

Oral Cenesthopathy Proceeding Dementia With Lewy Body: A Case Report

Yuta Sawada

Sangyo Ika Daigaku

Yuki Konishi

Sangyo Ika Daigaku

Atsuko Ikenouchi

Sangyo Ika Daigaku

Reiji Yoshimura (✉ yoshi621@med.uoeh-u.ac.jp)

University of Occupational and Environmental Health

Case report

Keywords: DLB, case report, hallucination, Oral.

DOI: <https://doi.org/10.21203/rs.3.rs-60931/v1>

License:   This work is licensed under a Creative Commons Attribution 4.0 International License.

[Read Full License](#)

Abstract

Somatic hallucination is defined as the false sensation of an occurrence in the body. When the hallucination is grotesque and visceral, it is classified as a cenesthopathy (1). Cenesthopathy is likely influenced by culture; one study reported that, of the seven unique countries from which patients were recruited, those from Ghana and those with chronic schizophrenia were significantly more likely to report cenesthesia (2).

Introduction

Somatic hallucination is defined as the false sensation of an occurrence in the body. When the hallucination is grotesque and visceral, it is classified as a cenesthopathy (1). Cenesthopathy is likely influenced by culture; one study reported that, of the seven unique countries from which patients were recruited, those from Ghana and those with chronic schizophrenia were significantly more likely to report cenesthesia (2).

Dementia with Lewy body (DLB) is a common cause of dementia in the elderly, accounting for 15–25% of dementia cases (3). Evidence suggests DLB may be underdiagnosed, often being mistaken for mood disorders (4). Some patients are even misdiagnosed with mood disorders or late-onset schizophrenia and treated with antidepressants or antipsychotics (4), which may subsequently worsen parkinsonism. A clinical distinction is important, as it has profound implications for management and prognosis.

We present herein a case of persistent cenesthopathy preceding DLB that was successfully treated with donepezil and levodopa.

Case Presentation

A 70-year-old female retired high school teacher presented with complaints of a sensation of her teeth melting and swelling in her gums. At 68 years of age, she consulted a dentist due to the feeling of incongruity in her oral cavity. Although her oral discomfort transiently improved after the oral care instructed by the dentist, the symptoms soon relapsed and persisted. She was diagnosed with major depression at an outpatient's clinic, treated with paroxetine (20 mg/day), and referred to our university hospital. Her psychiatric symptoms included oral cenesthopathy, depressed mood, persecutory delusion, auditory hallucination, and impaired attention and concentration. Her diagnosis was changed to late-onset schizophrenia, and she was treated with aripiprazole (24 mg/day) and paroxetine (20 mg/day). Her psychiatric symptoms were relieved. Six months later, she again presented with oral cenesthopathy accompanied by persecutory delusion, depressed mood, attention deficits, and anxiety. Aripiprazole was tapered off, and perospirone was gradually increased to 36 mg/day. She then presented with parkinsonism, including finger tremor, muscle rigidity at the upper and lower limbs, bradykinesia, hypersalivation, postural instability, orthostatic hypotension, constipation, and polyuria. Subsequently, she demonstrated rapid eye movement sleep behavior disorder. We reconsidered her diagnosis, and

suspected DLB. She retained independence for basic activities of daily living and had no trouble remembering remote past life events; memory problems were restricted to recent events. Her Mini-Mental State Examination score was 27. Magnetic resonance imaging (MRI), single photon emission computed tomography (SPECT), MIBG scintigraphy, and DAT scans were performed. The atrophy of medial central lobe was not pronounced in MRI (Fig. 1). MIBG scintigraphy showed a hypo-accumulation pattern (Fig. 2). Reduced uptake at the lateral basal ganglia was revealed in the DAT scan (Fig. 3). These findings supported the diagnosis of DLB. Perospirone and paroxetine were tapered off, and donepezil was started and increased to 5 mg/day. Four weeks later, the oral cenesthopathy, persecutory delusion, depressed mood, and cognitive flexibility were reduced; parkinsonism, however, persisted. Levodopa (75 mg/day) was added to the ongoing donepezil (5 mg/day), and her parkinsonism improved. Written informed consent was obtained from the patient for publication of this case report.

Discussion

This case sheds light on the rare psychotic symptoms that may occur before the development of DLB. Instead of memory deficits, it is common for the initial cognitive symptoms of DLB to be attention, executive, and visuospatial deficits (5). Psychiatric features are also present in DLB; visual illusion or hallucination, persecutory delusions, depressed mood, and mood fluctuation are often observed (6). Cenesthopathy presents with other psychiatric diseases, including schizophrenia, major depression, and somatoform disorders (7). A recent case report detailed a case of Ekbohm syndrome, which consists of the delusion of parasites accompanied by tactile hallucinations, associated with DLB (8). The authors reported rivastigmine, an acetylcholinesterase inhibitor, was effective, and no antipsychotic drugs were needed. Another case of delusional parasitosis in DLB was successfully treated with donepezil and aripiprazole (9). Umezaki et al. (10) first reported a case of oral cenesthopathy that developed into DLB, which was treated with rivastigmine. To the best of our knowledge, the case described herein was the second demonstrating oral cenesthopathy preceding DLB. Although the causal relationship between oral cenesthopathy and DLB remains unclear, close attention should be paid to late-onset psychotic symptoms as a DLB prodrome, even when cognitive impairment is not significant.

Declarations

Ethics approval and consent to participate

Written informed consent was obtained from the patient for publication of this case report.

*** Consent for publication**

Consent for publication was obtained from all authors.

*** Availability of data and material**

*** Competing interests**

The authors have no conflicts of interest to declare.

* Funding

This case report was performed without funding.

* Authors' contributions

YS was involved in designing the study, collecting the data, and writing the paper. YK and AI assisted with writing the paper. RY was involved in designing the study, supervising data collection, and writing the paper.

* Acknowledgements

None.

References

1. Kaplan HI, Sadock BJ. Comprehensive textbook of psychiatry. 5th ed. Baltimore: Williams and Wilkins; 1989.
2. Bauer SM, Schanda H, Karakula H, Olajossy-Hilkesberger L, Rudaleviciene P, Okribelashvili N, Chaudhry HR, Idemudia SE, Gscheider S, Ritter K, Stompe T. Culture and the prevalence of hallucinations in schizophrenia. *Compr Psychiatry*. 2011;52(3):319–25.
3. Vann Jones SA, O'Brien JT. The prevalence and incidence of dementia with Lewy bodies: a systematic review of population and clinical studies. *Psychol Med*. 2014;44:673–83.
4. Walker Z, Possin KL, Boeve BF, Aarsland D. Lewy body dementias. *Lancet*. 2015;386(10004):1683–97.
5. Mueller C, Ballard C, Corbett A, Aarsland D. The prognosis of dementia with Lewy bodies. *Lancet Neurol*. 2017;16(5):348.
6. Morenas-Rodríguez E, Sala I, Subirana A, Pascual-Goñi E, Sánchez-Saudinós MB, Alcolea D, Illán-Gala I, Carmona-Iragui M, Ribosa-Nogué R, Camacho V, Blesa R, Fortea J, Lleó A. Clinical subtypes of dementia with Lewy bodies based on the initial clinical presentation. *J Alzheimers Dis*. 2018;64(2):505–13.
7. Takahashi T, Fuke T, Washizuka S, Hanihara T, Amano N. A review of recent case reports of cenesthopathy in Japan. *Psychogeriatrics*. 2013;13(3):196–8.
8. Pontes de Mendonça FJ, Teixeira IA, Marinho V. Ekbom syndrome associated with Lewy body dementia: a case report. *Dement Neuropsychol*. 2020;14(1):83–7.
9. Ochiai S, Sugawara H, Kajio Y, Tanaka H, Ishikawa T, Fukuhara R, Jono T, Hashimoto M. Delusional parasitosis in dementia with Lewy bodies: a case report. *Ann Gen Psychiatry*. 2019;18:29.
10. Umezaki Y, Asada T, Naito T, Toyofuku A. A case of oral cenesthopathy in which dementia with Lewy bodies developed during treatment. *Psychogeriatrics*. 2020.

Figures

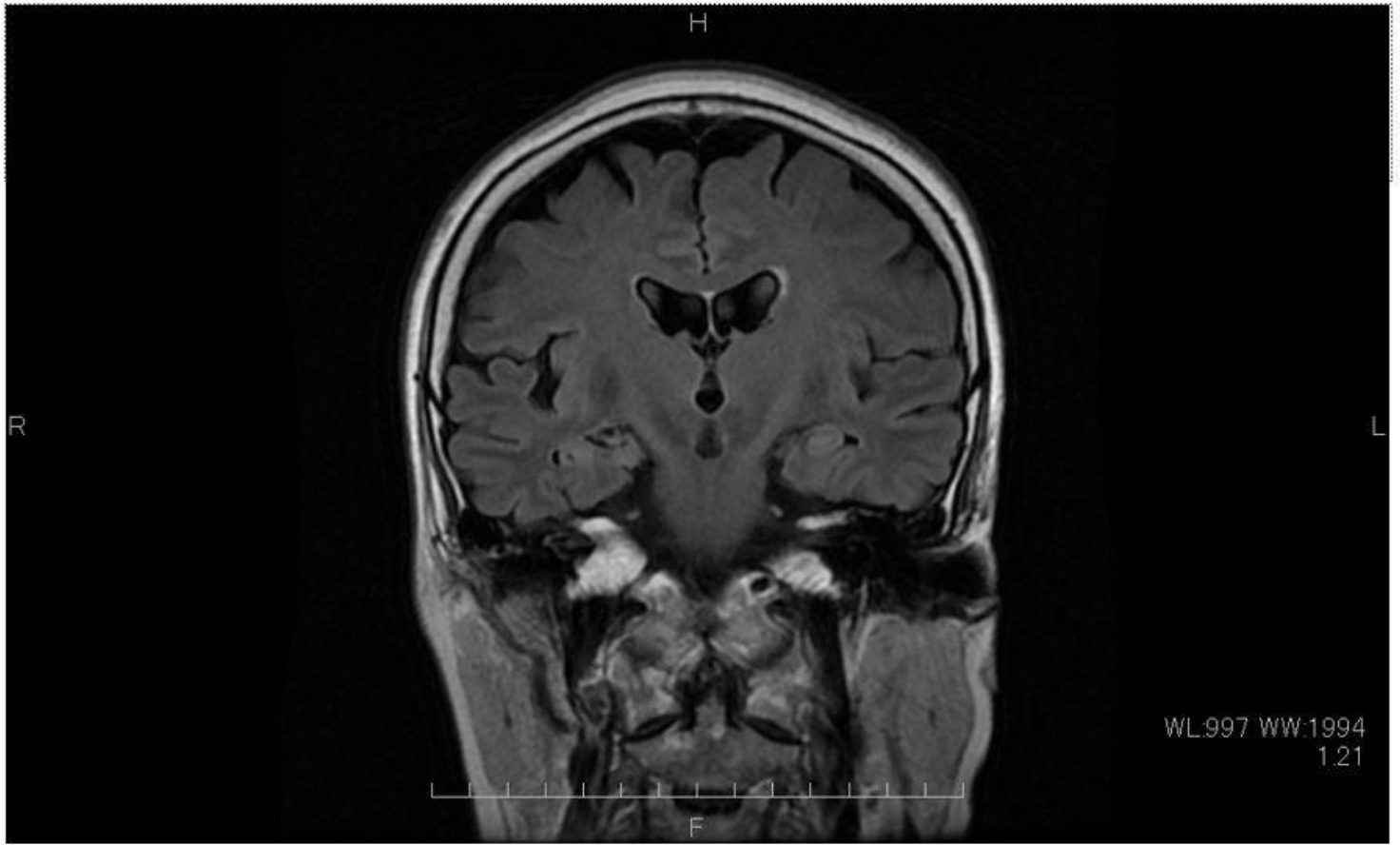


Figure 1

The patient's DAT scan reveals significantly reduced uptake at the bilateral basal ganglia.

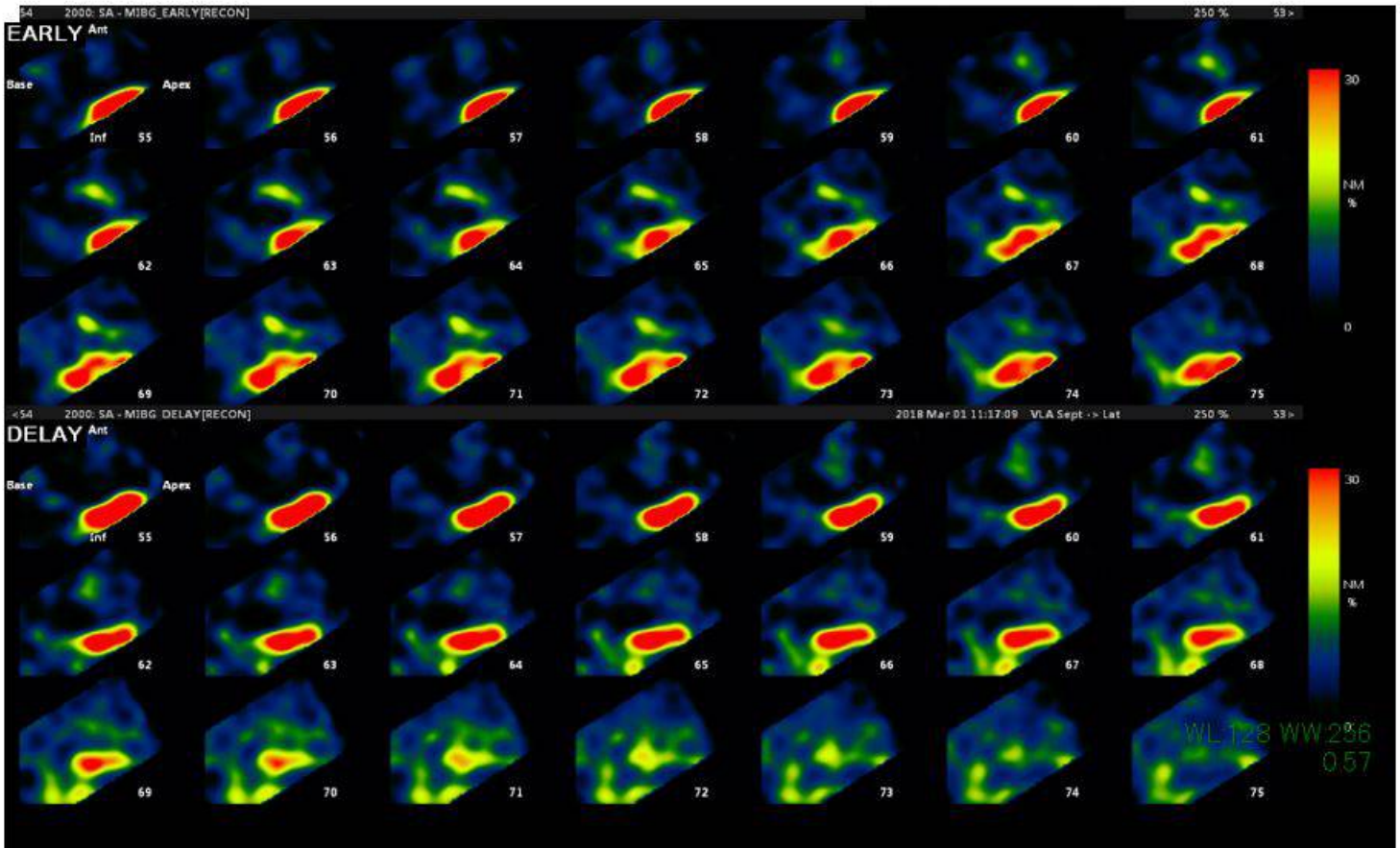


Figure 2

Figure 2

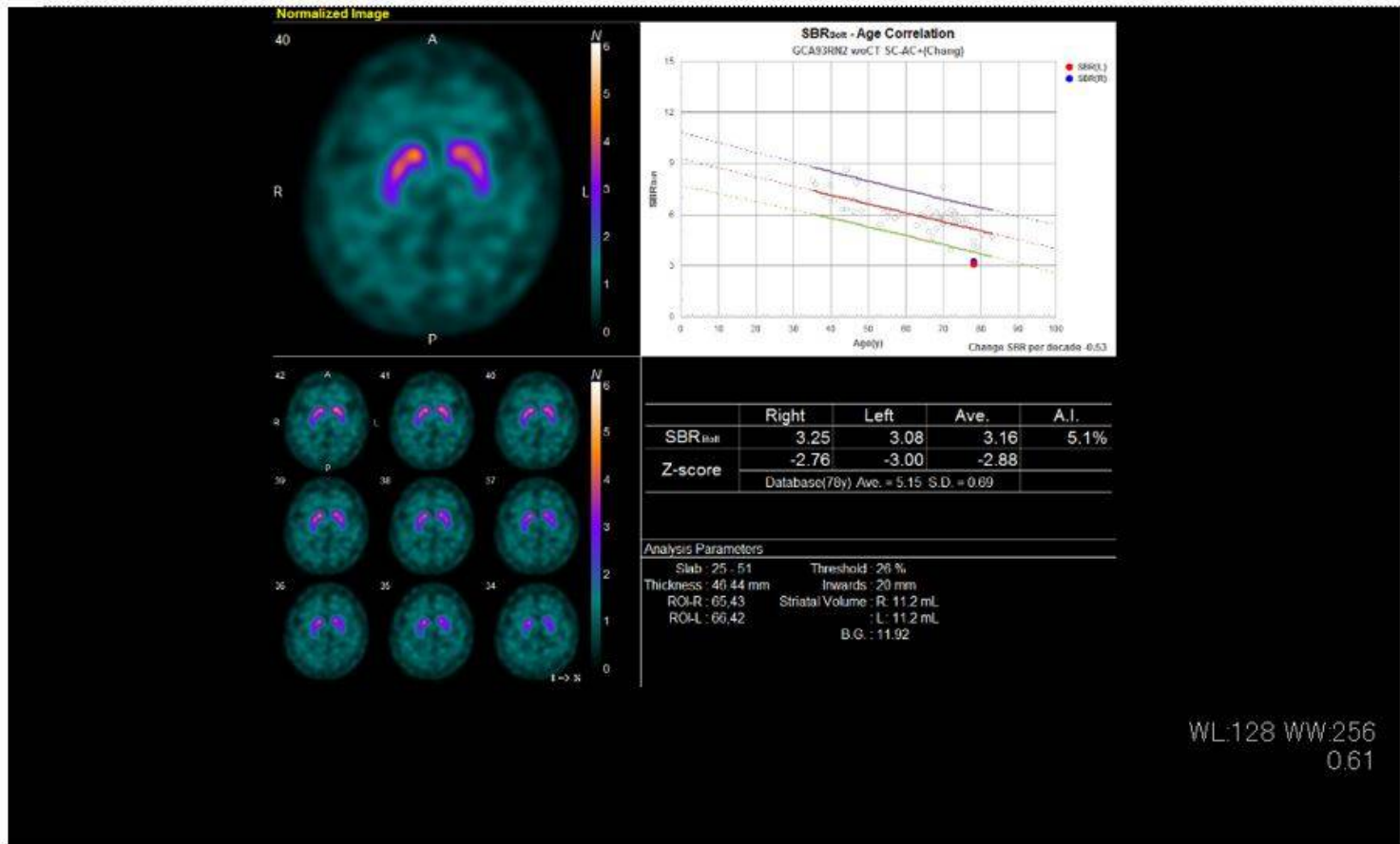


Figure 3

Figure 3