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【 CASE REPORT 】

Foix-Chavany-Marie Syndrome Induced by a Unilateral Brain Abscess

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Abstract:
Foix-Chavany-Marie syndrome (FCMS) is a rare cortical type of pseudobulbar palsy characterized by the loss of voluntary control of the facial, pharyngeal, lingual, and masticatory muscles with preserved reflexive and autonomic functions. FCMS is generally associated with cerebrovascular diseases affecting the bilateral opercular regions. We herein report the clinical features of an 84-year-old right-handed Japanese man with FCMS due to a unilateral brain abscess. The patient's symptoms were resolved after treating the brain abscess. The present clinical results suggest that a unilateral brain abscess in the temporal operculum with a persistent old lesion in the contralateral insular cortex can induce FCMS.

Key words: anterior opercular syndrome, brain abscess, Foix-Chavany-Marie syndrome, pseudobulbar palsy, temporal operculum

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Introduction

Foix-Chavany-Marie syndrome (FCMS), also known as anterior opercular syndrome, is a rare cortical type of pseudobulbar palsy characterized by the loss of voluntary control of the facial, pharyngeal, lingual, and masticatory muscles with preserved reflexive and autonomic functions (1, 2). FCMS is generally associated with cerebrovascular diseases affecting the bilateral opercular regions (3, 4). We herein report a case of FCMS induced by a unilateral brain abscess.

Case Report

An 84-year-old right-handed Japanese man presented to our department with weight loss because of an inability to consume food. He had a medical history of chronic hepatitis C and asthma with no medications. Two months earlier, the patient had been observed to have some difficulty speaking and swallowing. Thereafter, these symptoms gradually worsened, and his weight decreased from 42 kg to 31 kg within a 2-month period.

At admission, the results of a general physical examination were mostly normal, but a high fever (38.2 °C) and low body mass index (11.5 kg/m^2) were noted. On a neurological examination, the patient was alert but had anarthria and severe dysphagia because of an inability to voluntarily move his lower face and masseter muscles. He could not voluntarily protrude his tongue either. In particular, his mouth was continuously half open and could not be closed when he was asked to do so. However, he was able to close his mouth when he spontaneously smiled and yawned. His writing and comprehension were preserved. In addition, he had no motor or sensory deficits in the extremities, and the tendon reflex was normal in all four limbs. Based on these neurological findings, the patient was diagnosed with FCMS.

Routine hematological tests showed that the patient had normocytic normochromic anemia with a hemoglobin level of 10.9 g/dL (reference range: 12.0-16.0 g/dL). Biochemical tests found that the C-reactive protein level was elevated, with a value of 3.77 mg/dL (reference range: <0.3 mg/dL), and the serum albumin level was decreased, with a value of 2.7 g/dL (reference range: 3.7-5.2 g/dL). His serum sample was found to be positive for hepatitis C virus. A radiographic examination and magnetic resonance imaging (MRI) of the brain revealed old cerebral hemorrhaging in the right insular cortex, isolated sphenoid sinusitis, and a ring-

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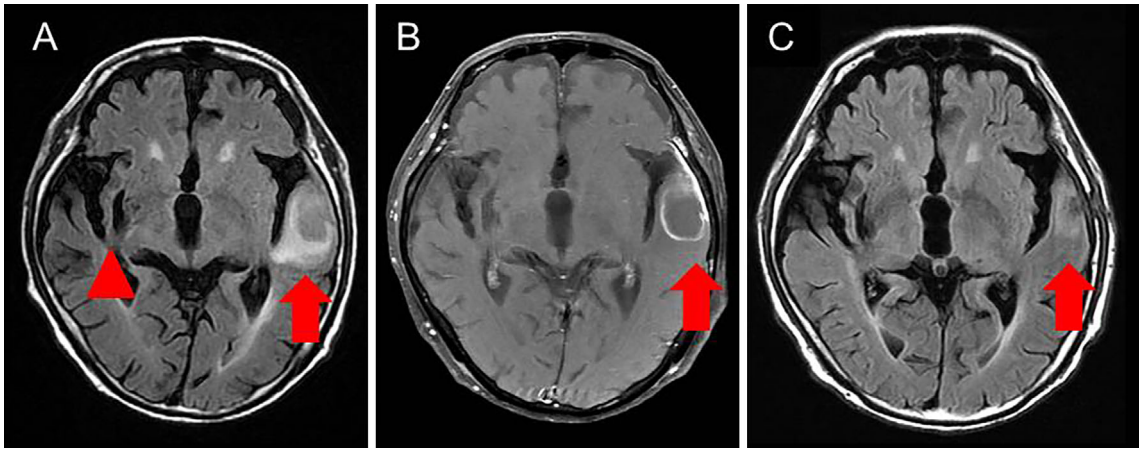

Figure. **Brain magnetic resonance imaging at admission (A and B) and after the treatment (C). (A) Fluid-attenuated inversion recovery (FLAIR) images show old cerebral hemorrhaging in the right insular cortex (arrowhead) and a cystic mass surrounded by brain edema in the left superior temporal gyrus (the temporal operculum) (arrow). (B) T1-weighted imaging with gadolinium enhancement showed ring-enhancing cystic mass lesions (arrow), confirming the presence of a brain abscess. (C) FLAIR images obtained after the treatment show that the cystic mass had almost disappeared (arrow).**

enhanced cystic mass surrounded by brain edema in the left superior temporal gyrus (the temporal operculum), suggesting the presence of a brain abscess (Figure A and B). The brain abscess may have spread from the sphenoid sinusitis.

The patient was initially treated with intravenous antibiotic therapy with ceftriaxone. Subsequently, sphenoidotomy was performed on day 8 of admission. Although his body temperature was normalized after these treatments, the neurological symptoms and the size of the brain abscess did not improve. On day 24, he underwent computed tomography-guided percutaneous drainage of the brain abscess, and approximately 8 mL of yellowish-brown pus was aspirated from the lesion. A small amount of pus was sent for a culture analysis, but no bacterial growth was detected. After percutaneous drainage, his neurological symptoms gradually improved. One week later, he regained his ability to speak and swallow. Brain MRI on day 60 after drainage revealed that the brain abscess had improved (Figure C).

Discussion

We encountered a case of FCMS with a unilateral opercular lesion. This patient had old cerebral hemorrhaging and a subacute brain abscess in the right subcortical insular regions and left temporal operculum, respectively. The symptoms were resolved after treatment for the brain abscess. To our knowledge, this is the first case report in which a patient with FCMS induced by a brain abscess showed a relatively good recovery, as assessed using MRI.

FCMS is a cortical type of pseudobulbar palsy characterized by a loss of voluntary control of the facial, pharyngeal, lingual, and masticatory muscles with preserved reflexive and autonomic functions (1, 2). FCMS is most commonly caused by sequential infarcts to the bilateral anterior opercular or subcortical insular regions (3, 4) and less commonly by a unilateral opercular lesion (5-8). The exact functional mechanisms underlying this rare syndrome remain unclear.

In general, the prognosis of FCMS caused by the formation of bilateral opercular lesions tends to be poor, and most patients have persistent clinical deficits. In contrast, patients with FCMS caused by a unilateral opercular lesion have a relatively good prognosis (8). In the present case, the symptoms were resolved after treatment for the brain abscess, suggesting that the prognosis was relatively good. Therefore, based on the clinical findings in this case, we speculate that a newly arising unilateral brain abscess in the left temporal operculum with a persistent old lesion in the contralateral insular cortex results in bilateral opercular dysfunction, which is categorized as FCMS.

One limitation associated with our case report is that the mechanisms of FCMS remain elusive due to the insufficient performance of examinations used to examine the blood flow and metabolic functions of the brain, such as single-photon emission computed tomography and positron emission tomography. However, the present findings are crucial for delineating the mechanisms of FCMS.

In conclusion, we encountered a case of FCMS induced by a unilateral brain abscess. Further investigations are needed to clarify the mechanisms and pathogenesis of FCMS.

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

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