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Two Cases of Oral Somatic Delusions Ameliorated With Brain Perfusion Asymmetry: A Case Report

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Background: Oral cenesthopathy is the complaint of abnormal oral sensation where no underlying organic cause can be identified. It is also called oral dysesthesia or oral somatic delusion and classified as delusional disorder, somatic type. The patients with oral cenesthopathy show right > left asymmetric regional cerebral blood flow (rCBF) in the broad brain region. However, the studies scrutinizing the rCBF change before and after the successful treatment are still a few so far.

Case: We present 2 cases of oral cenesthopathy, who responded well to aripiprazole. The asymmetric rCBF patterns were attenuated after successful treatment in both cases.

Conclusions: We found a marked improvement of oral cenesthopathy with aripiprazole. It is suggested that right > left rCBF asymmetry in the frontal and temporal lobes and thalamus, and the dopaminergic and serotonergic dysfunctions are involved in the pathology of oral cenesthopathy.

Key Words: aripiprazole, cenesthopathy, oral DRS, oral somatic delusion, single-photon emission computed tomography

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Cenesthopathy is the complaint of abnormal bodily sensation where no underlying organic cause can be identified.¹ The oral area is the most affected region of cenesthopathy.² It is also called oral dysesthesia,³ oral parasitosis,⁴ or oral somatic delusion⁵ and classified as delusional disorder, somatic type, or somatic symptom disorder in the *Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition*.⁶ The complaints are various, sometimes understandable such as “sticky saliva in my mouth” or “feels bubbles around the teeth,” and sometimes bizarre such as “wires and coils are hanging on my teeth” or “my palate melts like a cheese.” Although the pathophysiology of oral cenesthopathy is still unclear, we recently reported that the patients with oral cenesthopathy showed right > left asymmetric regional cerebral blood flow (rCBF) in the broad brain region including the frontal and temporal lobes⁵ regardless of presence or absence of depression.⁷ In some case reports, although typical antipsychotics hardly improved the symptoms of oral cenesthopathy,² the right-side-predominant rCBF disappeared after successful treatment using paroxetine,⁸ risperidone,⁹ or modified electroconvulsive

therapy (mECT).^{10,11} However, the studies scrutinizing the rCBF change before and after successful treatment are still a few so far. We present 2 cases of oral cenesthopathy, who responded well to aripiprazole, a dopamine partial agonist, and whose asymmetric rCBF patterns were attenuated after successful treatment. Written informed consent was obtained from the patients before this presentation.

Case Presentation

Patient 1

A 72-year-old housewife was referred to our clinic from her family physician with a complaint that “there are many balloons and holes on my palate all day.” The patient has no significant medical or psychiatric history.

Two years before and 3 months after eradicating *Helicobacter pylori*, she felt as if there were many holes on her palate and saw a gastroenterologist, but there were no abnormal findings. In addition, she felt that there were many balloons and needles on her palate. She saw the gastroenterologist again and was prescribed amitriptyline 10 mg/d, resulting in no improvement of her symptoms. Then she presented to our clinic by herself. Because there were no delusional or hallucinatory symptoms other than in oral regions, considering the clinical history together, we diagnosed her condition as oral cenesthopathy. On evaluation with the Oral Dysesthesia Rating Scale (Oral DRS),¹² the Symptom Severity Scale (SSS) score was 8 and Functional Impairment Scale (FIS) score was 7. We initially prescribed aripiprazole 1.5 mg/d. Single-photon emission computed tomography (SPECT) using technetium Tc 99m ethylcysteinate dimer was undertaken at 1 month after the first visit, when the symptoms had not changed. The right > left asymmetric rCBF was seen in the frontal and temporal lobes and thalamus (Fig. 1A). The magnetic resonance imaging showed no atrophy or infarction. Two months after the first visit, the feeling of holes on her palate still existed but did not irritate her. The symptoms gradually alleviated with the same dosage. Fourteen months after the first visit, she reported that her symptoms had resolved (Oral DRS scores: SSS, 2; FIS, 3) without any adverse effects. The SPECT imaging at that time showed the attenuation of the asymmetric rCBF (Fig. 1B).

Patient 2

A 73-year-old man, former office worker in the accounting department of a construction company, was referred by his family physician. His chief complaints were “sticky sensation in my mouth” and “gritty sensation and pressure-like sensation in the lower incisor.” The patient had histories of angina pectoris, diabetes, and osteoporosis, all of which were under control at initial presentation. No psychiatric history was detected.

Three years before, he felt the gritty sensation in his teeth spontaneously. He had visited 4 dental clinics and undergone dental hygiene treatment, resulting in no change. In addition to the gritty sensation, a sticky sensation developed 6 months later. In

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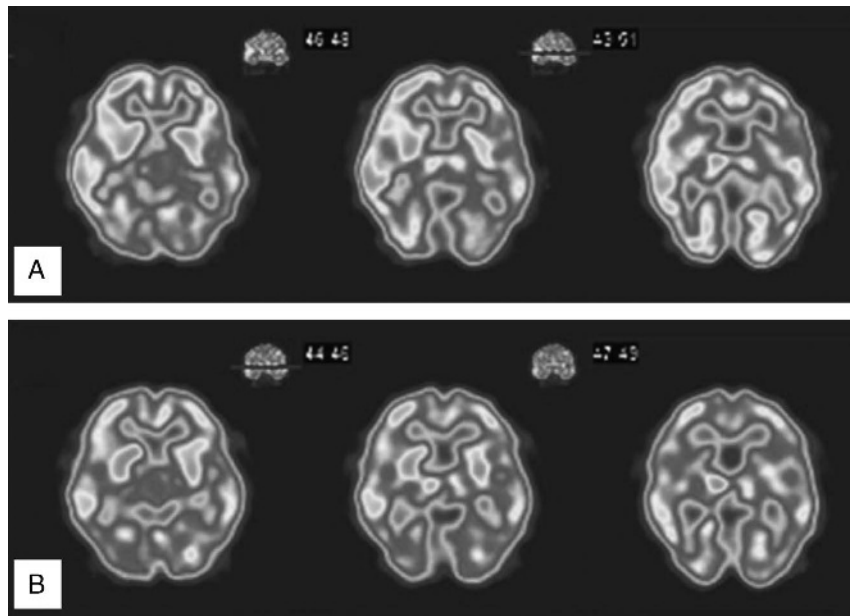


FIGURE 1. Single-photon emission computed tomography qualitative imaging in patient 1. The right > left asymmetric rCBF is shown in the frontal and temporal lobes and thalamus before the alleviation (A). The asymmetric rCBF was attenuated after the alleviation (B).

our first examination, no organic cause was detected. Considering the history of illness, the patient’s condition was diagnosed as oral cenesthopathy. Aripiprazole 1 mg/d was prescribed at first, and mirtazapine 7.5 mg/d was added to avoid sleep disturbance induced by aripiprazole 2 weeks after the first visit. The first SPECT was carried out 1 month after the first visit, when he had slight improvement (Oral DRS scores: SSS, 6; FIS, 2). The right > left rCBF asymmetries were seen in the inferior frontal lobe, temporal lobe, and thalamus, and slight rCBF decrease was detected in

the parietal lobes bilaterally (Fig. 2A). After the dosages of aripiprazole and mirtazapine were gradually increased, his complaints decreased. At the time of the second SPECT, which is 14 months after the first visit, he reported mostly being unaware of the sticky and gritty sensation throughout the day (Oral DRS scores: SSS, 2; FIS, 0) with the combination of aripiprazole 1.5 mg/d and mirtazapine 45 mg/d. No adverse effects were reported. The asymmetric rCBF in the inferior frontal and temporal lobes and thalamus and the decreased rCBF in the bilateral

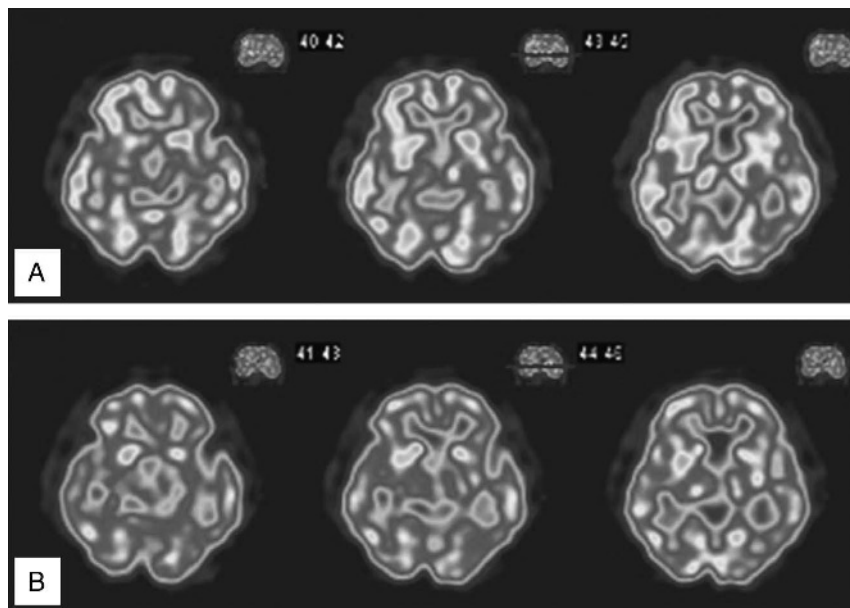


FIGURE 2. Single-photon emission computed tomography imaging in patient 2. The right > left rCBF asymmetries are shown in the inferior frontal lobe and temporal lobe and thalamus, and slight rCBF decrease was detected in the bilateral parietal lobes before the remission (A). The asymmetric rCBF in the inferior frontal and temporal lobes and thalamus and the decreased rCBF in the bilateral parietal lobes were attenuated after the remission (B).

parietal lobes were attenuated in the second SPECT (Fig. 2B). However, right > left asymmetric rCBF was detected in the superior frontal lobe only in the second SPECT; the pathological implication is unknown.

CONCLUSIONS

The 2 cases are the first to be reported of oral cenesthopathy alleviated with aripiprazole concurrently with attenuation of rCBF asymmetry. Although the detail is different, the right > left rCBF asymmetries in the frontal and temporal lobes and thalamus disappeared after successful treatment in both cases. The rCBF changes might reflect the symptom's improvement, because the prescriptions were almost the same between the first and second SPECT images in both cases.

In the literature, some neuroimaging case reports on oral cenesthopathy or delusional disorder, somatic type, in the oral area have shown the attenuation of right-side-predominant rCBF asymmetry in parallel with the improvement of symptoms by paroxetine,⁸ mECT,¹¹ and the combination of mECT and perospirone.¹⁰ In the present cases, we found similar rCBF change to former case reports regardless of treatment method. It suggests that the rCBF change might be an objective therapeutic indicator for oral cenesthopathy. It would be useful for many types of oral cenesthopathy, because the right-side-predominant rCBF asymmetry is common between presence and absence of a history of depression,⁷ while these cases were not second symptoms of psychiatric disorders.

Aripiprazole is a dopamine partial agonist that has high occupancy at D₂ receptor and lower occupancy at 5-HT_{1A} and 5-HT₂ receptors.¹³ Drug occupancy levels at D₂, 5-HT_{1A}, and 5-HT₂ receptors were correlated with plasma drug concentrations. Because of this unique profile, it is suggested that dose-effect relationship for aripiprazole exists in the oral cenesthopathy treatment. In the present cases, low-dose aripiprazole improved the symptoms, suggesting that the complex dopaminergic and serotonergic system is involved in the pathology of oral cenesthopathy.

In patient 2, the combination therapy with mirtazapine and aripiprazole had marked improvement. Schüle et al¹⁴ showed that the combination therapy with mirtazapine and aripiprazole accelerated the onset of their effects, and some case reports^{15,16} described that the combination therapy was safe and well tolerated and that mirtazapine reduced the aripiprazole-induced adverse effects and vice versa. The present cases suggested that not only aripiprazole but also the combination of aripiprazole and mirtazapine could be treatment options for oral cenesthopathy.

In conclusion, we found a marked improvement of oral cenesthopathy with aripiprazole. Considering SPECT images and the effective drug profile, it is suggested that right > left rCBF asymmetry in the frontal and temporal lobes and thalamus and the dopaminergic and serotonergic dysfunctions are involved in the pathology of oral cenesthopathy. To further reveal the association between symptoms and brain perfusion asymmetry, a study with a

larger number of cases that quantitatively evaluate symptoms by using the Oral DRS is warranted.

REFERENCES

- Dupré E, Camus P. Les cénesthopathies. *Encéphale* 1907;2:616–631.
- Takahashi T, Fuke T, Washizuka S, et al. A review of recent case reports of cenesthopathy in Japan. *Psychogeriatrics* 2013;13:196–198.
- Umezaki Y, Miura A, Watanabe M, et al. Oral cenesthopathy. *Biopsychosoc Med* 2016;10:20.
- Maeda K, Yamamoto Y, Yasuda M, et al. Delusions of oral parasitosis. *Prog Neuropsychopharmacol Biol Psychiatry* 1998;22:243–248.
- Umezaki Y, Katagiri A, Watanabe M, et al. Brain perfusion asymmetry in patients with oral somatic delusions. *Eur Arch Psychiatry Clin Neurosci* 2013;263:315–323.
- Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition*. American Psychiatric Association, Washington, DC; 1994.
- Watanabe M, Umezaki Y, Miura A, et al. Comparison of cerebral blood flow in oral somatic delusion in patients with and without a history of depression: a comparative case series. *BMC Psychiatry* 2015;15:42.
- Hayashi H, Oshino S, Ishikawa J, et al. Paroxetine treatment of delusional disorder, somatic type. *Hum Psychopharmacol* 2004;19:351–352.
- Akahane T, Hayashi H, Suzuki H, et al. Extremely grotesque somatic delusions in a patient of delusional disorder and its response to risperidone treatment. *Gen Hosp Psychiatry* 2009;31:185–186.
- Uezato A, Yamamoto N, Kurumaji A, et al. Improvement of asymmetrical temporal blood flow in refractory oral somatic delusion after successful electroconvulsive therapy. *J ECT* 2011;1:50–51.
- Ota M, Mizukami K, Katano T, et al. A case of delusional disorder, somatic type with remarkable improvement of clinical symptoms and single photon emission computed tomography findings following modified electroconvulsive therapy. *Prog Neuropsychopharmacol Biol Psychiatry* 2003;27:881–884.
- Uezato A, Toyofuku A, Umezaki Y, et al. Oral Dysesthesia Rating Scale: a tool for assessing psychosomatic symptoms in oral regions. *BMC Psychiatry* 2014;14:1696.
- Mamo D, Graff A, Mizrahi R, et al. Differential effects of aripiprazole on D₂, 5-HT₂, and 5-HT_{1A} receptor occupancy in patients with schizophrenia: a triple tracer PET study. *Am J Psychiatry* 2007;164:1411–1417.
- Schüle C, Baghai TC, Eser D, et al. Mirtazapine monotherapy versus combination therapy with mirtazapine and aripiprazole in depressed patients without psychotic features: a 4-week open-label parallel-group study. *World J Biol Psychiatry* 2007;8:112–122.
- Dimopoulos NP, Mitsionis CI, Psarra VV. Delusional disorder, somatic type treated with aripiprazole—mirtazapine combination. *J Psychopharmacol* 2008;22:812–814.
- Poyurovsky M, Bergman J, Pashinian A, et al. Beneficial effect of low-dose mirtazapine in acute aripiprazole-induced akathisia. *Int Clin Psychopharmacol* 2014;29:296–298.