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Case Report

Dreamy State, Delusions, Audiovisual Hallucinations, and Metamorphopsia in a Lesional Lateral Temporal Lobe Epilepsy Followed by Ipsilateral Hippocampal Sclerosis

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Keywords
Dreamy state · Delusions · Audiovisual hallucinations · Metamorphopsia · Lesional lateral temporal lobe epilepsy · Hippocampal sclerosis

Abstract
We report a 65-year-old man who was diagnosed with focal status epilepticus generating a dreamy state, delusions with anxiety, complex audiovisual hallucinations, elementary auditory hallucinations, and metamorphopsia with a growing large lateral temporal lobe lesion. After administrating anti-seizure drugs, all the symptoms disappeared, and brain magnetic resonance imaging revealed ipsilateral hippocampal sclerosis. To the best of our knowledge, this is the first report to present all the symptoms in one epilepsy case. On the basis of semiology, electroencephalography, and brain magnetic resonance imaging, we speculated that epileptic activities that have originated from the lateral lesion might have propagated to the ipsilateral mesial temporal lobe, causing hippocampal sclerosis.

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Introduction

Temporal lobe epilepsy (TLE) is common, the estimated prevalence of which is 1.7 per 1,000 people [1]. Eighty percent of TLE is of hippocampal onset, which is characterized as mesial TLE [2]. Compared to mesial TLE, however, fewer reports are available on lateral TLE, especially lesional TLE [3, 4], although discrete lesions in the temporal lobe tend to be more frequently on the lateral side than on the mesial side [3]. In addition, not only mesial TLE but also lateral TLE develops psychic symptoms, such as elementary auditory hallucination; however, few reports are available on psychic symptoms related to lateral lesional TLE. To recognize these psychic symptoms as epilepsy correctly, it is necessary to accumulate evidence on the relationship between symptoms and findings of electroencephalography (EEG) and magnetic resonance imaging (MRI) of the patients.

Here, we report a case of epilepsy that generated a dreamy state, delusions with anxiety, complex audiovisual hallucinations, elementary auditory hallucinations, and metamorphopsia with a large lateral temporal lobe lesion followed by ipsilateral hippocampal sclerosis. To our knowledge, this is the first report of a case that presented with all the symptoms in one epilepsy case.

Case Report

A 65-year-old right-handed man was admitted to our hospital because of a sudden and intermittent dreamy state, delusions with anxiety, complex audiovisual hallucinations, elementary auditory hallucinations, and metamorphopsia. Nine years before admission, he developed focal to bilateral tonic-clonic seizures. A lesion in the right lateral temporal lobe with calcification was pointed out, and valproic acid (VPA) 800 mg/day had been administered. Seven years before admission, the lesion had flow voids and started to grow. The lesion's size was 10 mm in the major axis and 7 mm in the minor axis in the axial slice at the midbrain level, but VPA was reduced to 600 mg/day by the patient. Three days before admission, he suddenly started to experience weird sensations intermittently and randomly for several minutes or hours without obvious loss of consciousness . He had a sensation that he went to another unfamiliar world. He felt that a thief had broken into his room and that someone was peeping at him from behind. He heard voices and ringing of a bell. He saw a cat on the wall. In addition, he felt that objects, including faces, looked swollen, and it happened more frequently with objects in the left than with those in the right visual field. The most annoying sensation for him was hearing the ringing of a bell to the extent that he had difficulty in falling asleep.

On admission, his body temperature was 36.3°C, blood pressure was 131/89 Torr, and heart rate was 62 beats/min and regular. Neurological examination showed that his consciousness was alert, and the Mini-Mental State Examination score was 29/30, with one point loss in calculation. The patient's drawing of a cuboid plastic bottle was swollen compared to the original shape (Fig. 1). He had no prosopagnosia based on the identification test of faces such as the Prime Minister of Japan and the President of the USA. Ophthalmological examinations, such as visual acuity test and fundoscopy test, were normal, except that Goldmann's perimetry test showed a slight reduction of sensitivity in the left homonymous visual field, suggesting impairment of the function between the right optic tract and the occipital lobe. Otolaryngological examinations, such as pure tone and speech audiometry tests, were unremarkable.

We considered his experiences that "he went to another unfamiliar world" as dreamy state, "he felt that a thief had broken into his room and that someone was peeping at him from behind" as delusions with anxiety, "he heard voices and ringing of a bell" as complex auditory hallucination and elementary auditory hallucination, "he saw a cat on the wall" as complex visual hallucination, and "he felt that objects, including faces, looked swollen" as metamorphopsia.

Routine blood examinations were unremarkable, except that serum VPA concentration was 9 μg/ml, which was below the therapeutic level. Cerebrospinal fluid examinations, including cytology, were normal. EEG showed right-dominant diffuse delta or theta waves on basic activity (Fig. 2a). Brain MRI revealed that the lesion in the right lateral temporal lobe had grown up to 50 mm in the major axis and 20 mm in the minor axis in the axial slice at the midbrain level. The lesion was in the middle temporal gyrus and reached the border between the temporal and occipital lobes on axial T2 star-weighted imaging (Fig. 3a). In addition, abnormal hyperintensity in the right mesial temporal lobe emerged on T2-weighted imaging (T2WI), diffusion-weighted imaging (DWI; Fig. 3b), and fluid-attenuated inversion recovery (FLAIR; Fig. 3c, d).

On the basis of the sudden-onset and intermittent series of symptoms, insufficient serum VPA concentration, and the findings of MRI and EEG, we considered his symptoms as focal status epilepticus with awareness or impaired awareness. The lesion in the right lateral temporal lobe was considered as an old hemorrhage of which the pathology might be vascular malformation. Given the course of the MRI, cavernous angioma was most likely, except that no vascular flow void was seen on MRI on admission. Abnormal hyperintensity in the right mesial temporal lobe was considered to be a secondary alteration caused by an epileptic seizure.

After administrating levetiracetam 1,000 mg/day and increasing VPA to 800 mg/day on day 1, delusions with anxiety and complex auditory hallucinations disappeared on day 3, and slow waves in EEG on day 8 were markedly reduced (Fig. 2b). Single-photon emission computed tomography (SPECT) using *N*-isopropyl-[^{123}I]-*p*-iodoamphetamine on day 6 revealed no hyperperfusion area but a hypoperfusion area in the right temporal cortex, which could correspond to the old hemorrhage. After administrating lacosamide 100 mg/day on day 8, the dreamy state disappeared, and abnormal hyperintensity in the right mesial temporal lobe on DWI, T2WI, and FLAIR in MRI was reduced on day 9. Elementary auditory hallucinations, complex visual hallucinations, and metamorphopsia disappeared on day 12. The patient was discharged on day 18 and restarted paper work as before. Eight months after discharge, MRI showed atrophy of the right mesial temporal lobe (Fig. 3e, f), suggesting hippocampal sclerosis. Finally, we diagnosed him with focal status epilepticus with impaired awareness or awareness, related to the vascular malformation in the right lateral temporal lobe and secondary ipsilateral hippocampal sclerosis.

Discussion

We reported a case of epilepsy that generated focal status epilepticus with impaired awareness or awareness with a right lateral temporal lobe lesion followed by ipsilateral hippocampal sclerosis. We speculated the following mechanism.

In general, seizure onset zones that originated from epileptogenic lesion and symptomatogenic zones do not always match because epilepsy has the nature of propagation based on epilepsy networks. In addition, scalp EEG and SPECT only provide a presumable irritable

zone, functional deficit zone, or symptomatogenic zone, not a seizure onset zone. Therefore, the detection of the detailed seizure onset zone or propagation process of this case without long-term video-intracranial EEG monitoring was impossible. However, known symptomatogenic zones corresponding to each symptom as follows and MRI findings could provide clues to presume the mechanisms of epilepsy of this case.

A dreamy state is related to temporal lobe activity [5, 6]. Delusions are related to the mesial and lateral temporal lobes [5] of the minor hemisphere. Complex hallucinatory states such as complex audiovisual hallucinations are considered to involve the limbic cortex as a prerequisite [7]. Complex auditory hallucinations are related to temporal auditory association cortices in the lateral temporal lobe [8]. Complex visual hallucinations are related to the boundaries of parietal, occipital, and temporal lobes of the minor hemisphere [8], which contain the visual association cortex and limbic system [9]. Elementary auditory hallucinations are related to the primary auditory cortices, that is, the transverse temporal gyrus in the superior temporal gyrus [10]. Metamorphopsia is a rare symptom, and its symptomatogenic zone is controversial among the temporal lobe [11], occipital lobe [12], parietal lobe [11], and both the occipital and temporal lobes [12]. In addition, a slight reduction of sensitivity in the left homonymous visual field on admission and higher frequency of metamorphopsia in the left than in the right visual field suggested the possibility of impairment of the function between the right optic tract and the occipital lobe; however, the visual field test was not followed after the administration of anti-seizure drugs.

The most anatomically specific symptom in this case was elementary auditory hallucination [7, 9], which was one of the symptoms that persisted from the beginning to the end of the course of the series of symptoms. The transverse temporal gyrus, being adjacent to the middle temporal gyrus where the lesion was mainly located, was speculated to be the surest symptomatogenic zone.

We considered two hypotheses of the mechanisms of epilepsy of this case. First, epileptogenicity originated from one lesion: epileptogenicity was generated from the large lateral lesion on the back of the growing lesion and accelerated by the reduction of the anti-seizure drug, because in general epileptogenicity tends to be generated in the area adjacent to the lesion based on the local neuronal injury, glial proliferation, vascular changes, and neurotransmitter changes [4]. In this case, hemosiderin deposition of the lesion might be a main epileptogenicity. In addition, it has been reported that many clinical seizures were recorded from the lateral cortex of the lesional temporal lobe using long-term intracranial EEG [4]. The epileptic activities quickly propagated to the ipsilateral mesial temporal lobe [13, 14], and the propagation caused the excitotoxic neuronal cell death [15] of the ipsilateral mesial temporal lobe because the hippocampal sites, in particular, in the ipsilateral mesial temporal lobe have a lower threshold of excitability. Through the propagation, epileptic activities might have spread to a part of the right temporal, parietal, and occipital lobes, as suggested by the findings of EEG on admission, and extended to symptomatogenic zones, as implied by the series of symptoms. Second, epileptogenicity originated from two regions: we cannot deny the possibility that the mesial structure might have had primary epileptogenicity by nature or gained epileptogenicity independently from the old hemorrhage. Anyway, epileptic activities that originated from the considerably large lateral temporal lesion under the administration of an anti-seizure drug might be the reason for the various symptoms without loss of consciousness or generalized tonic-clonic seizure. If we could perform long-term intracranial EEG on this case, we could answer the questions of which hypothesis was correct and whether this case had dual pathology [15] or not.

In conclusion, we reported the first case that presented with a dreamy state, delusions with anxiety, complex audiovisual hallucinations, elementary auditory hallucinations, and metamorphopsia as focal status epilepticus with impaired awareness or awareness, of which epileptogenicity presumably originated from the lesion with considerable hemosiderin deposition in the right lateral temporal lobe. Further studies were needed to prove epileptogenicity using not only scalp EEG and SPECT but also 18-fluorodeoxyglucose-positron emission tomography on the early days after admission. In addition, to clarify the detailed propagation process or the range of the spread of the epileptic activities, video EEG monitoring, continuous EEG, or intracranial EEG were needed. However, elementary auditory hallucination, which has high specificity on the symptomatic zone, and hippocampus sclerosis, which was revealed after the symptoms were diminished, were the important clues to presume the mechanisms of the epilepsy of this case.

Statement of Ethics

Informed consent was obtained from the subject.

Disclosure Statement

The authors declare no conflict of interest.

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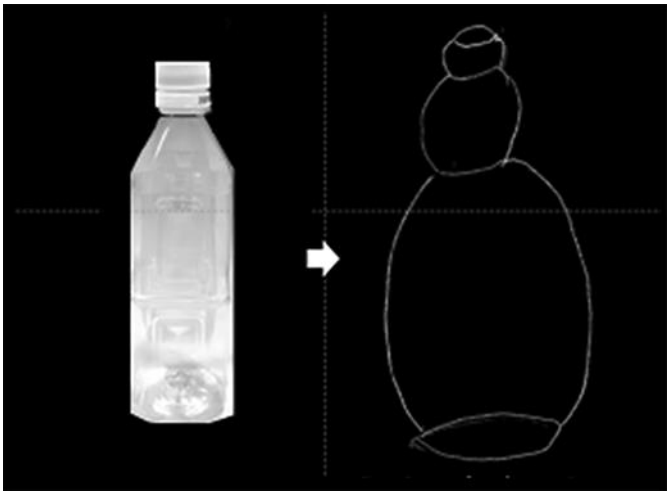

Fig. 1. Metamorphopsia drawn by the patient. The patient's drawing of a cuboid plastic bottle (right) was swollen compared to the original shape (left).

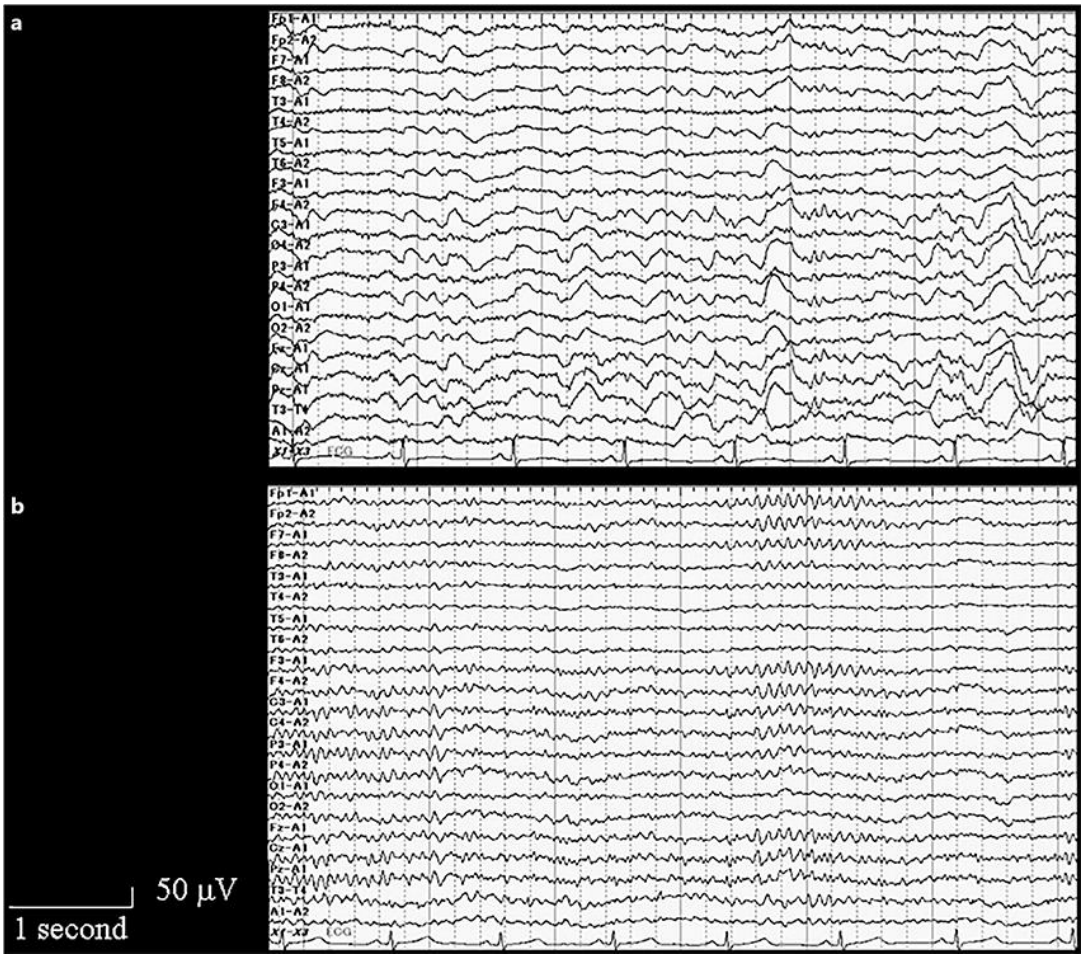

Fig. 2. EEG on admission and after increasing the anti-seizure drugs. **a** Monopolar recording on admission. Continuous right-dominant diffuse delta or theta waves on basic activity were noted. **b** Monopolar recording on day 8, after increasing the anti-seizure drugs. Slow waves were markedly reduced.

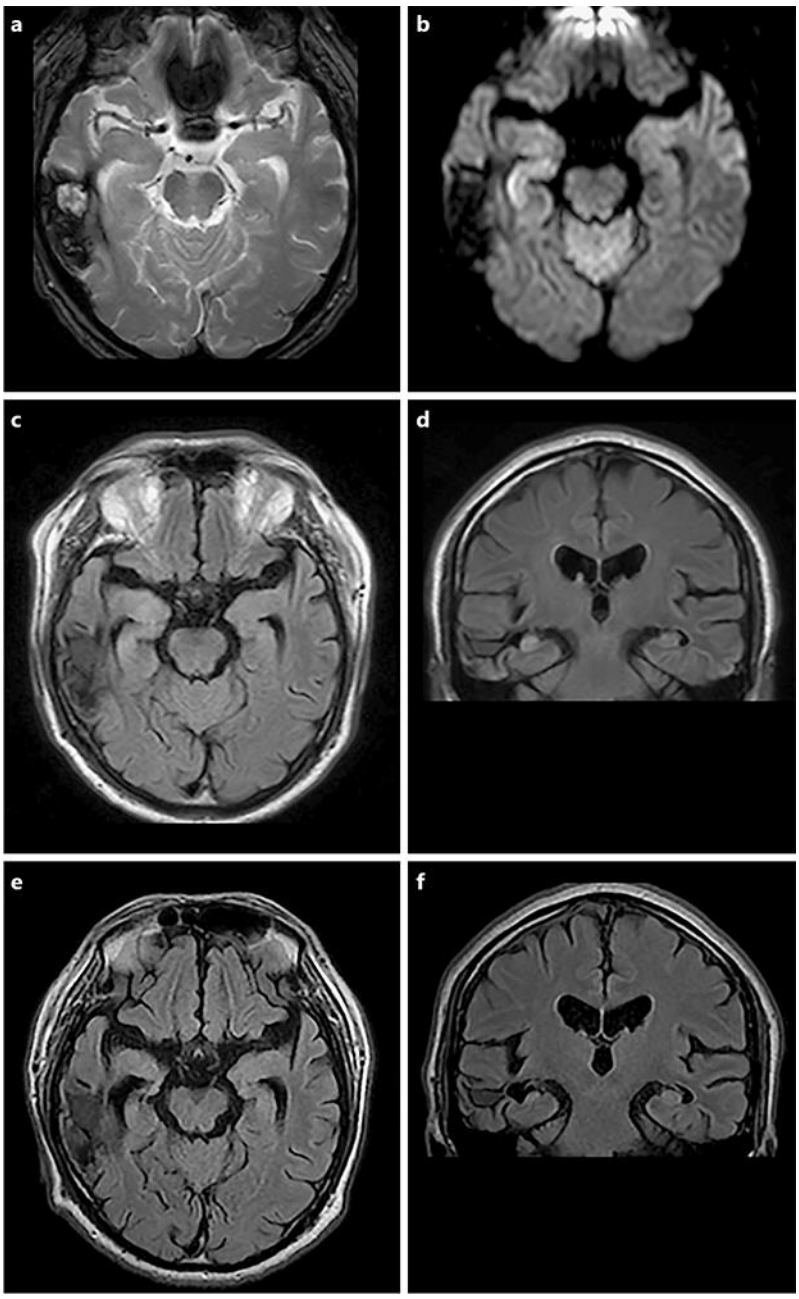

Fig. 3. MRI on admission and 8 months after all the symptoms had disappeared. **a** On admission. Axial T2 star-weighted image illustrating the right lateral temporal lobe oval lesion, which was 50 mm in the major axis and 20 mm in the minor axis, mainly in the middle temporal gyrus in the axial slices at the midbrain level, accompanying a cyst of 13 mm in diameter. **b** On admission. Axial DWI illustrating abnormal hyperintensity in the right mesial temporal lobe. **c**, **d** On admission. Axial (**c**) and coronal (**d**) FLAIR images illustrating abnormal hyperintensity in the right mesial temporal lobe and abnormal hypointensity in the right middle temporal gyrus. **e**, **f** Eight months after all the symptoms had disappeared. Axial (**e**) and coronal (**f**) FLAIR images illustrating atrophy of the right mesial temporal lobe.