OLGU SUNUMU/
CASE REPORT

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Herpes Simplex Encephalitis Presenting with Isolated Ninth Cranial Nerve Palsy: A Case Report

İzole Dokuzuncu Kranial Sinir Paralizisi İle Başvuran Herpes Simpleks Ensefaliti: Olgu Sunumu

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ABSTRACT

Herpes simplex virus is the most common cause of acute sporadic viral encephalitis. Mortality is over 70% in untreated cases. Clinical diagnosis of the disease is difficult. Routine laboratory results are generally nonspecific. Here, we consider the present case worthy to be presented because we demonstrated for the first time that herpes simplex encephalitis may manifest with isolated ninth cranial nerve palsy based on a patient, who presented at the age of 27 years with dysphagia and loss of sense of taste, had MRI and EEG findings consistent with herpes encephalitis, and improved with acyclovir therapy without complication.

Key words: Herpes simplex, encephalitides, glossopharyngeal motor neuropathies, acyclovir

ÖZ

Herpes simpleks virusu akut, sporadik viral ensefalitlerin en sık tanımlanan etkenidir. Tedavi edilmeyen olgularda mortalite %70'in üzerindedir. Klinik olarak hastalığın tanısını koymak güçtür. Rutin laboratuvar sonuçları genellikle nonspesifiktir. Burada 27 yaşında yutamama ve tat alamama şikayeti ile başvuran, MRG ve EEG bulguları herpes ensefalitini destekleyen, asiklovir tedavisi ile komplikasyonsuz iyileşen, ilk defa izole dokuzuncu kranial sinir paralizisi ile de başvurabileceğini göstermiş olduğumuz için sunmaya değer bulduk.

Anahtar kelimeler: Herpes simpleks, ensefaliti, glossopharyngeal sinir nöropati, asiklovir

Introduction

Herpetic encephalitis is the most common and the highest mortality of encephalitis (1-5). It is clinically characterized by high fever, headache, and confusion. Focal or generalized seizures may also occur. There may be severe neurological condition progressing from psychotic behavioral disorders, hemiplegia, speech disorders and amnesia to stupor and coma (4-9). Here we present the case with herpes encephalitis who presented with dysphagia and loss of sense of taste and improved without complication with acyclovir therapy after diagnosed with herpes encephalitis, we have shown that it is possible to present with 9th cranial nerve paralysis for the first time in herpes encephalitis cases that may present with atypical admissions.

Case Report

Personal and family history of the 27-year-old female patient, who presented with reduced oral intake, difficulty in swallowing and loss of sense of taste, was unremarkable. Her physical examination revealed good general status and clear consciousness. Her arterial blood pressure was 120/80 mmHg, radial pulse was 88/minute, body temperature was 38°C, and respiration rate was 22/minute. Her gag reflex was diminished and her voice was hoarse with a nasal twang. Her tongue deviated to the right side when asked to protrude. There was no neck stiffness or signs of meningeal irritation. Her laboratory analyses demonstrated a leukocyte count of 13.100/mm^3 (5.200–12.400/mm^3), hemoglobin value of 13.4 g/dl (12–18 g/dl), hematocrit concentration of 40.1% (37–52%), and

C-reactive protein value of 4.8 mg/dl (0.1–0.5 mg/dl). Thrombocyte count and biochemical parameters were within the normal limits. During follow-up, her general status worsened suddenly and she lost her consciousness. Reevaluating the patient, neck stiffness and the signs of meningeal irritation were determined, pupils were mid-dilated, and her light and corneal reflexes were negative. The patient was intubated as her general status and arterial blood gases were extremely impaired. Observing such a fast clinical worsening in a very short time, cerebral MRI was performed and hyperintense areas were seen in the bilateral temporal lobes. CSF specimen, which was taken because of prediagnosis of viral encephalitis, was clear in appearance with slightly increased CSF pressure, leukocyte count of 60/mm^3, and cellular predominance of lymphocyte by 90%. CSF glucose was 76 mg/dL and protein was 21 mg/dL. EEG showed mild diffuse background abnormality and focal paroxysmal abnormal activity in the frontotemporocentral region composed of the combination of slow theta waves and low-amplitude sharp waves. Prediagnosing the patient with encephalitis based on the clinical, laboratory and radiological findings, 30 mg/kg/day acyclovir therapy was commenced in addition to antiedema therapy (mannitol 1 mg/kg) and anti-epileptic drug treatment (Levetiracetam 1000 mg/day). CSF culture revealed no growth. Regarding Herpes simplex virus-DNA (HSV) level studied in the CSF by PCR method, HSV-DNA type–1 was positive. As she was suffering from impaired taste and difficulty in swallowing, recently contrasted foci were observed in the right frontal and right occipital regions on the control cerebral MRI. Based on these findings, she was thought to have ninth cranial nerve palsy. As the patient's general status improved, she was extubated. The patient's body temperature decreased on the eighth day of hospitalization. She developed no complication, and acyclovir therapy was stopped on the 21 th day of hospitalization. Control LP performed at the end of treatment revealed completely improved CSF. On the control visit performed after 6 months, all parameters including radiological findings were normal.

Discussion

It is possible to make the diagnosis of herpes encephalitis early by cranial imaging considering the disease in the patients presenting with headache, fever, epileptic seizure, etc.; furthermore, prevention of the related potential severe morbidity and mortality is also possible with early treatment (1-3,7-9). Early diagnosis and prompt treatment are likely in the patients presenting with typical clinical signs, whereas it is usually difficult to make the diagnosis in those presenting with atypical signs. Herein, we reported this herpes encephalitis case because of atypical clinical presentation who had not previously been reported in the literature.

HSV enters the system most frequently through the oropharyngeal mucosa and rarely through the conjunctiva and damaged skin. The virus needs to reach to the central nervous system via hematogeneous route or via olfactory nerves to cause encephalitis. While encephalitis clinic occurs due to primary disease in children and young adults, it occurs due to retrograde spread of the virus from the peripheral neurons to the brain in advanced ages (3-5,8,9).

Laboratory findings are not specific to herpes encephalitis. Although CSF pressure is usually high, the literature comprises also the cases reported to have normal CSF pressure. Polymorphonuclear leukocytes increase in early phases together with lymphocytic pleocytosis and elevated protein concentration. CSF contains erythrocytes in hemorrhagic encephalitis caused by herpes virus. This is helpful in diagnosis, but not diagnostic (2-5,8,9). In the present case, the CSF was clear in appearance with slightly elevated pressure. Microscopic examination of CSF, mildly elevated protein concentration and normal glucose concentration confirmed the literature.

MRI is another auxiliary method in diagnosing herpes encephalitis. It is the first-line radiological method to be considered immediately in the cases suspected to have herpes encephalitis. Detection of focal hemorrhagic necrotic areas in the temporal lobe is specific to HSV encephalitis. This sign is critical for distinguishing HSV from the other types of encephalitis. Another radiological finding is the unilateral involvement of the temporal lobe with the ganglia preserved [3-5]. The findings of cranial MRI performed in the present case on the first day of hospitalization confirmed the diagnosis.

Clinical signs of HSV encephalitis start with two-three-day of prodromal period, which generally include fatigue, fever and headache. Subsequently, the clinical picture may progress to seizure, hemiplegia, dysarthria, amnesia, stupor and even coma (2-5). The present case did not have a prodromal period and also presented with loss of sense of taste and dysphagia contrary to the expected neurological picture. Afterwards, the clinical picture of the case worsened rapidly.

Dysphagia is a symptom occurring due to mechanical prevention of the food transfer from mouth to the stomach and decreased strength or impaired coordination of the muscles that help swallowing. Oropharyngeal dysphagia results usually from neurological, myopathic and metabolic reasons. The reason may sometimes be a simple sore throat, or it can occur due to stroke, head trauma, Bell's paralysis or a metabolic encephalopathy (5-9). The loss of sense of taste, which was the other complaint of the present case, is usually ignored as it is not a life-threatening sign. The differential diagnosis includes many reasons as dysphagia. It can develop due either to a simple reason as poor oral hygiene or to a neurological disease such as head trauma, multiple sclerosis and Parkinson disease (5,7). Regarding this case presenting with dysphagia and loss of sense of taste, the literature reports no herpes encephalitis case presenting with these complaints. In this case, in which ninth cranial nerve palsy was suspected based on her complaints, the radiological methods ruled out tumor, varicella zoster infection and cranial hemorrhage, which might cause similar symptoms, in the differential diagnosis. The fact that the patient's clinical picture improved with the improvement of herpes encephalitis confirmed the diagnosis.

Conclusion

The present study considered worth reporting in order to underline that herpes encephalitis, which is a rare condition with high mortality and morbidity rates in untreated or late-treated cases, may present with isolated ninth cranial nerve palsy, which has not been reported in the literature yet, and that it can be even improved with treatment without sequel.

AUTHOR CONTRIBUTIONS:

Concept: SŞB, SO, EŞ, YT, BY; **Design:** SŞB, SO, EŞ, YT, BY; **Supervision:** SŞB, SO, EŞ, YT, BY; **Resources:** SŞB, SO, EŞ, YT, BY; **Materials:** SŞB, SO, EŞ, YT, BY; **Data Collection and/or Processing:** SŞB, SO, EŞ, YT, BY; **Analysis and/or Interpretation**: SŞB, SO, EŞ, YT, BY; **Literature Search:** SŞB, SO, EŞ, YT, BY; **Writing Manuscript**: SŞB, SO, EŞ, YT, BY; **Critical Review:** SŞB, SO, EŞ, YT, BY.

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