

The efficacy of vortioxetine for the delusional disorder of cenesthopathy

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Abstract

Cenesthopathy is a rare syndrome characterized by strange bodily and oral sensations and is classified as a delusional disorder, somatic type, according to the fifth edition of the Diagnostic and Statistical Manual of Mental Disorders. Cenesthopathy has been considered difficult to treat. However, to improve cenesthopathy, many pharmacotherapeutic options are reported, including antidepressants and antipsychotics. In this case report, vortioxetine significantly alleviated the distress of oral cenesthopathy in a patient with cerebral ischemia and depression without any adverse effects. To the best of our knowledge, this is the first report on the efficacy of vortioxetine in treating cenesthopathy. Though it is unclear why vortioxetine was effective for cenesthopathy in our case, we stated two possibilities for improving his oral cenesthopathy. When treating oral cenesthopathy in elderly patients, clinicians consider to be one of the options to prescribe vortioxetine.

KEYWORDS

cenesthopathy, delusional disorder, norepinephrine, serotonin, vortioxetine

1 | INTRODUCTION

Cenesthopathy is a rare syndrome characterized by strange bodily and oral sensations. Patients with this syndrome often make statements such as "something is moving in my head," "something has been placed in my oral cavity," or "I have a burning feeling in my mouth."^{3,6,9-11} According to the fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5),¹ this syndrome is classified as a delusional disorder.^{4-6,10,11} This phenomenon was first presented in a French report³ in the early 20th century, and its etiology has been controversial. Cenesthopathy was initially reported as an aspect of psychosis or paranoia;^{4,5,10,11} however, it has recently been reported that it may also be found in patients with depression or dementia.⁹⁻¹¹

We here report a case in which vortioxetine improved oral cenesthopathy in a patient with cerebral ischemia and depression.

2 | CASE REPORT

A 77-year-old male who had had ischemic disease in the left lower temporal lobe of his brain 5 years previously was referred to our clinic due to a 1-year history of bizarre sensations in his teeth and tongue. He had hypertension and diabetes mellitus, for which he had been receiving treatment for 10 years more. Neither the patient nor his family members had any history of alcohol abuse or tobacco use, or any psychiatric history. The patient's oral hygiene was good, and there were no other pathological organic findings. He was

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diagnosed with a delusional disorder according to DSM-5; however, he had other psychiatric symptoms in addition to the abnormal oral sensations. His visual analogue scale (VAS) rating for the degree of discomfort of the oral conditions was 75/100 at the first interview. Although he had already been prescribed only imipramine (10mg/day) by brain surgery, he reported that he did not take the drug because of urinary retention and constipation.

The patient was informed about oral cenesthopathy and the treatment for it, as well as the possibility that the effects of the drug might be limited and adverse effects might occur. We changed his medication from imipramine to vortioxetine monotherapy (5mg/day). Two weeks after the initiation of vortioxetine, his oral sensations were somewhat reduced, his VAS was 50, and there were no adverse effects. At 1 month after the initiation of vortioxetine, the patient reported that there was little discomfort in his mouth, and his VAS was 20–30. He chose not to change the dosage of the drug much more because the degree of oral sensation was acceptable and tolerable for him and his daily life was not disturbed by symptoms. At his follow-up, slight symptoms remained, and he continued to take the same dose of vortioxetine with no side effects.

3 | DISCUSSION

To the best of our knowledge, this is the first report on the efficacy of vortioxetine in treating cenesthopathy. According to some reviews on cenesthopathy, oral cenesthopathy is a common subtype, especially in elderly patients. In some reports, the bizarre sensation is mainly related to cerebral dysfunction such as ischemic disease^{7,10} or mood disorders.^{7,9} In our case, cenesthopathy appeared 5 years after a brain injury, and our patient did not experience depression, loss of interest, or other mood symptoms. To improve cenesthopathy, many pharmacotherapeutic options are available, including antidepressants such as amitriptyline, mianserin, paroxetine, fluvoxamine, mirtazapine, and milnacipran. Additionally, antipsychotics such as haloperidol, pimozide, tiapride, risperidone, olanzapine, perospirone, and aripiprazole have been reported to improve somatic delusional symptoms.^{4,9–11} However, the mechanism of cenesthopathy has not been clarified. Some reports indicated that cenesthopathy resulted from damage of thalamocortical tract,^{9,10} other studies showed that various changes in cerebral blood perfusions were observed in cenesthopathy.¹⁰ It remains unclear which neurotransmitters would be associated with the development of cenesthopathy.^{10,11}

In Japan, vortioxetine was approved in 2019 for the treatment of major depressive disorder. Vortioxetine is a multimodal mechanism antidepressant. It is a 5-HT₃, 5-HT_{1D}, and 5-HT₇ antagonist, a 5-HT_{1A} agonist, and a 5-HT_{1B} partial agonist.² These actions might regulate many neurotransmitters such as noradrenaline, dopamine, acetylcholine, glutamate, and gamma-aminobutyric acid.^{2,8} In the present case, we decided to use vortioxetine not only for efficacy, to reduce the patient's oral discomfort, but also for safety, considering his brain ischemic injury. Given that the response to various types of treatments including electroconvulsive therapy for cenesthopathy is

reported to be lower than 50%,^{10,11} it is unclear why vortioxetine was effective for oral cenesthopathy in the present case. One possibility is that the patient had masked depression, and the improvement in his depressive symptoms led to reduced oral symptoms. Another possibility is that the patient's cognitive function was improved as a result of the vortioxetine regulating various neurotransmitters and that this in turn improved his oral cenesthopathy.

Clinicians should consider vortioxetine to treat oral cenesthopathy in elderly patients. However, further investigation with a greater number of cases is needed. In addition, affected regions of cenesthopathy are not only oral cavity but also any other unusual bodily sensations.^{3,11} Other studies are also necessary to reveal that whether types of medications would improve symptoms differently in cenesthopathy.

AUTHOR CONTRIBUTIONS

SS is the main author of the manuscript, and KI assisted in authoring the discussion of this paper.

FUNDING INFORMATION

No funding was received to assist with the preparation of this manuscript.

CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data of this study are not publicly available because informed consent on disclosing them has not been obtained.

ETHICS STATEMENT

Informed consent: Written informed consent was obtained in accordance with the World Medical Association's Declaration of Helsinki.

Approval of the research protocol by an Institutional Reviewer Board: N/A.

Registry and the Registration No. of the study: N/A.

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How to cite this article: Sato S, Imai K. The efficacy of vortioxetine for the delusional disorder of cenesthopathy. *Neuropsychopharmacol Rep*. 2023;00:1–3. <https://doi.org/10.1002/npr2.12384>