

Case report

Simple autonomic seizures and ictal enuresis

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ARTICLE INFO

Article history:

Received 12 January 2011

Received in revised form 28 February 2011

Accepted 22 April 2011

Keywords:

Simple autonomic seizures

Ictal enuresis

Cortical control of micturition

ABSTRACT

Human micturition is physiologically controlled via a complex and finely tuned network of cortical and subcortical regions, hitherto not fully understood. We report the case of a 42-year-old man with juvenile myoclonic epilepsy (JME) and post-traumatic epilepsy. Ictal enuresis was the only manifestation of the *de novo* post-traumatic simple autonomic seizures. Source analysis of the ictal epileptiform discharges showed activation of the cortical areas surrounding the right inferior frontal sulcus, adjacent to the lesional zone. The case depicted here further endorses recent neuroimaging studies which strongly implicate this cortical region in the regulation and the initiation of the micturition.

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1. Introduction

Urinary incontinence poses a major clinical and social problem affecting all age groups. In elderly, dysfunction of certain brain areas has been implicated as one of the major culprits leading to the incontinence.¹ Micturition is a highly complex process synchronized by multiple centres of the central and peripheral nervous system and involves the coordination of functions that are under both involuntary and voluntary control. Thus far relatively little has been known about the exact brain areas involved in the control of micturition. It is generally accepted that the central nervous control is not essential for the basic micturition reflex but it is thought to determine the beginning of micturition.² The phenomenon of ictal enuresis as a sole manifestation of simple seizures was first described by Penfield and Kristiansen in 1951.³ Simple autonomic seizures and their ictal EEG presentation have traditionally been of great scientific interest as indirect means to decipher the true functional role for the implicated neuroanatomical area.^{3–6} Similarly one could suggest that ictogenic focus resulting in enuresis may help provide an insight into the functional organization of the cortical centres of micturition. However, only a few cases of true simple ictal enuresis have been reported in the scientific literature to date.^{3–6} We report an unusual case of paroxysmal episodes of urinary incontinence developing *de novo*, after the post-traumatic brain injury, in a patient with juvenile myoclonic epilepsy (JME). Ictal EEG

suggested seizure onset from the posterior parts of the right middle and inferior frontal gyrus (Fig. 1C), the brain area previously implicated by the neuroimaging studies in the cortical control of micturition.^{7–10}

2. Case description

A 42-year-old right-handed man with a long history of epilepsy, starting when he was 10 years old, was referred to a tertiary epilepsy centre for video-telemetry investigation of his newly developed paroxysmal enuresis. No family history of epilepsy or childhood illnesses, inclusive of febrile seizures, were reported. During the juvenile period his habitual seizures were infrequent on antiepileptic medication and consisted of generalised tonic clonic seizures (GTCS), absences and myoclonia. He was diagnosed with JME.

In his late 30s, the attacks increased in frequency. Daily absences, weekly GTCSs and additional polymorphic myoclonic jerks were all reported. During one of his seizures he suffered a traumatic fall with loss of consciousness resulting in significant post-contusional bleeding, mainly to the right frontal and temporal lobes. Two urgent neurosurgical evacuations of haematomas via burr holes were necessitated before he was deemed clinically stabilised.

The exact temporal onset of paroxysmal attacks of urinary incontinence following his traumatic brain injury is unclear. However, once established, attacks happened daily, up to several times per day. No aura or associated change in the mental state were reported. Albeit brief in duration, habitually lasting up to 15 s, the patient perceived them as highly disturbing and they begot a great social and psychological handicap for him. The semiology and frequency of his other seizures (GTCS, absences, myoclonic

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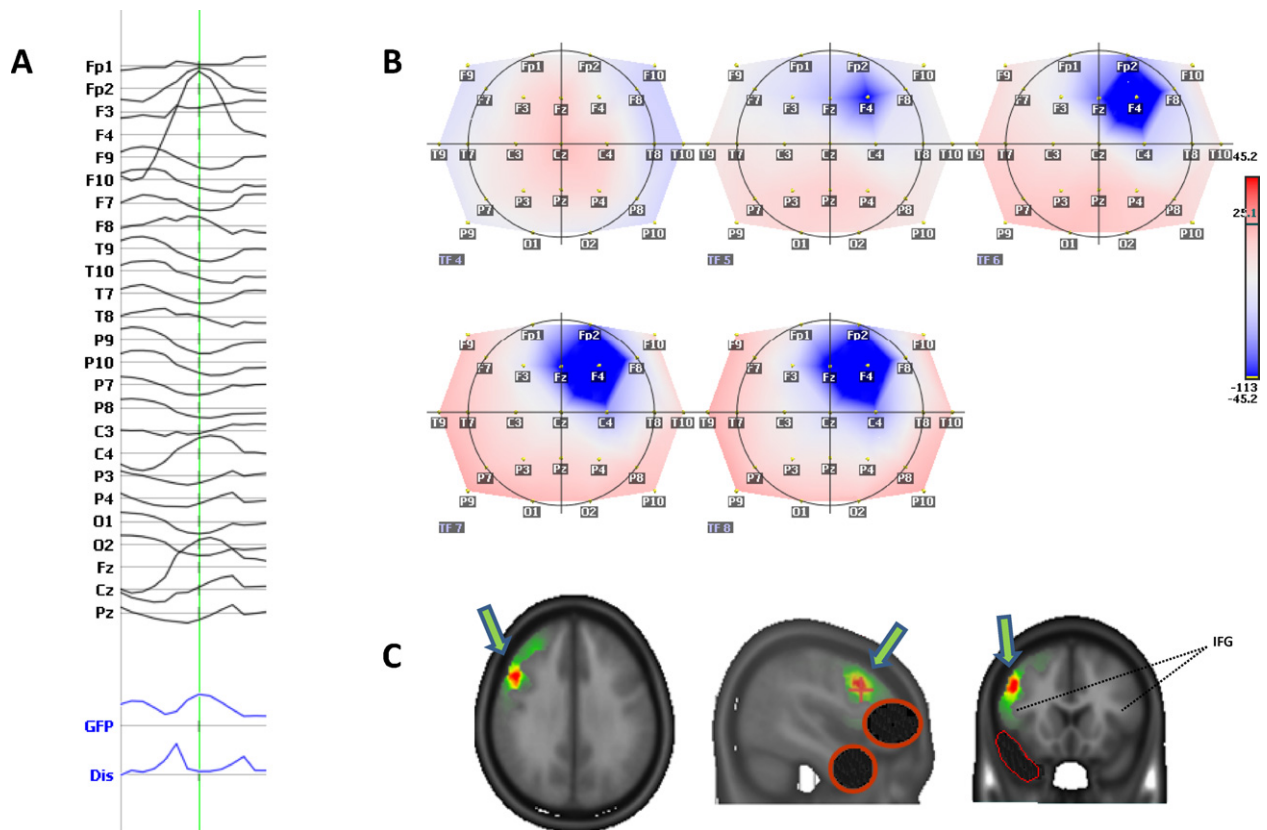


Fig. 1. The averaged-wave of the ictal discharges (A) and the corresponding voltage-maps are shown (B). To increase the signal/noise ratio, waves from the same ictal discharge were averaged (i.e. aligned to the peak negativity). The frequency of the ictal discharge was determined in successive windows of 1 s duration, with an overlap of 0.5 s. The waves in the initial time-segment were included into the averaging when their frequency was noted constant and the voltage-map of the individual waves had similar field distribution. Finally, the ictal discharges were analysed using the source localisation (LAURA) method and then superimposed (green arrows) on the standardised MRI scan of head (C). Here the anatomical sites, which correspond to postcontusional atrophy and encephalomalacia, are also depicted (red circles). IFG = inferior frontal gyrus. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of the article.)

appeared unchanged and their corresponding EEG (e.g. generalised spike-wave) discharges were unaltered. No breach rhythm was observed.

Neuroimaging revealed several areas of encephalomalacia probably corresponding to resolution of the postcontusional injury and haemorrhage (see Fig. 1C) and the neuropsychology tests revealed patchy cognitive deficits suggestive of fronto-temporal dysfunction. Electromyography and video urodynamics test were reported normal. He was otherwise neurologically intact.

EEG videotelemetry was performed and several episodes of paroxysmal enuresis were recorded and electrographically confirmed. Ictal EEG showed paroxysmal 16–20 Hz burst of spikes in the right frontal region (Figs. 1 and 2). The source analysis of the ictal EEG discharges with LAURA (Local AutoRegressive Average) method¹¹ suggested the region surrounding the posterior portion of the inferior frontal sulcus (inferior part of the middle frontal gyrus, and the superior part of the inferior frontal gyrus) on the right side as the most likely ictogenic locus (Fig. 1C).

3. Discussion

Urinary incontinence is frequently found in patients with stroke or with CNS diseases.⁸ It commonly complicates the end of the clonic phase of a tonic-clonic epileptic seizure when the sphincter muscle relaxes in a patient with a full bladder. It may also occasionally complicate absence and focal seizures, such as it did presumably in our patient, possibly resulting from a combination

of increased intravesicular pressure and loss of cortical inhibition of the micturition reflex.^{2,8,9} Moreover, ictal urge incontinence is considered to be a relatively frequent lateralising sign for the non-dominant temporal epilepsy.^{12–15} Conversely, simple ictal enuresis, which was described in this case, is considered a truly rare simple autonomic phenomenon.^{3–6} In our patient, the ictal EEG source analysis broadly suggested cortical areas surrounding the non-dominant right inferior frontal sulcus as the ictal zone of the onset. This brain region has been consistently shown as significantly activated in several PET and other imaging studies of human micturition.^{7–10}

The dorsal region of right inferior frontal gyrus (FG) is involved in attention mechanisms and response selection.⁸ It also modulates the emotional network including nucleus accumbens and hypothalamus. In animal studies, the preoptic area of hypothalamus has been shown to project directly to the M-region, the important pontine micturition centre.² In this context, it is tempting to speculate that, in respect to micturition, the right inferior FG region might play a role in making the decision as to whether or not micturition should take place under the given circumstances.^{8,16} Moreover, lesions in the prefrontal cortex, including right inferior FG, have been reported to cause urge incontinence.^{2,8} The prefrontal cortex has multiple connections with the anterior cingulate gyrus, which has also been implicated in the micturition control by the neuroimaging studies. Both regions have direct or indirect connections with the periaqueductal grey area, the hypothalamus and other areas that are associated with autonomic control.^{2,16} Fowler and colleagues

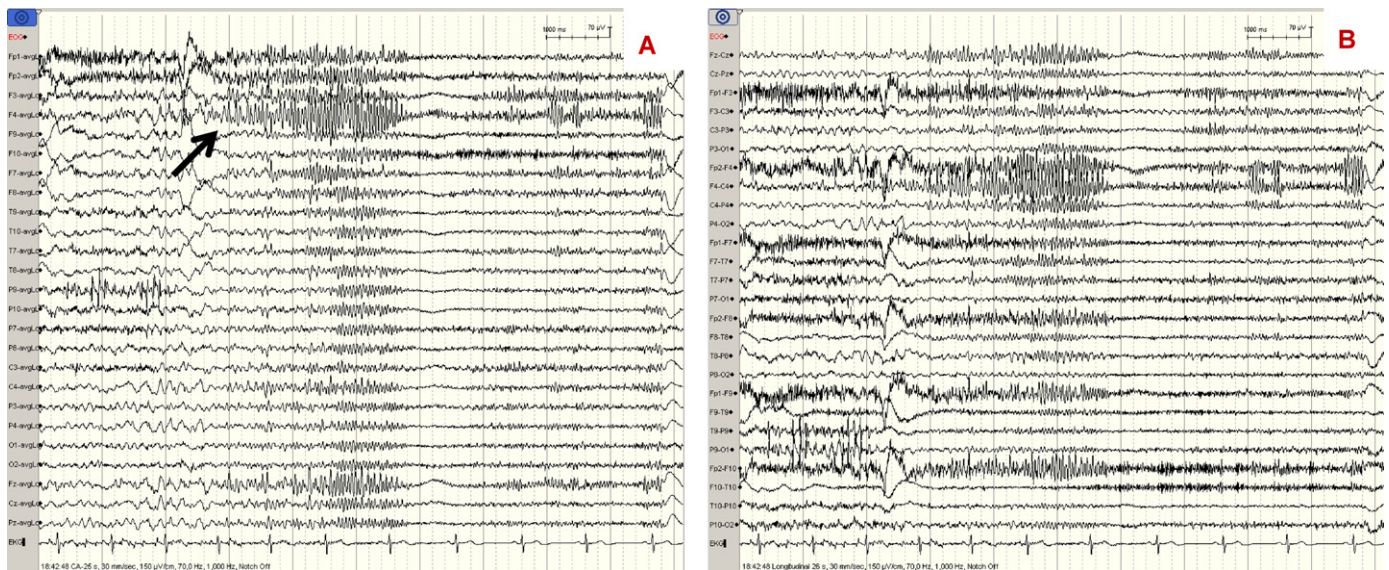


Fig. 2. The electroencephalogram depicting the ictal discharge is shown in two different montages (A—common average, B—longitudinal bipolar). The start of the clinical symptom of enuresis corresponded to paroxysmal 16–20 Hz burst of spikes in the right frontal region (arrow).

recently suggested that this prefrontal cortical ‘inhibitory network’ might be responsible for tonic suppression of voiding, which is then relaxed when voiding is both desired and socially appropriate.¹⁶

Hence, it would hypothetically follow that in our patient the abnormal ictal activation in the cortical area surrounding the posterior portion of the inferior frontal sulcus acts to disinhibit the cortical ‘inhibitory network’ and that this then can potentially lead to the onset of the involuntary micturition. The normal electromyography and urodynamics test potentially also provide some support to this cortical disinhibition theory. On the other hand, the prior diagnosis of JME in our patient might arguably suggest *a priori* aberrant neuronal connectivity and structural abnormalities predating the trauma.¹⁷ By the same token, one cannot fully exclude the possibility that the post-traumatic ictogenic focus somehow utilises or is modulated by the pre-existing idiopathic ictogenic networks. However, the fact that trauma does not appear to have altered generalised spike-wave discharges, in our opinion, makes this notion less compelling. Nonetheless, the traumatic etiology of the *de novo* ictogenic locus, which is clearly adjacent to the areas of encephalomalacia (Fig. 1C), has to be taken into account when gauging the true functional role of the brain region in question.

In conclusion, in this unusual and rare case the causal relationship is highly intimated by the anatomical overlap with the previously reported centres of micturition.^{7–10} The phenomenon of ictal enuresis as a singular autonomic manifestation, resulting from the post-traumatic ictogenic focus in a patient with JME, suggests an important role for the cortical areas surrounding the right inferior frontal sulcus in the cortical regulation of human micturition.

Conflict of interest

None. Dr. Rosenzweig was supported by the EFNS Department to Department Programme grant during her stay at the Department of Neurophysiology, Danish Epilepsy Centre in Dianalund, Denmark.

Acknowledgements

We are grateful to following people for the help with the manuscript and their generous input in this clinical case: Dr. Jørgen Alving, Dr. Noémi Andersen and Dr. Marit Dahl.

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