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Case Report

Fantastic confabulation in right frontal lobe epilepsy

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ABSTRACT

Background: Interictal behavioral symptoms in frontal lobe epilepsy (FLE) are variable and often difficult to discriminate from other localization-related epilepsies.
Methods and results: We report two female patients with right FLE who exhibited fantastic confabulations. One of the patients had a 14-year history of hypermotor seizures, and the other had a 10-year history of dyscognitive seizures with automatism. Their fantastic confabulations arose in the context of moderate-to-severe cognitive impairment and of a variety of behavioral abnormalities, including emotional withdrawal and compulsive behaviors.
Conclusion: Fantastic confabulations are rare but may be a relatively specific behavioral marker for FLE-associated psychosis.

1. Introduction

A wide variety of ictal and interictal symptoms have been documented in frontal lobe epilepsy (FLE), which reflects the functional heterogeneity of the frontal lobe. The frontal lobe comprises several motor areas and regions associated with cognition and behavior. The latter is largely divided into three parts: the dorsolateral prefrontal cortex, the medial prefrontal/anterior cingulate cortex, and the orbitofrontal cortex [1]. Damage to each region has been associated with executive cognitive dysfunction, volitional/motivational impairment, and impulsive and compulsive behaviors, respectively. Most cases with FLE present with diffuse, mild underlying neural damage. As a result, the interictal cognitive and behavioral symptoms are typically diminished and mixed and, therefore, have little diagnostic value in differentiating FLE from temporal lobe epilepsy (TLE) or other types of localization-related epilepsies [2,3]. At the individual patient level, however, cognitive and behavioral features can be important clues in identifying epilepsy focus. Fantastic confabulation is a specific type of confabulation characterized by implausible bizarre descriptions of false realities. This condition arises in association with frontal lobe dysfunction in Wernicke–Korsakoff syndrome, after subarachnoid hemorrhage associated with the rupture of anterior communicating artery aneurysms, and other conditions [1,4,5]. Here, we report two patients with right FLE who exhibited fantastic confabulations during interictal periods. The diagnostic utility and possible underlying mechanisms of this symptom will be discussed in relation to FLE-associated psychosis.

2. Case presentation

All procedures in this study followed the clinical study guidelines of Tohoku University Hospital and were approved by its ethics committee. All patients and their caregivers provided written informed consent.

2.1. Case 1

A right-handed female with no family history of neurological or psychiatric diseases experienced her first hypermotor seizure during sleep at the age of six. Her seizures resolved after 2–3 months with medical treatment. At the age of 11, her hypermotor seizures recurred and were frequently followed by secondary generalization. Although her cognitive and psychological development was normal until the recurrence of seizures, her school performance gradually declined and interpersonal conduct deteriorated after the age of 12. She became socially withdrawn and indifferent to her family and friends, and her behavior became repetitive and stereotypic. Her interest focused on a few specific things, such as reading and drawing cartoons.

At the age of 20, she was referred to our epilepsy monitoring unit (EMU) for a comprehensive assessment. Interictal EEG revealed generalized intermittent rhythmic slow activity without clear

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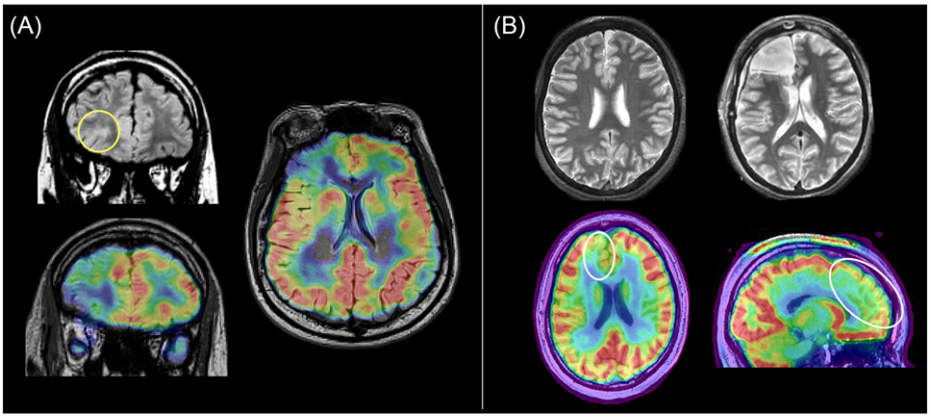

Fig. 1. Neuroimaging findings. (A) Case 1. A coronal attenuation inversion recovery (FLAIR) image shows a transmantle sign in the ventrolateral portion of the right frontal lobe (highlighted in yellow circle). Extensive hypometabolism is evident in the right frontal lobe on FDG-PET. (B) Case 2. Presurgical and postsurgical axial T2-weighted images are shown in the upper left and upper right, respectively. Axial and sagittal FDG-PET images (shown in the lower row) show hypometabolism in the right medial frontal cortex in figure 1B (highlighted in white circle).

epileptiform discharges. Her habitual hypermotor seizures were captured during sleep with nonlocalizable EEG seizure patterns during video-EEG monitoring. Magnetic resonance imaging (MRI) revealed a transmantle sign in the right frontal lobe, suggesting focal cortical dysplasia (Fig. 1A). 18F-fluorodeoxyglucose positron emission tomography (FDG-PET) showed extensive hypometabolism in the right frontal lobe (Fig. 1). Based on these findings, a diagnosis of right FLE was made. Her verbal intelligence quotient (VIQ) and performance intelligence quotient (PIQ) on the Wechsler Adult Intelligent Scale (WAIS)-III were 73 and 76, respectively. On the Wechsler Memory Scale — Revised (WMS-R), she scored 74, 93, and 73 on the verbal memory index, visual memory index, and delayed recall index, respectively. Abundant confabulatory responses were noted on the picture naming test, in which she invented new words to denote common objects in an impromptu manner. During interviews, she was easily distractible, inappropriately changed topics at her will, and only responded to topics that interested her. She never interacted with other patients yet did not seem distressed from social isolation over the course of her hospital stay. Her affect was flat, and her mood was neither depressed nor manic. In her free time, she spent a significant amount of time drawing cartoon characters, which cover each page entirely in a notebook (Fig. 2). She frequently produced confabulations in response to questions by hospital

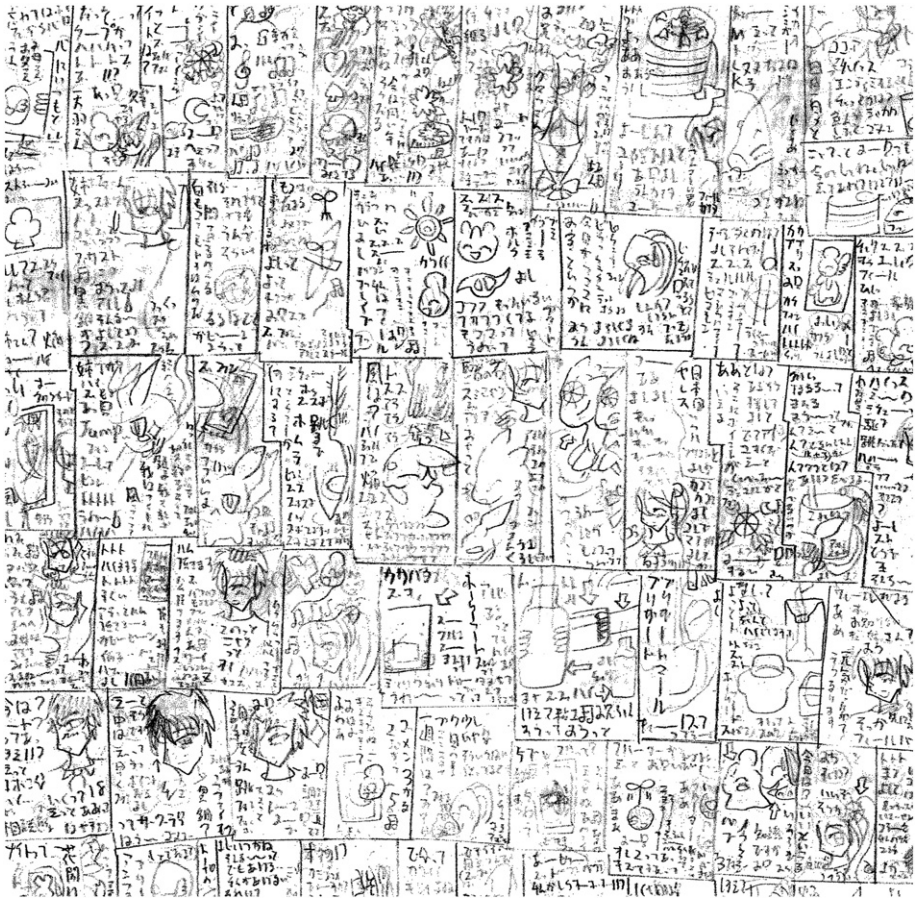

Fig. 2. Compulsive drawing in Case 1. Characters from Japanese cartoons and words written in small letters cover the entire width of the page. Most words are onomatopoeic.

staff. For instance, she said that she came to our hospital to see her previous counselor, whom she falsely insisted was secretly working in our hospital. When asked why she withdrew indoors and no longer went outside, she responded, "It's because of exhaust gas in the air." The themes of her remarks varied each time, suggesting that she had no firm delusional beliefs.

2.2. Case 2

A right-handed female with a history of febrile seizures and no family history of neurological or psychiatric diseases experienced her first seizure at the age of 13. Her initial seizure was a dyscognitive seizure with automatism and secondary generalization. Despite the treatment with valproic acid, carbamazepine and zonisamide, her seizures persisted weekly. At the age of 16, she developed monology and impulsivity and received an inpatient psychiatric treatment. Even after the discontinuation of zonisamide, her behavioral problems persisted.

At the age of 23, she was referred to our EMU for presurgical evaluation of epilepsy. Interictal EEG showed right frontotemporal and generalized spikes. Her habitual dyscognitive seizures with automatism were captured with right frontotemporal onset on video-EEG. Magnetic resonance imaging disclosed no structural abnormalities. 18F-fluorodeoxyglucose positron emission tomography revealed extensive hypometabolism in the right frontal cortex (Fig. 1). Her full IQ was below 50 on the shortened version of WAIS. On WMS-R, she scored 84, 58, and 78 on the verbal memory index, visual memory index, and delayed recall index, respectively. During neuropsychological testing, she was cooperative but quickly responded to questions without considerable thought. She was emotionally flat and indifferent to others. Abundant confabulatory responses were observed on conventional neuropsychological assessments. To further investigate her confabulation, we administered the Confabulation Battery [6]. She made a significant number of confabulations in response to the "I don't know" semantic and episodic questions. For example, in response to the question, "How far is it from the earth to Saturn?", she quickly responded with "one kilometer." In response to the question, "Did you go to the station in early September, 2004 (five years before the examination)?", she answered, "In 2004, I went to the station. (For what?). For shopping. (The weather?) Fine, I think."

She was diagnosed with right FLE and underwent a right frontal lobectomy after intracranial EEG confirmed the ictal-onset zone. Although no significant cognitive deterioration was observed, her confabulation disappeared after surgery, despite persistent monthly seizures.

3. Discussion

Confabulation covers a wide variety of qualitatively different memory-related phenomena from mild memory distortions to implausible, bizarre descriptions of false realities [7]. Our patients' behavior corresponded to the latter type of confabulation, which is called fantastic confabulation. Although, by definition, confabulations and delusions differ in the absence or presence of firm beliefs regarding false realities, there are phenomenological and neurological similarities between fantastic confabulations and delusions. Both conditions involve implausible and, often, bizarre narratives and the failure to correct false reports in the face of contradictory evidence. Frontal lobe dysfunction has been implicated in both fantastic confabulations and delusions [5,8], and they occasionally coexist in patients with frontal lobe lesions [9]. These facts suggest that fantastic confabulation may be a psychosis-related symptom akin to delusion [10].

Several lesion analysis studies demonstrated that orbitofrontal or medial ventral frontal damage was predictive of the emergence of confabulations [5,11]. However, confabulations occasionally occur in association with hypothalamic or thalamic lesions [12,13]. This suggests that orbitofrontal dysfunction associated with frontal–subcortical circuitry disruption, not the structural frontal damage itself, plays a pivotal role in causation. The severity and bizarreness of confabulations correlate with the extensiveness of lesions and the severity of cognitive dysfunction [11,14]. In agreement with the above findings, our two cases indicated extensive frontal dysfunction in FDG-PET, and moderate-to-severe impairments on multiple domains of neuropsychological examination. In addition, both of our patients developed fantastic confabulations with a variety of behavioral abnormalities including emotional withdrawal, blunted affect, and stereotyped behaviors [1]. While these behavioral symptoms have been associated with frontal lobe dysfunction, they can be observed in both FLE- and TLE-associated psychoses [15,16]. We suggest that fantastic confabulations may be a rare but relatively specific behavioral marker for FLE-associated psychoses. This prediction should be further investigated through systematic studies.

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Conflicts of interest statement

The authors declare that there are no conflicts of interest.

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