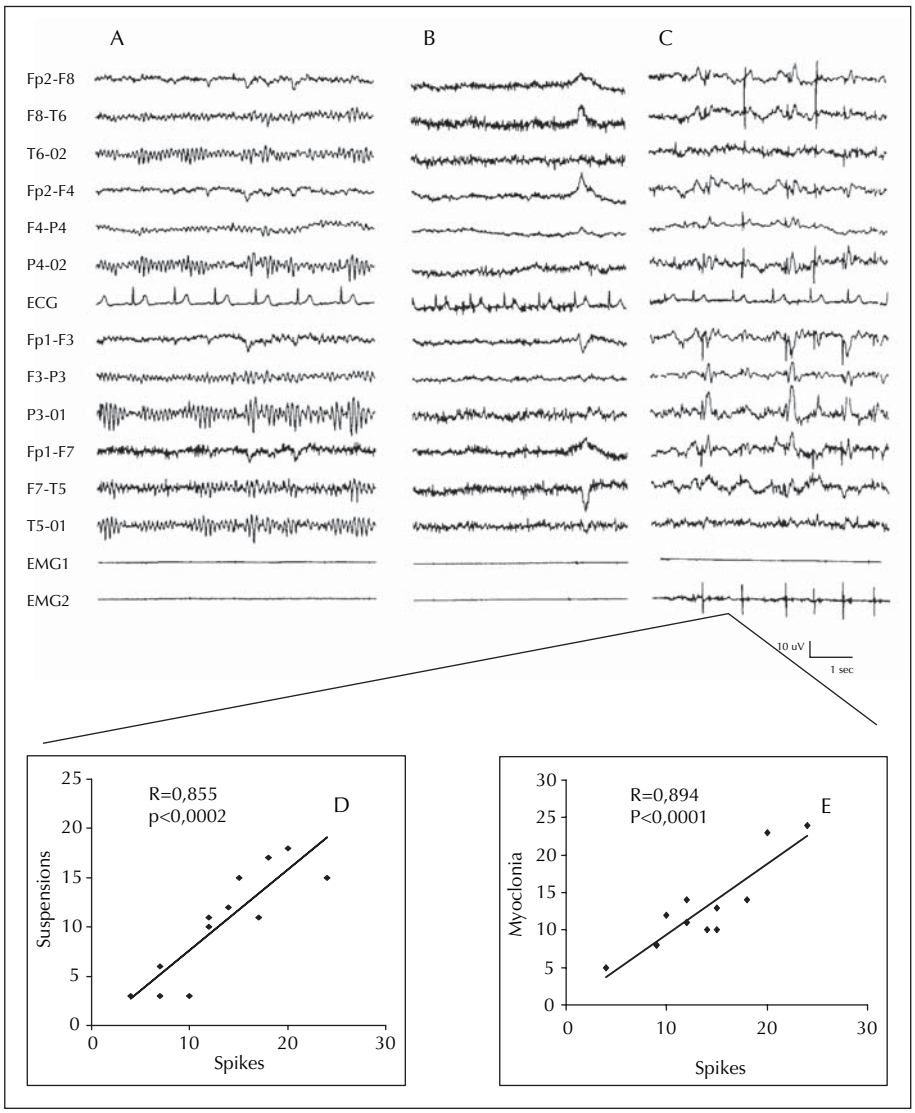


**Figure 1.** *EEG recording (montage longitudinal, cond:10µV/mm, 0.3s, 15 Hz) 1A: Normal EEG recorded during relaxed wakefulness with eyes closed. 1B: Normal EEG recorded during writing and reading silently. 1C: Left fronto-temporal spike-wave discharges during reading aloud associated with electromyographic discharges recorded from the right orbicularis oris (EMG2). Note correlation between myoclonia, spikes and suspension of language during reading. We computed the number of spikes and language suspensions every minute, and found a good correlation between spikes and myoclonia (R=0.894, p<0.0001) (1D) and between spikes and language suspensions (R=0.855, p<0.0002) (1E).*

The present case report raises a thought-provoking question: are some forms of stuttering due to partial epileptic seizures? A potential link is suggested by several arguments. The pathophysiology of stuttering remains obscure and a number of causative factors have been proposed such as a speech motor control disorder, a language disorder with worsening stuttering during language skill acquisition, and the failure of auditory processing [11]. Although specific control-correlates of stuttering have not yet been found by neuroimaging studies, a general consensus is emerging. Several PET studies have shown that during dysfluent speech tasks such as spontaneous speech or reading aloud, stutterers present hypoactivity in cortical areas associated with language processing but hyperactivity in areas associated with motor function [3]. Moreover, Salmelin and coworkers [11] used magnetoencephalography to explore the temporal dynamics of cortical activations. After seeing each word, stutterers presented cortical activation first in the motor cortex and premotor area (associated with motor programs), and then in the left inferior frontal region (associated with articulatory and linguistic processing). Interestingly, non-stutterers showed the reverse pattern. Stutterers thus seem to initiate motor programs before the articulatory code is prepared. Above

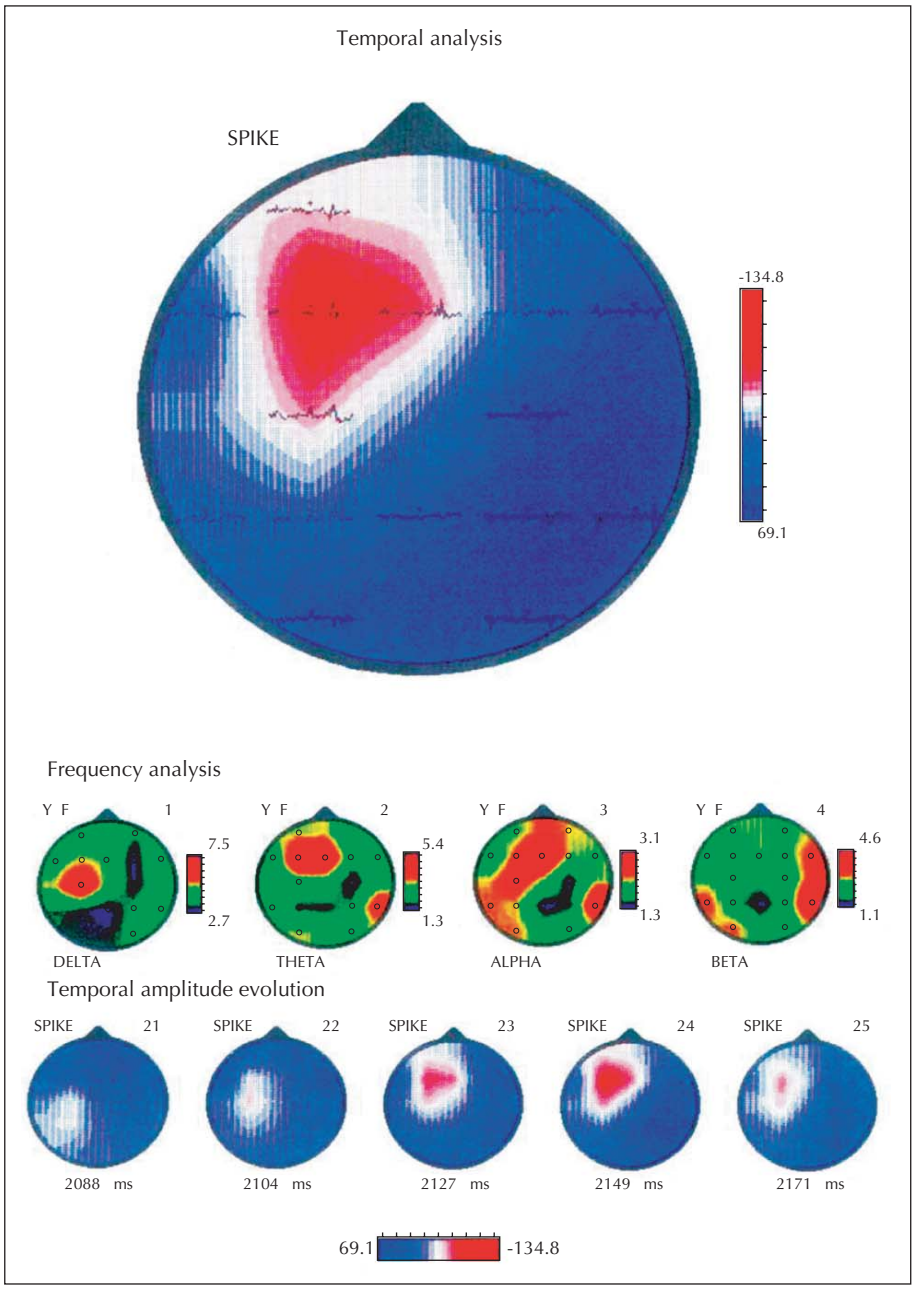


**Figure 2.** *Topographical EEG mapping of sharp-wave discharges. The map series shows the spatiotemporal evolution and frequency evolution of the discharges by sequential amplitude maps recorded versus common average references. The series indicates that the discharges show a left frontal field.*

all, the perisylvian speech and language areas in stutterers are anatomically anomalous [12]. Some stutterers also exhibit abnormal gyration, a malformation which is known to be potentially epileptogenic.

Our observation suggests that at least some forms of acquired stuttering could be linked to epileptic activity in speech-language areas. In this respect, it would be useful to investigate patients exhibiting atypical forms of stuttering with video-electrophysiological techniques. □

**Legend for the video**

The patient reads a newspaper. The stammering begins a few minutes after starting to read, and is accentuated with the length of time of reading. Language becomes more dysfluent and more incomprehensible when the paroxysmal discharges are clustered.

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