

Ictal Forced Repetitive Swearing in Frontal Lobe Epilepsy: Case report and review of the literature

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SUMMARY

Introduction. Dominant presentation of ictal forced repetitive swearing has been rarely addressed and could be misdiagnosed.

Case report. We report a 45-year-old man with a long history of right frontal lobe epilepsy (FLE) who developed forced repetitive swearing during hypermotor seizures. His seizures were refractory to different antiepileptic drugs (AEDs). Scalp video-EEG telemetry suggested a right frontal epileptic focus. Magnetic resonance imaging (MRI) suggested focal cortical dysplasia (FCD) in the right mesial frontal lobe. Intracranial implantation with video-EEG recordings confirmed seizures originating from the MRI lesion. Patient underwent right frontal lobe resection followed by seizure freedom in the last five years on a single AED. Neuropathology confirmed FCD type IIB.

Discussion. The following aspects of the case are discussed: FLE and ictal vocalization, swearing, FLE and aggression. We emphasize the differences among ictal vocalisation, verbal automatism and ictal speech. We propose that ictal swearing might fit a verbal automatism definition.

Conclusion. Ictal forced repetitive swearing can be a manifestation of hypermotor seizures in FLE and should not be misdiagnosed.

Key words: ictal swearing • frontal lobe epilepsy • lateralization

INTRODUCTION

Frontal lobe seizures may manifest as bizarre behaviors such as kicking, thrashing, unusual facial expressions and genital manipulation (Williamson et al., 1985). Articulate vocalizations with variable complexity with uttering words and shouted obscenities have been described in frontal lobe epilepsy (FLE) (Williamson et al., 1985; Jobst et al., 2000). These patients can be misdiagnosed and mismanaged for years (Jobst et al., 2000). We studied an adult man with a long history of FLE who developed forced repetitive swearing as a dominant clinical feature during his hypermotor seizures.

CASE REPORT

A 45-year-old right handed man was referred to our Sei-

zure clinic by his family physician for an opinion regarding episodes of bizarre behavior. The patient's prenatal delivery and development history was unremarkable. He presented with generalized tonic-clonic seizure at age four that recurred on two other occasions at 18 and 28 years of age and were generally controlled on antiepileptic drugs (AEDs). A few years after the first seizure he started having stereotypical attacks characterized by sudden awakening from sleep with rocking on bed, flailing his arms, shouting swears on Jesus for about 15 seconds (see a video). Before the episodes he reported tingling in his back for a moment. He did not have falls, urinary incontinence or tongue biting. There was no confusion after episodes. He was aware

that he had a behavioral change, would usually apologize right after seizures but was amnesic for details of his behavior. Attacks occurred up to 40 times per day. They appeared mainly from nocturnal sleep but few episodes have been reported from day time naps and wakefulness. He tried many AEDs for these attacks including phenytoin, phenobarbital, carbamazepine, valproic acid, levetiracetam and clobazam with seizure reduction only to 4–5 per day.

The patient never physically attacked another person or objected during or between attacks. His attacks were diagnosed as Gilles de la Tourette's vocal tics and treated as such during his adolescence. Attacks frightened other people and at some point were thought to represent a psychiatric disorder. Otherwise, his medical history was unremarkable. He was not working. He lived in a Salvation Army facility and was on disability for his attacks. His maternal grandmother had epileptic seizures. His neurological examination was normal.

Noninvasive presurgical evaluation

The patient had prolonged surface video-EEG telemetry for seven days in August, 2008. We captured 23 seizures during sleep, 4 while waking up from sleep and 4 seizures in the wake state. All seizures were stereotypical as previously described. The patient was amnesic for the major part of each seizure, but could postictally identify the person who assessed him during the

seizure. The patient followed simple commands and demonstrated comprehension of spoken language after each episode when he was examined by staff. He usually would apologize for his behavior right after his seizures.

Surface EEG showed ictal diffuse, low amplitude, fast activity at a frequency of 15–20 Hz with maximum over the right frontal region. The same fast activity over the right frontal region preceded the onset of some seizures by a few seconds (fig. 1). Interictal EEG was remarkable for mild to moderately active low amplitude often ill-defined epileptiform discharges seen over the right frontal, right frontocentrottemporal and to a lesser extent the left frontal regions. Mild intermittent diffuse cerebral dysfunction was recorded without specific lateralization.

Brain magnetic resonance imaging (MRI; 3Tesla T1, T2, 3D FSPGR and FLAIR sequences) showed right mesial frontal cortical thickness, blurring of the gray white matter junction and high signal tapering from cortex to the lateral ventricle which were characteristic imaging findings for focal cortical dysplasia (FCD; fig. 2). FDG-PET revealed diffuse hypometabolism in the right frontal lobe, especially at the location of lesion on MRI (fig. 3).

Invasive surgical evaluation

The patient underwent intracranial implantation with video-EEG recording. Subdural strip electrodes were



Figure 1. Surface EEG showed low amplitude fast activity at a frequency of 15–20 Hz that started diffusely over both cerebral hemispheres, with maximum over the right frontal region (arrow head). Short run for few seconds of fast rhythmic activity at right frontal leads Fp2, F4 and F8 preceded the onset of seizures by at least 8 seconds (arrow).

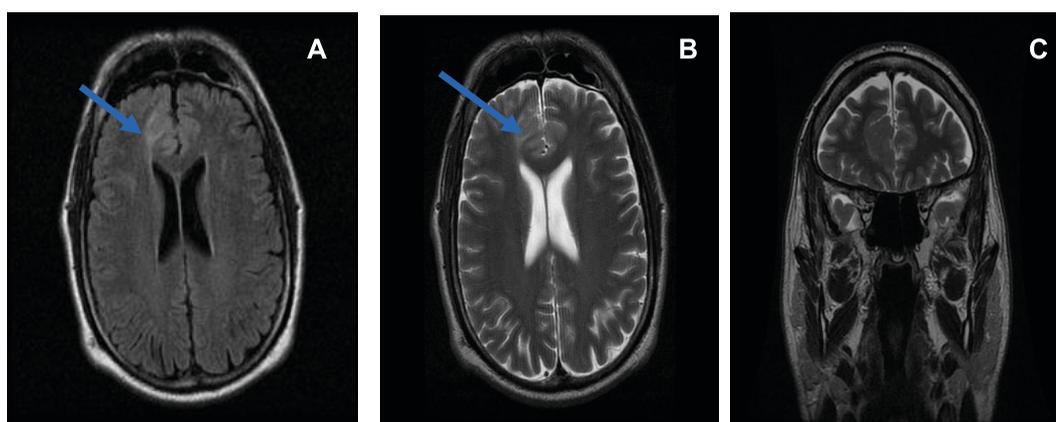


Figure 2. Brain MRI, Axial FLAIR A, Axial T2 B and Coronal T2 C shows right mesial frontal lobe cortical thickness, blurring of the gray white matter junction and tapered signal as it extends to the lateral ventricle (arrows).

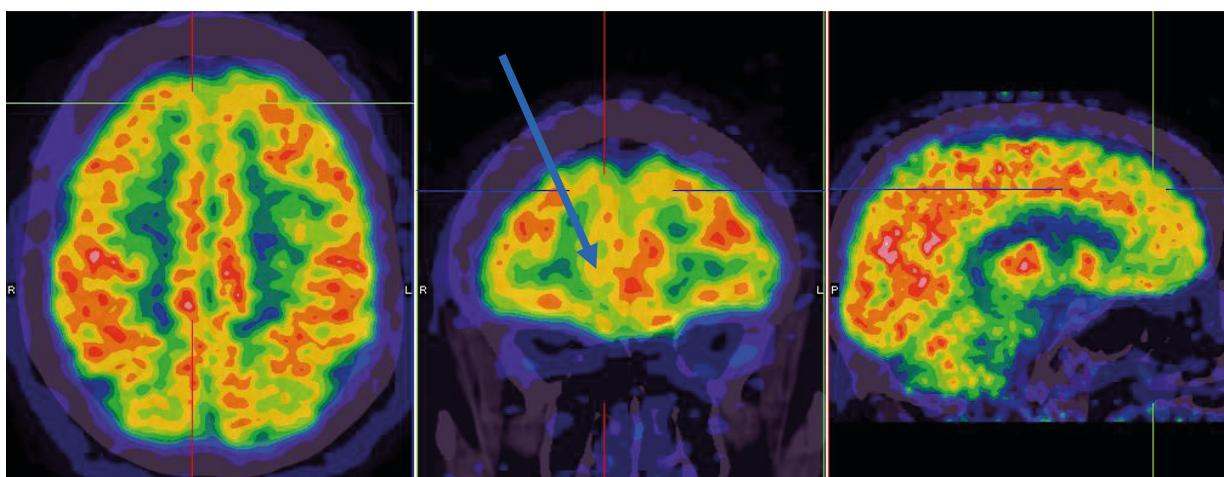


Figure 3. FDG-PET with hypometabolism in the right mesial frontal region (arrow).

implanted over the right frontal region. Two depth electrodes with 10 contacts each were implanted parasagittally in the lesion. One strip consisted of two rows of two-sided contacts and was placed in the anterior hemispheric fissure. Four other strips electrodes and two peg electrodes were placed to record from different areas of surrounding brain. 27 seizures were recorded over a 3 day period. All seizures were originating in the right mesial frontal region inside and overlying the MRI lesion area with quick involvement of other areas of right frontal and left frontal regions but seizure activity remained maximal inside the lesion and overlying cortex until the end of seizures. The interictal EEG was remarkable for very active epileptiform discharges inside the right mesial frontal region involving the MRI lesion and overlying the cortex area (fig. 4).

Epilepsy surgery and postoperative course

The patient underwent right frontal lesionectomy. The neuropathological findings confirmed the diagnosis of FCD Type IIb (fig. 5). The postoperative course was uneventful. The patient has been free of seizures since epilepsy surgery for 5 years on levetiracetam 750 mg twice a day. One attempt to discontinue medication activated typical brief seizure.

DISCUSSION

FLE and ictal vocalizations

We investigated a patient in whom forced swearing has been a dominant clinical feature during seizures and which frightened other people. FLE may manifests as bizarre behaviors including articulate vocalizations with

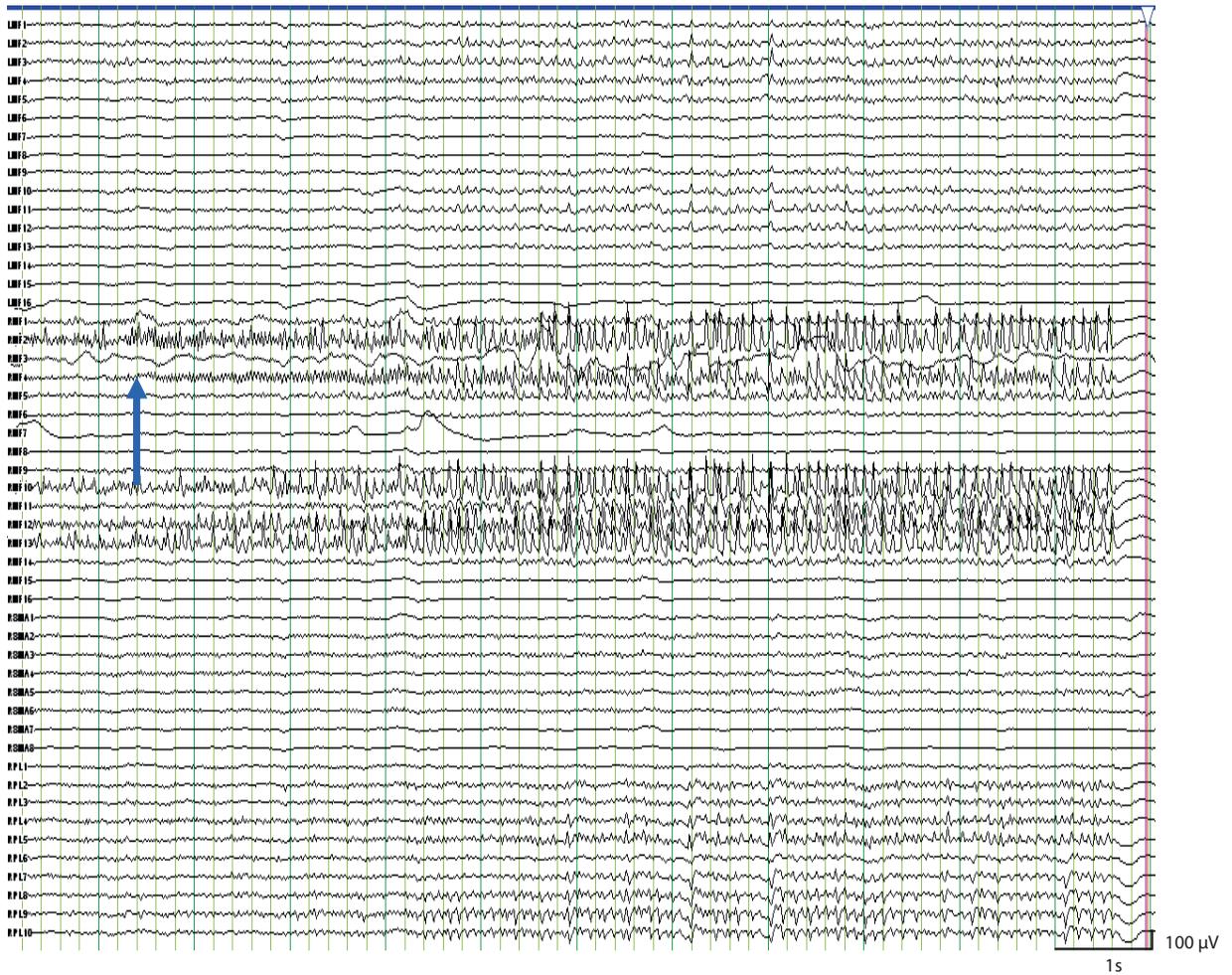


Figure 4. Intracranial EEG showed seizures originating in right mesial frontal (RMF) region with quick involvement of other areas of right frontal lobe but seizure activity remained maximum inside the lesion and overlying area until the abrupt end of seizure.

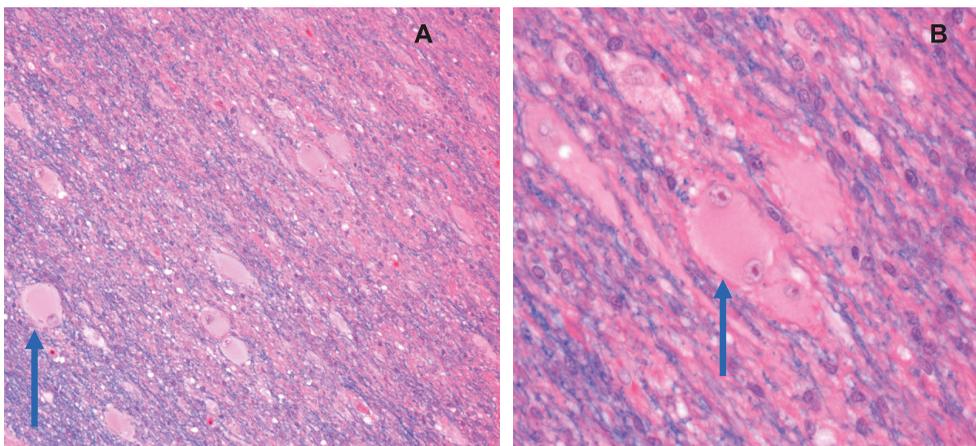


Figure 5. H and E showed Balloon cells (arrow) with eccentric nuclei in the white matter **A**, Some of these cells were binucleated **B**, with lack of neuronal edges basophilia.

variable complexity. Shouted ictal swearing has been briefly mentioned in such patients (Williamson et al., 1985; Jobst et al., 2000; Sumer et al., 2007; Manford et al., 1996). Up to 30% of FLE patients in one series were reported to have ictal articulate vocalizations including swearing that may be part of a complex gestural-hyperkinetic motor activity. It is not clear how many patients presented with swearing, but probably swearing is not a very rare phenomenon (Jobst et al., 2000).

The definition of vocalization was not uniform across studies. Speech, vocalizations, or physical sounds may be present during seizures and should not be confused with each other. Pure ictal vocalizations have been recently defined as audible sounds that lack speech quality and do not accompany apnea or clonic or generalized tonic-clonic seizures.

No uniform agreement exists regarding lateralizing value of ictal vocalizations in FLE. Janszky et al. (2000) reported pure ictal vocalisations in 11 patients with FLE, of whom nine had a left frontal epileptogenic zone. The lateralization of pure ictal vocalization to left frontal lobe was statistically significant. It was therefore concluded that ictal vocalisations could be an additional lateralising sign in FLE (Janszky et al., 2000). Another report on a series of FLE patients, who underwent resective epilepsy surgery, noted ictal vocalizations (less stringently defined as a "phonation of any kind") in approximately two-thirds of the patients; however, an association between vocalization and lateralization was not observed (Manford et al., 1996). In two other studies ictal vocalization was observed to originate from the right hemisphere but these results were not statistically significant (Marsh and Krauss, 2000; Bonelli et al., 2007).

Ictal speech is defined as clearly intelligible propositional speech during a period of altered consciousness (Chase et al., 1967). Ictal speech is a well-accepted lateralizing sign in temporal lobe epilepsy. However, ictal speech is rare in frontal lobe seizures, although non-speech vocalizations often occur. Janszky et al. (2000) reported that 2 of 12 patients with ictal speech had intelligible speech that was lateralized to the dominant frontal lobe.

Verbal automatism or ictal verbalization is described as a single or repetitive utterances consisting of words, phrases, or brief sentences, such as uttering, shouting, talking or singing words, sentences or phrases. Verbal automatism of coherent speech is associated with non-

dominant hemisphere. Both ictal vocalization and verbalization can occur in extratemporal epilepsies such as frontal lobe seizures. They often occur in childhood and juvenile absence epilepsy (Williamson et al., 1998; Yen et al., 1996).

Our patient's ictal swearing occurred as repeated utterances, and not in the form of propositional speech and it was localized to the right frontal lobe. Thus, ictal swearing and ictal speech might probably be generated through different neurophysiological processes. We propose that ictal swearing is a form of ictal verbalization based on the above definitions.

Swearing

Swearing is a common human act and is frequent in neurological disorders. Despite its obvious role in communication, detailed studies of pathophysiology, localization and lateralization of swearing are lacking. Coprolalia, a type of swearing, encompasses words and phrases that are culturally taboo or generally unsuitable for acceptable social use, when used out of context. Compared to voluntary normal swearing, it is usually expressed out of social or emotional context, and may be spoken in a louder tone or different cadence or pitch. Coprolalia is most common in GTS syndrome, but is not unique to tic disorders and may occur in other neurological disorders such as stroke, traumatic brain injury, post-herpetic encephalitis, choreoacanthocytosis, seizures, etc. Brain imaging, neurophysiological, and post-mortem studies support involvement of cortical-striatal-limbic pathways in GTS, but the definitive pathophysiological mechanism or neurotransmitter abnormality is unknown. Recent evidence, however, suggests a prefrontal dopaminergic abnormality. Traditional neuroleptics are the standard treatment for GTS which correlates with this hypothesis (Van Lancker and Cummings, 1999). There is a case report of loss of swearing in a 75-year-old right-handed man after having a right basal ganglia stroke. Post-stroke, he never swore or cursed; also he could not guess the correct expletives for the situations explained to him, nor could he complete a curse (Speedie et al., 1993). Georgiou et al. (1995) presented evidence that GTS involves dysfunction of basal ganglia and interconnections with frontal lobes. Imaging studies of GTS patients showed atrophy of basal ganglia compared to controls. Voluntary normal cursing and epileptic swearing may share the anatomy and physiology of coprolalia (Georgiou et al., 1995).

Ictal swearing/coprolalia

Ictal swearing in our patient with right FLE suggests that the speech output network in the left dominant hemisphere may not be involved in the ictal discharge. Recently, the largest case series of ictal swearing has been published that reviewed two old case reports of ictal swearing and presented 8 new cases. Results indicated that ictal swearing occurs more commonly in male subjects and lateralizes to the non-dominant hemisphere, but has poor localization value, arising either from the frontal, parietal, temporal or occipital lobes in different patients (Birca et al., 2013). Another case report of ictal swearing and aggression in FLE also lateralizes to the non-dominant hemisphere (Shih et al., 2009).

An immediate mechanism of forceful ictal swearing might be episodic disinhibition of frontal lobe control on swearing words and might be a release phenomenon. The famous frontal lobe patient Phineas P. Gage is said to become “fitful, irreverent, indulging at times in the grossest profanity” (Harlow, 1868). In theory, ictal frontal lobe seizure might disinhibit swearing regardless of lateralization of seizure onset, but this has not been studied. Interestingly, patients with left hemisphere stroke or left hemispherectomy and global aphasia have been showed to preserve swearing (Van Lancker and Nicklay, 1992).

People swear to let off steam, to shock or insult, or out of habit. Swearing was shown to increase pain tolerance, increase heart rate and decrease perceived pain compared with not swearing (Stephens et al., 2009). Out of 10 patients with hypermotor seizures reported by Williamson et al. (1985), only two had tightness in the chest as an aura and ictally swore. Our patient also reported unpleasant tingling in the back as his seizure aura. Attempt to reduce stress and pain by swearing might be an easy explanation in everyday life but less likely an explanation in epileptic patients. The same “output neuronal pathway” might be used in seizure or pain trigger. The seizure event can trigger hypermotor clinical manifestations as well as interfering with inhibitory frontal lobe function leading to a release of swearing behavior.

Generally men swear more than women. Men with epilepsy might swear during seizures more than women with epilepsy (Birca et al., 2013).

FLE and aggression

Aggressive behavior is very rare as an ictal phenomenon, while it may be seen in the context of postictal confu-

sion. A large survey of several thousand seizures documented on video-EEG reports an incidence of 1 out of 1000 seizures with aggressive behaviors (Delgado-Escueta et al., 1981). In all of these cases, violent motor automatisms during seizures were mistaken as threatening or assaultive. Although the aggressive act may seem coordinated, it does not include complicated and purposeful acts or detailed behaviors (Janszky et al., 2000). The aggressive behavior is shown towards nearby objects or persons, mostly as pushing and shoving. Classic epileptic presentations such as staring and oral and motor automatisms may be present. Patients usually have no recall of these episodes, and express deep regret for their action (Delgado-Escueta et al., 1981). Differentiation of violent behaviors from an ictal event is not always easy, and video-EEG monitoring can help to do so. Treiman (1986) recommended five criteria to determine whether a specific violent act was the result of an epileptic seizure: (a) an established diagnosis of epilepsy, (b) a video-EEG documentation of epileptic automatisms, (c) a video-EEG documentation of the aggressive behavior, (d) the aggressive act should be characteristic of the patient’s habitual seizures, and (e) a clinical judgment should be made by the neurologist as to the possibility that the violent act was part of a seizure. Although it seems established that aggressive automatisms are related to epileptic activity rising from the amygdala and spreading through the diencephalic regions, no clear supporting data on laterality are available, although most data point to the nondominant hemisphere (Mula, 2014).

CONCLUSION

Ictal swearing is a rare phenomenon but has been described in FLE. The pathophysiology and anatomy of swearing, in general and in FLE remains elusive. We report a FLE case whose ictal swearing went undiagnosed for several years because of its rare ictal presentation. Similar to the case report by Shih et al. (2009), he remained interactive during his seizures. In our patient the origin of seizures was from right frontal lobe secondary to FCD, based on MRI and prolonged scalp and intracranial EEG/video telemetry studies. Post-operatively the patient was seizure-free during a 5 year follow-up period on one AED and this supports the accuracy of presurgical localization. Our case and the similar case report by Shih et al. (2009), suggest lateralization of ictal swearing to non-dominant frontal lobe. However, further studies using the Wada test are need-

ed to investigate the lateralization of ictal swearing with regards to the language-dominant hemisphere.

This study highlights that frontal lobe seizures presenting with swearing may be mistaken with vocal tics, violent profanity and may have potential adverse legal ramifications. In recent years, recognition of hypermotor seizures has improved but still remains a challenge in some cases. Surface EEG may not show ictal epileptiform discharges and might be obscured by artifacts. Clinical pattern and stereotypical presentation is a key. The diagnosis of seizures should always be considered in cases of episodic stereotyped behavior.

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CONFLICT OF INTEREST DISCLOSURE

The authors have no conflicts of interest.

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