Seizure: European Journal of Epilepsy 71 (2019) 80–82

Contents lists available at ScienceDirect

Seizure: European Journal of Epilepsy

journal homepage: www.elsevier.com/locate/seizure

Clinical letter

Post-ictal Cotard delusion in focal epilepsy patients

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ARTICLE INFO

Keywords:
Cotard delusion
Post-ictal delusion
Focal epilepsy

1. Introduction

Cotard delusion is a nihilistic delusion that presents as denial of self-existence and is a rare neuropsychiatric phenomenon associated with diverse neurologic or psychiatric disorders [1]. Various case reports and case series have indicated associations between Cotard delusion and neurologic or psychiatric diseases; these include depression, schizophrenia, traumatic brain injury, multiple sclerosis, stroke, and Parkinson's disease [1]. However, there have been few reports of Cotard delusion in epilepsy patients [2]. Here, we describe two patients who exhibited transient post-ictal Cotard delusion with spontaneous recovery after focal or focal to bilateral tonic-clonic seizure.

1.1. Case 1

A 52-year-old right-handed male patient was referred to our team from the Cardiology Department because of repeated transient unresponsiveness. He had end-stage renal disease with daily peritoneal dialysis and congestive heart failure. He had been originally admitted to our institution because of aggravated dyspnea; chest X-ray then showed pneumonic consolidation and he was treated with a third-generation cephalosporin. Two weeks post-admission, his caregiver noted that he showed jerky movement and intermittent unresponsiveness.

To evaluate unresponsiveness in the patient, we performed brain imaging and video-electroencephalogram (EEG) monitoring. On brain computed tomography, we observed a small calcified lesion in the right insular area (Fig. 1A); on brain magnetic resonance images, we observed a right insular cortex calcified lesion with partial volume loss (Supplementary Fig. 1). Video-EEG showed 2-Hz generalized triphasic waves and captured several episodes of electroclinical seizures originating in the right hemisphere (Fig. 1B). He was diagnosed with beta-lactam-induced encephalopathy with focal seizures and was treated with antiepileptic drugs and massive dialysis. His seizure stopped and he responded promptly. Approximately 1.5 days after seizure control, he stated, "I was already dead," and denied his existence for approximately one day. He then spontaneously recovered to his baseline condition.

1.2. Case 2

A 33-year-old right-handed female patient presented to our emergency department for acute-onset bizarre delusion. She exhibited magnetic resonance imaging-negative temporal lobe epilepsy (Fig. 1C) and was taking three antiepileptic drugs for seizure control. One day before presentation to the emergency department, she experienced several episodes of dialeptic seizures and one episode of focal to bilateral tonic-clonic seizure. After spontaneous cessation of seizures, there were no remarkable events until her family observed bizarre delusion. She denied her existence and the existence of her family. She stated "This is not me. My true self has died. My family members are not themselves, but are cyborgs resembling my family." Her statements were compatible with Cotard delusion and Capgras delusion. Her delusion persisted for more than one day, then disappeared spontaneously. Her interictal EEG after delusion resolution showed quasi-periodic sharp wave discharges over the right temporal area (Fig. 1D).

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https://doi.org/10.1016/j.seizure.2019.06.015
Received 22 April 2019; Received in revised form 11 June 2019; Accepted 12 June 2019
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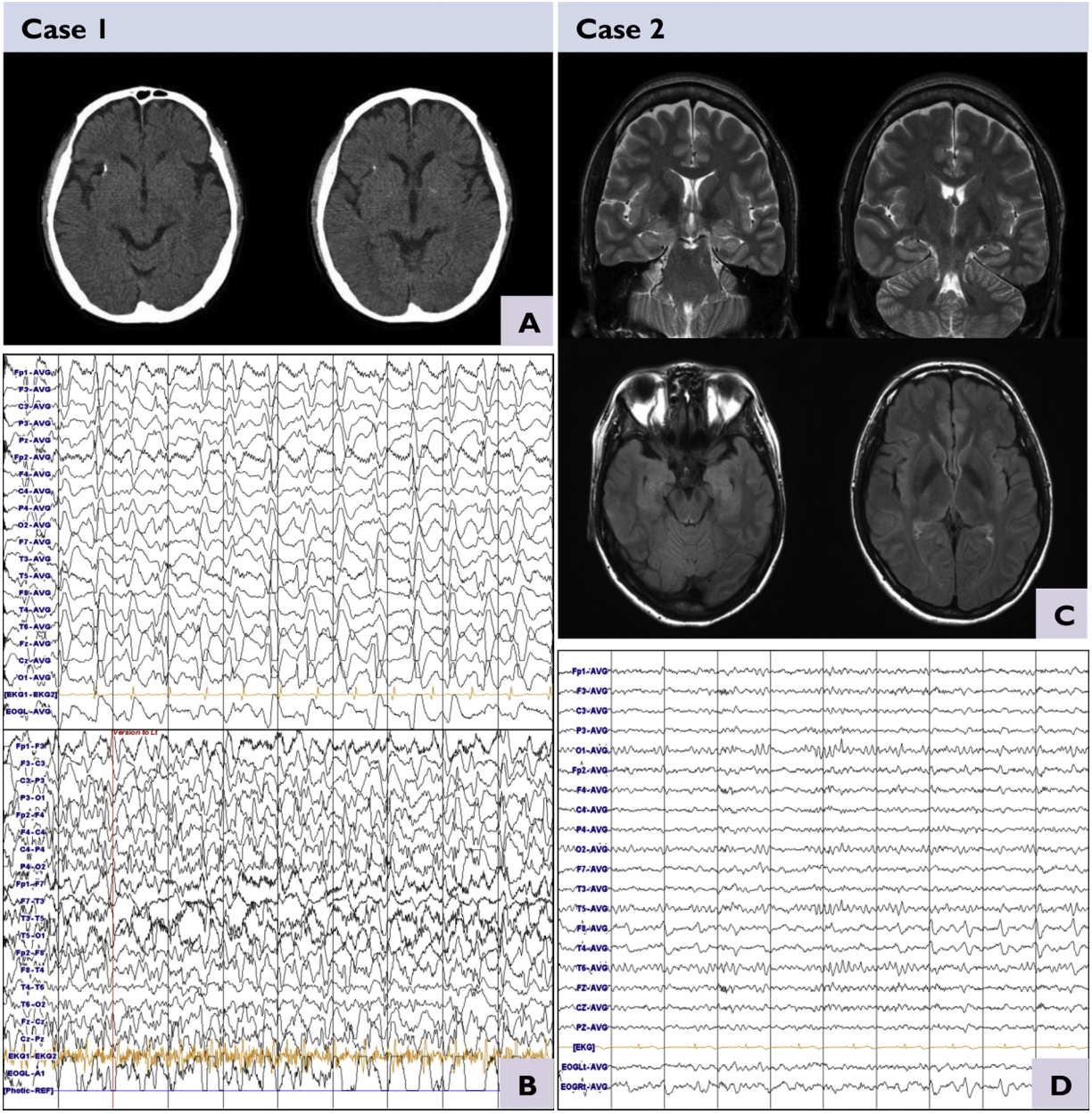

Fig. 1. Brain imaging and electroencephalogram of cases 1 and 2. A and B depict brain imaging and electroencephalogram (EEG) of case 1. Brain computed tomography showed small calcified lesion with volume loss on Rt. insular cortex (A). EEG showed 2-Hz generalized triphasic wave with ictal rhythm originating from the right hemisphere (B-upper: interictal, B-lower: ictal phase). C and D depict brain imaging and EEG of case 2. Brain magnetic resonance imaging performed after emergency department visit showed unremarkable abnormality (C). EEG after resolution of delusion showed quasiperiodic sharp wave discharges over the right temporal area (D).

2. Discussion

We have described two patients who exhibited focal or focal to bilateral tonic-clonic seizure of right hemispheric origin. Both patients showed normal appearance after seizure, known as “lucid interval.” However, the patient in case 1 exhibited Cotard delusion, denying his existence, while the patient in case 2 exhibited Capgras delusion and Cotard delusion. After symptoms lasting approximately one day, each delusion spontaneously disappeared.

In 1880, Jules Cotard described a middle-aged woman who denied existence of portions of her body. Subsequently, Cotard delusion was defined as delusional misidentification syndrome with nihilistic delusion, which presents with various degrees of severity (e.g., from mild desperate feeling to denial of one’s own existence). Capgras delusion is the belief that someone significant or well-known to an individual is no longer who that person was [3]. Cotard delusion and Capgras delusion are typically associated with psychiatric disease, but have been reported in patients with neurological disease.

Hypotheses vary regarding underlying causes of Cotard delusion and Capgras delusion, but are generally explained by two models. Notably, there is a two-factor model of delusional belief—experiential and inferential [4]. The first factor is unusual perceptual experiences due to dysfunction of neural pathways, such as frontal, temporal, and parietal lobes of the non-dominant hemisphere. Connections among occipital, parietal, and temporal cortices and the limbic system are also affected. The second factor is attributional style of experience. Patients with Cotard delusion choose an internalizing attributional style to interpret the same unusual emotional experience, while patients with

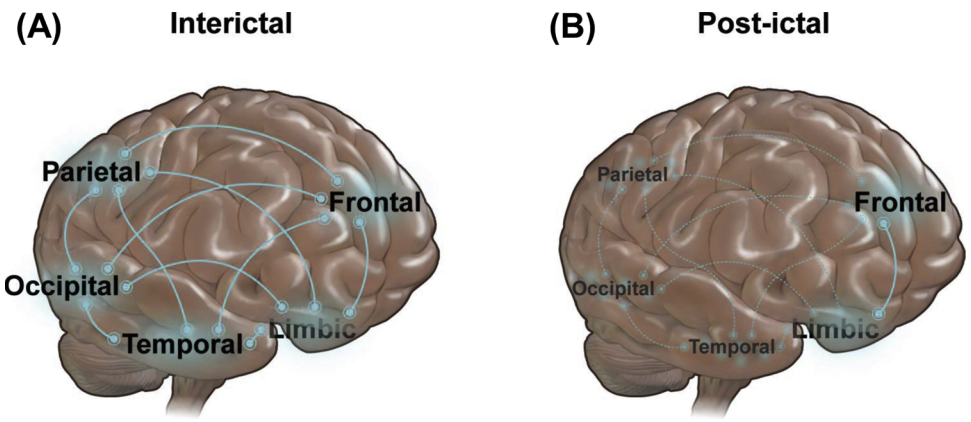

Fig. 2. Neuroanatomical circuit of Cotard and Capgras delusion. A. Interictal status of circuit activation. B. Post-ictal status. Cotard delusion is associated with right frontal, temporal, and parietal areas. The Capgras delusion is mainly explained by the lesion of the right hemisphere, which is related to the face recognition. After right hemispheric origin seizure, global dysfunction of the right hemisphere, as well as dysfunction of specific parts of the pathway among occipital, parietal, and temporal cortices, contribute to instability in the existing pathway in the association cortex. Functional disruption of the right hemisphere after seizure is represented by Cotard delusion, with or without Capgras delusion. In particular, abnormality around the limbic system, which is associated with familiarity and self-existence, is presumed to be the main pathognomic pathway of post-ictal delusion. Thick arrows: normal circuit; thin arrows: dysfunction of pathway.

Capgras delusion employ an externalizing attributional style. The other model is as follows: Capgras delusion may be due to disruption within facial recognition pathways, while Cotard delusion may be caused by global disconnection of all sensory areas from the limbic system.

Overall, neuroanatomical lesions for Cotard delusion with or without Capgras delusion often involve a series of pathways associated with the non-dominant or right frontal, temporal, and parietal areas in many patients. Most patients who exhibit Cotard delusion in combination with neurologic disease show right hemispheric lesions caused by various insults, such as cerebral infarction, brain tumor, arteriovenous malformations, or intracranial hemorrhage; left hemispheric lesions have also been reported. In patients with Capgras delusion, unilateral right hemisphere lesions are observed more frequently than left hemisphere lesions. In addition to structural neurologic disease, there have been case reports of Cotard delusion with or without Capgras delusion with epilepsy; however, most older reports lack detailed imaging and EEG data.

Cotard delusion with or without Capgras delusion in both patients in this report is consistent with post-ictal delusion criteria: a series of symptoms that occur in association with seizure, following a brief lucid period, which last approximately one day and cannot be explained by another etiology. The mechanism of post-ictal delusion, particularly in patients with temporal lobe epilepsy, is explained by disinhibition of the limbic system. Our patients showed Cotard delusion with or without Capgras delusion after focal seizures originating from right hemisphere. Global disconnection of the right hemisphere, as well as dysfunction of specific portions of the pathway among occipital, parietal, and temporal cortices, could be associated with post-ictal delusion, especially Cotard delusion. Right hemisphere dysfunction after seizure may be represented by Cotard delusion with or without Capgras delusion (Fig. 2). Our findings suggest a potential mechanism for post-ictal Cotard delusion and indicate the lateralizing significance of post-ictal Cotard delusion originating from the non-dominant right hemisphere.

Declaration of Competing Interest

The researchers claim no conflicts of interest.

Funding acknowledgments

This research was supported by the National Research Foundation (NRF) funded by the Korean government (MSIT) (NRF-2018M3A9E8023853, NRF-2018R1C1B6006145 and NRF-2019R1A5A2026045).

Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:https://doi.org/10.1016/j.seizure.2019.06.015.

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