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【 CASE REPORT 】

Loss of Taste as an Initial Symptom of a "Facial Diplegia and Paresthesia" Variant of Guillain-Barré Syndrome

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Abstract:
Loss of taste is a relatively common symptom of coronavirus disease 2019 (COVID-19) and has also been considered a rare Guillain-Barré syndrome (GBS) symptom. We herein report a case of a facial diplegia and paresthesia (FDP) variant of GBS that initially presented as a loss of taste occurring two weeks after COVID-19 mRNA vaccination. The patient recovered completely after intravenous immunoglobulin therapy. Clinicians should consider the possibility of post-vaccination FDP manifesting as facial palsy and should be aware that GBS, including the FDP variant, can initially present as an isolated loss of taste.

Key words: loss of taste, gustatory disorders, facial diplegia and paresthesia, Guillain-Barré syndrome, coronavirus disease 2019, vaccination

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Introduction

The rapid spread of coronavirus disease 2019 (COVID-19), which was first reported in December 2019, has led to significant changes in healthcare systems and protocols worldwide. Although the predominant clinical presentation of COVID-19 is respiratory disease, gustatory dysfunction is also a common symptom (1). In addition, neurological manifestations, including Guillain-Barré syndrome (GBS), are potential complications of COVID-19 illness (2).

GBS can also be associated with COVID-19 vaccination (3). Loss of taste is a relatively common symptom of COVID-19 and has also been considered a rare GBS symptom (4, 5). However, the results of a study that investigated gustatory impairment in patients with GBS using electrogustometry suggested that taste impairment in GBS may be more frequent than previously thought (6).

We herein report a case of a facial diplegia and paresthesia (FDP) variant of GBS that initially presented as a loss of taste occurring two weeks after Pfizer COVID-19 mRNA vaccination.

Case Report

A previously healthy 19-year-old Japanese man presented with loss of taste 16 days after receiving the second dose of the Pfizer mRNA COVID-19 vaccine. The patient noticed complete loss of taste with all five taste modalities: sweet, sour, bitter, salty, and umami. Three months prior to receiving the second dose of the vaccine, the patient had contracted COVID-19 with a fever as an isolated symptom; he did not have dysosmia or dysgeusia. He completely recovered from COVID-19 within two days.

However, two days after the onset of his loss of taste (day 3), he noticed numbness in his left foot, which extended to the distal parts of his four extremities two days later. On day 8, he developed difficulty closing his left eye, and on day 9 he noticed the left side of his mouth drooping and visited our hospital. The patient was initially diagnosed as suffering Bell's palsy and treated using oral valacyclovir and prednisolone. Following the worsening of his symptoms, he developed difficulty closing his right eye on day 11. The patient was then admitted on day 12.

A physical examination revealed that the patient had isocoric pupils, no limitation of eye movements, and no blepharoptosis. In addition, the patient showed bilateral periph-

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eral type facial weakness (House-Brackmann grade 4, bilaterally). However, his muscle strength and deep tendon reflexes were normal. He had no ataxia, and his gait was normal. Pinprick sensation and joint position sensation were normal.

Blood testing showed that his peripheral white blood cell count was 8,600/μL with 58.5% lymphocytes. His aspartate aminotransferase level was 51 IU/L, whereas his alanine aminotransferase level was 191 IU/L. A serological investigation revealed that the patient was positive for Epstein-Barr virus (EBV) viral capsid antigen (VCA) IgM, negative for VCA IgG, positive for EBV early antigen, and showed an equivocal EBV nuclear antigen result. Results of serological tests for hepatitis A, B, and E viruses, herpes simplex virus, varicella-zoster virus, cytomegalovirus, mycoplasma, and human immunodeficiency virus were all negative for recent or active infection. Serum IgG and IgM antibodies against glycolipids GM1, GM2, GM3, GD1a, GD1b, GD3, GT1b, GQ1b, and GalNAc-GD1a were negative. A cerebrospinal fluid analysis showed that the patient had 1 white blood cell/mm^3 and a protein level of 76 mg/dL. COVID-19 polymerase chain reaction, which was performed using a nasopharyngeal swab, was negative. A stool culture was negative for *Campylobacter jejuni*.

Electrodiagnostic studies in his right extremities showed decreased compound muscle action potentials in the median nerve (2.4 mV) and decreased sensory nerve action potentials in the ulnar nerve (5 μV). The distal latencies, conduction velocities, and F-wave latencies were all within normal ranges. His clinical history, physical examination results, and laboratory findings were consistent with a diagnosis of FDP.

The patient underwent intravenous immunoglobulin therapy (0.4 g/kg for 5 consecutive days), starting on the day of admission. His taste sensation improved from day 15, facial palsy from day 17, and paresthesia from day 20. His loss of taste and bilateral facial palsy completely resolved on days 35 and 45, respectively. Deep tendon reflexes of the four extremities tended to be mildly inactive after admission but were at no point lost.

Discussion

This report describes a case of FDP following the second dose of the Pfizer mRNA COVID-19 vaccine, highlighting two important clinical issues. First, this case suggests that the Pfizer COVID-19 vaccine may cause FDP. Second, FDP can initially present as a loss of taste.

Classical GBS is subdivided into demyelinating GBS and axonal GBS. Other variants are (in order of frequency) Miller Fisher syndrome, the pharyngeal-cervical-brachial variant, FDP, and the paraparetic variant (7). Classical GBS is the most frequent type, whereas FDP is a very rare variant. The frequency of FDP among all cases of GBS has been reported to be as low as 1% in Japan (8). Most patients (75.5%) with COVID-19-associated GBS have a demyelinating subtype (9). For GBS following COVID-19 vaccination, the most frequent subtype is the classic form (56%), and the second-most frequent subtype is FDP (31%) (3). In fact, FDP after receiving the AstraZeneca COVID-19 vaccine has been reported in two case series (7, 10). The World Health Organization database analysis suggests that COVID-19 vaccination, especially vaccination using adenovirus vaccines, such as the AstraZeneca vaccine, is associated with GBS with facial palsy (11). The association between FDP and COVID-19 vaccination may be confirmed if post-COVID-19 vaccination GBS displays a specific phenotype, particularly GBS with facial palsy and FDP (11). Our case suggests that, in addition to the AstraZeneca vaccine, the Pfizer mRNA vaccine may trigger FDP. Most cases of GBS are easy to diagnose; however, it is often difficult to diagnose variants of GBS, particularly FDP, in the early stages. Like in the present case, early-stage FDP may be misdiagnosed as Bell's palsy. In fact, COVID-19 vaccination can cause Bell's palsy (12), but the treatment of Bell's palsy generally differs from that of GBS. Although the immune-treatment response and indication for FDP after COVID-19 vaccination remain unclear, the patient in this study showed satisfactory recovery after intravenous immunoglobulin treatment.

The second important issue to note is that loss of taste in this patient was the initial symptom of FDP, which can also be an initial symptom of classical GBS, albeit extremely rarely (5, 13, 14). The facial nerve is predominantly made of large, myelinated fibers, while the chorda tympani, a branch of the facial nerve, comprises small and thinly myelinated fibers (4). This difference in fiber size may be one possible reason for our case's initial symptoms: the small fibers might have been damaged first, resulting in the initial taste loss. Gustatory dysfunction has been reported in 88.8% of COVID-19 cases and is a common initial symptom (1). Furthermore, COVID-19 vaccination itself also can cause gustatory dysfunction (15). Therefore, given the COVID-19 pandemic, it may be difficult to diagnose a patient who initially presents with gustatory symptoms.

One limitation of this case report is that the serological results indicated a recent EBV infection, which previous reports indicated as a potential preceding sign of GBS, questioning whether this case's FDP was triggered by EBV or vaccination. Notably, the patient did not have a fever, sore throat, swollen tonsils, or lymphadenopathy. In contrast, the results of a Japanese study on FDP indicated that the serological EBV positivity rate for patients with FDP was not significantly higher than that of patients with Bell's palsy or healthy controls, suggesting that EBV is not a significant preceding sign of FDP (16).

In conclusion, this case suggests that the Pfizer COVID-19 vaccine may cause FDP and indicates that the FDP variant of GBS can initially present as a loss of taste. Clinicians must consider the possibility that post-vaccination facial palsy may be FDP. Furthermore, physicians should be aware that GBS, including the FDP variant, can initially present as

an isolated loss of taste.

The authors state that they have no Conflict of Interest (COI).

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