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Case report

Surgical treatment of enterovirus D68 brainstem encephalitis-induced dysphagia☆

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ABSTRACT

Cluster of acute flaccid paralysis and cranial nerve dysfunction was associated with a 2014 outbreak of enterovirus D68 (EV-D68) respiratory illness in US. We describe a 33 year-old male patient of refractory dysphagia due to EV-D68-induced brainstem encephalitis successfully treated by surgery. Following acute upper respiratory tract infection, he developed dysphagia and bilateral facial paralysis. A coughing reflex was readily produced when the laryngopharyngeal fiberscope touched the epiglottis, however, water infusion induced only very weak and slow swallowing reflex, suggesting that only motor component was impaired but sensory function was preserved during swallowing. Despite eight months-conservative rehabilitations, Food Intake Level Scale (FILS) remained level 4. Therefore, corrective surgeries including cricopharyngeal myotomy, laryngeal suspension, and pharyngeal flap were performed. Thirty-six days after surgery, FILS rapidly and dramatically improved to level 8. This is the first report describing a successful surgical intervention for EV-D68-induced refractory dysphagia. Surgical treatment was suitable for EV-D68-induced dysphagia, perhaps because sensory function was preserved and only motor disturbance was present during the pharyngeal stage of swallowing.

1. Introduction

In 2014, an outbreak of enterovirus D68 (EV-D68) occurred in North America [1,2]. According to the report of this outbreak [1], the number of children requiring hospital admission to the Children's Hospital Colorado due to EV-D68 respiratory disease during three months-period was three times higher than the maximum number of cases during the preceding 4 years. Among these patients, 12 had not only respiratory illness but also neurological diseases such as flaccid paralysis and acute cranial nerve dysfunction. It included flaccid limb weakness (n = 10), bulbar weakness (n = 6), and cranial nerve VI (n = 3) and VII (n = 2) dysfunction. Although intravenous immunoglobulin and methylprednisolone were administered in 75% and 42% of patients, respectively, no patients showed complete recovery of limb weakness and only two out of six patients showed improvement in bulbar weakness. Mild improvement in limb weakness has been noted in some children with physical therapies; however, all these children had persistent motor deficits. Therefore, prognosis of neurological deficits was very poor once they occurred.

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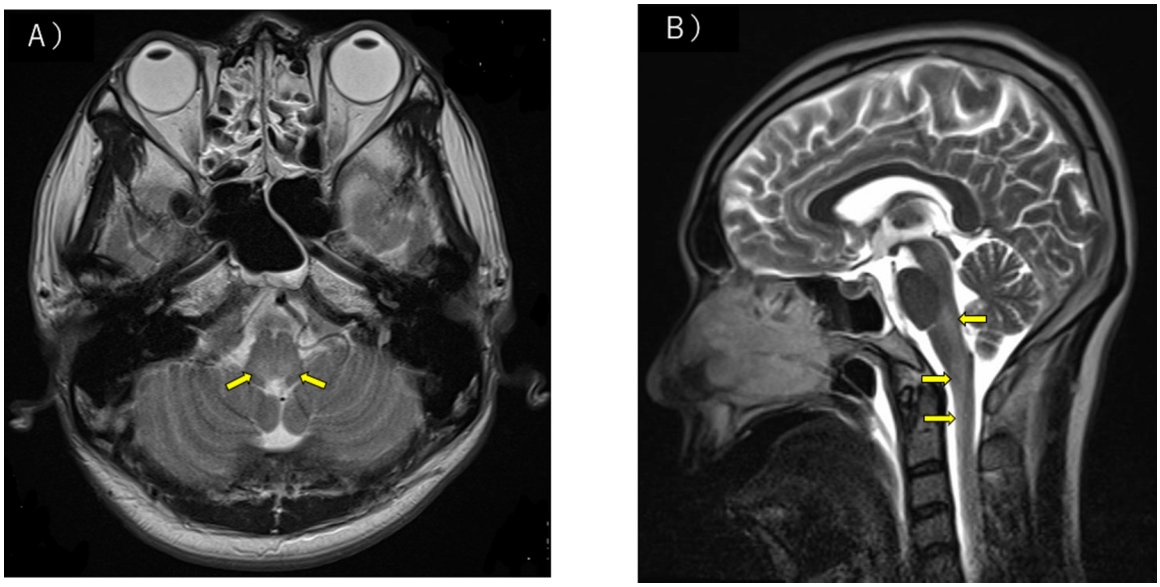

Fig. 1. T2-weighted magnetic resonance (MR) images on day 3.
A): axial images B): sagittal images.
Hyperintense region extending from the pons and dorsal medulla oblongata to the ventral side of cervical spinal cord (arrows) was seen, suggesting brainstem encephalitis.

In autumn 2015, an EV-D68 outbreak also occurred in Japan [3]. This report describes an adult patient with sequelae from EV-D68 infection, including severe treatment-refractory dysphagia. Ultimately, surgical treatment restored satisfactory swallowing function. To our knowledge, no previous report described successful surgical treatment of EV-D68 infection-related dysphagia. We believe that surgical treatment can be useful probably because EV-D68 infection causes paralysis of motor function but not of sensory function in the affected area.

2. Case report

Following acute upper respiratory tract infection, a 33-year-old man developed dysphagia and bilateral facial paralysis, for which he sought treatment at our neurological department in September 2015. On admission his consciousness was clear, pupil size was normal, and he had no nuchal rigidity, abnormality in eye movement, or loss of facial sensation. Tongue protrusion and soft palate elevation was insufficient without laterality. Gag reflex was weak. Limb movement was not impaired, but muscle strength in his neck and trunk was slightly diminished. These neurological findings suggested bulbar paralysis and cranial nerve VII dysfunction. An emergency CT scan revealed no evidence of cerebral hemorrhage or infarction (data not shown). On the second day, endotracheal intubation was performed because of aspiration due to severe dysphagia. On the third day, T2-weighted magnetic resonance imaging (MRI) revealed a hyperintense region extending from the pons and dorsal medulla oblongata including facial nucleus to the ventral side of cervical spinal cord (Fig. 1A, B). It is suggested that brainstem encephalitis caused bulbar paralysis and cranial nerve VII dysfunction. We started him on cocktail treatment including intravenous immunoglobulin (0.4 g/kg/day), ceftriaxone (2 g/day), and steroid pulse therapy (1 g methylprednisolone followed by 60 mg of prednisone) for three days. Aspiration continued to be severe, and the patient underwent tracheotomy on day 11. On day 13, a considerable amount of saliva was beginning to accumulate in the pyriform sinus and larynx. The patient was unable to elevate his larynx, and a curtain sign was noted on the right side, along with rhinopharyngeal dysraphism. The above findings were consistent with a diagnosis of severe brainstem encephalitis-induced dysphagia, which was classified as level 2 (no oral intake and swallowing training not using food is performed) on the Food Intake Level Scale (FILS), a 10-point observer-rating scale to measure the severity of dysphagia [4]. The FILS score was reported to be highly correlated with the functional oral intake scale (FOIS) [5], but is potentially sensitive to changes in swallowing status [4]. The patient was started on a program of indirect swallowing rehabilitation. On day 99, he underwent percutaneous endoscopic gastrostomy. On day 151, balloon dilatation training was begun. The patient was then able to swallow a little water, however, FILS still remained at level 4 (oral intake and alternative nutrition, easy-to-swallow food less than the quantity of a meal (enjoyment level) is ingested orally) until around day 250. The Japanese National Institute of Infectious Diseases analyzed serum obtained from the patient. The findings revealed EV-D68 infection, which was thus confirmed as the cause of brainstem encephalitis.

Figs. 2(A, B) and 3A show the videoendoscopic and videofluoroscopic examination of swallowing seven months after the onset, respectively. When the patient was tested for oral infusion of colored water, very slow and weak swallowing reflex was observed but most of it accumulated in the hypopharynx (Fig. 2 A). A coughing reflex was readily produced when the laryngopharyngeal fiberscope touched the epiglottis. Moreover, when the colored water entered the larynx, the expected coughing reaction prevented aspiration. These findings suggested a preservation of sensation. However,

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