



Case Report

A Different Perspective on the Phenomenon of Hemiplegic Encephalitis: A Case Report

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Abstract

Encephalitis is an acute inflammation of the central nervous system that is characterized by fever, headache, and sleep disorders. Among the causes of meningoencephalitis are bacterial and viral infections, autoimmune diseases, and drug reactions. This case report describes a male patient who experienced hemiplegia following encephalitis. A 35-year-old patient consulted a physician with complaints of fever and headache. The clinical evaluation was encephalitis and the patient was treated accordingly. A severe neurological deficit developed, and the patient became bedridden. After treatment, the patient was included in an inpatient physical therapy program and was subsequently discharged with the ability to walk using parallel bars. The aim of this case report was to draw attention to the rehabilitation process applied after the diagnosis and treatment of a patient with hemiplegia that developed as a complication of encephalitis.

Keywords: Encephalitis; hemiplegia; meningoencephalitis.

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Encephalitis is defined by cerebral inflammation associated with neurological dysfunction.^[1] Fever, headache, and variable states of consciousness are common features of meningitis. Acute mental confusion, behavioral changes, focal neurological findings, and seizures are also frequently seen in cases with encephalitis.^[2]

The season of onset and development, geographical characteristics, prevalent diseases in the community, history of travel, profession, contact with animals, vaccinations, and immunodeficiency should be examined in the investigation of etiology.^[2, 3]

The causative agent of encephalitis may be determined from cultures of body fluids or biopsy specimens, the de-

tection of a relevant antigen during analysis of samples or nucleic acid amplification, or the measurement of serum immune globulin M level. In addition, radiological examinations, such as magnetic resonance imaging (MRI), computed tomography (CT), electroencephalography (EEG) and 2-fluoro-2 deoxy d-glucose-positron emission tomography may be helpful.^[2] Once the agent has been identified, the appropriate treatment and rehabilitation programs should be initiated for hemiplegia or other sequelae.

The objective of this case presentation is to briefly describe the diagnosis and treatment of encephalitis, and provide guidance about what a physical therapy and rehabilitation clinic can achieve during the rehabilitation process.

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Case Report

A 35-year-old male patient presented at the outpatient clinic with the complaints of a loss of strength on the left side and difficulty walking. The patient had previously gone to a medical center with complaints of fever and a headache, and he had been sent home with medical treatment. However, 1 week later, the symptoms of diplopia and a drooping mouth developed, and he consulted a private hospital. The first neurological examination revealed bilateral nystagmus on lateral gaze, peripheral facial paralysis on the right side, hypoesthesia of the left half of the face, and unresponsive bilateral plantar skin reflex. A cranial MRI detected brainstem edema with a homogenous contrast-enhanced focus, and hyperintensity on T2-weighted fluid-attenuated inversion recovery sections, which suggested the presence of vasculitis, postviral demyelinating pathology, or encephalitis. The results of a biochemical analysis of cerebrospinal fluid (CSF) retrieved via lumbar puncture (LP) included protein: 74 mg/dL, white blood cell count: 115/mm³, erythrocyte count: 4/mm³. Some parameters of vasculitis and viral diseases, such as antinuclear antibody, perinuclear antineutrophil cytoplasmic antibody, cytoplasmic antineutrophil cytoplasmic antibody, anti-Ro, and anti-La antigens were analyzed, A CSF culture, tuberculosis culture, and testing for herpes simplex virus (HSV) types 1 and 2 using a polymerase chain reaction (PCR) test were performed. Treatment with acyclovir (3x750 mg) and ceftriaxone (2x2 g) was initiated, and the patient was sent to another teaching and research hospital. On the 11th day of treatment, neurological manifestations worsened, and another cranial MRI examination demonstrated that the earlier lesion in the pons had disappeared, but a new contrast-enhanced lesion was observed. Behçet's disease testing was performed, and intravenous methylprednisolone pulse (IVMP) therapy was initiated and continued for 10 days. Left-sided hemiplegia developed on the eighth day of IVMP treatment. Autoimmune encephalitis was considered, and intravenous immunoglobulin treatment of 5 daily doses of 0.4 mg/kg was added to a daily dose of oral methyl prednisolone of 64 mg. On the third day of this treatment, clouded consciousness developed. A repeat cranial MRI detected a new supratentorial lesion to the left of the midline. A decompressive right frontoparietal craniotomy was performed, and samples were obtained from the arachnoid and pial regions and sent for culture and antibiogram. The department of infectious diseases initiated meropenem treatment of 2 g 3 times a day. The histopathology report of the biopsy specimen indicated diffuse macrophage infiltration consistent with demyelinating disease or chronic inflammation. The PCR analysis of vasculitis and viral in-

fection parameters of HSV, Ehrlich-Ziehl-Neelsen staining, incubation on Löwenstein media, and other cultures for tuberculosis and *Brucella* screening yielded negative results. However, *Burkholderia mallei* was present in the samples, so trimethoprim/sulfamethoxazole was added to the antibiotherapy treatment. Subsequent control cranial MRIs revealed an abscess, and vancomycin was added to the treatment. The patient had a heightened startle response to sound and a once daily dose of sertraline of 100 mg was also added. He was discharged from the neurology clinic with left hemiplegia, but was conscious, alert, oriented, and cooperative. The patient underwent physical therapy and an exercise program at an external center for nearly 3 months, after which he returned to the outpatient clinic with the complaint of ongoing weakness on the left side.

The patient was fully conscious, oriented, and cooperative. He could respond to single and multiple commands. He had good sitting balance, but could not lie down without assistance. He could stand, but could not support his full weight while standing. When he attempted to walk, inversion of the left foot was observed, as well as a distinct genu recurvatum deformity. The Brunnstrom staging score^[4] for the left side was shoulder: 2 points, hand: 1 point, and lower extremity: 2 points. Right side lower, and upper extremity muscular strength was intact. The Modified Ashworth Scale^[5] indicated that the left upper and lower extremities had grade 2 spasticity. The deep tendon reflexes of the left side were hyperactive, and more than 10 clonic convulsions of the Achilles tendon were observed. He had a restricted right gaze (total visual loss). A sulcus sign test was positive, though the range of joint mobility was restricted. Clinical scales were used to determine the severity of the palsy. The patient scored 25 points out of a possible 30 on a Mini-Mental State Test.^[6] His Barthel Index of Activities of Daily Living^[7] score was 25 out of a total of 100 points, which indicated that he was extremely dependent on a caregiver.

Routine biochemical and hematological analyses performed during his hospitalization did not detect any abnormalities. A once daily dose of 6 mg tizanidine were increased to 8 mg, and then to 10 mg. Botox injections to the upper and lower extremities were administered under ultrasonographic guidance. Since the John Cunningham (JC) virus had previously been detected, the department of infectious diseases was consulted. Samples sent for examination of anti-HIV and viral hepatitis markers all yielded negative results. Since most people test positively for the JC virus antigen, it was discarded from consideration as an etiological agent.

The patient was included in a physical therapy and exercise

program that included mat exercises, weight bearing while standing, walking in parallel bars, and active assisted joint range of motion exercises for the upper and lower extremities. Neuromuscular electrical stimulation was applied on the left deltoid, wrist extensor, and tibialis anterior muscles. Mirror therapy to assist with recovery of manual function was performed for 30 minutes every day. His physical examination findings at discharge were a Brunnstrom score for the left side of shoulder: 2 points, hand: 1 point, and lower extremity: 2-3 points. A marked regression in spasticity was not observed after the application of Botox (Allergan plc, Dublin, Ireland). His Barthel Index score was 30 points. When he was discharged, he could walk in the parallel bars with assistance.

Discussion

Encephalitis is an acute inflammation of the central nervous system parenchyma characterized by fever, headache, behavioral changes, and drowsiness. Encephalitis may be caused by viral, bacterial, or parasitic infections; vasculitides; Behçet's disease; Whipple disease; collagen vascular diseases; paraneoplastic syndromes; or drug reactions.^[2, 8] The most frequent cause of sporadic encephalitis is HSV type 1. HSV characteristically affects the temporal lobes and may lead to hemorrhagic necrotizing encephalitis.^[9]

Etiological agents should be investigated in a patient presenting with manifestations of encephalitis. Recent travel to epidemic regions, vaccination status, profession, immunosuppressive causes (posttransplantation immunosuppressive drug treatment, or acquired immune deficiency), transfusion of blood or blood products, and exposure to insect or other animal bites should be explored. The associated clinical symptoms are also important: the patient may present with fever, headache, seizures, neurological deficit, or coma.

There are several methods that may be used in the diagnosis of encephalitis. Generally, nonspecific hematological tests, detailed biochemical and coagulation tests, and a chest X-ray are performed initially, followed by tests seeking to identify the specific agent. Body fluid samples may be cultured, biopsy samples may reveal an antigen, and nucleic acid amplification or histopathological analyses may be performed. As was the case with our patient, frequently CSF samples obtained with LP may be used. Polynuclear pleocytosis can be detected in an early phase of encephalitis, and later mononuclear pleocytosis can be observed. If it persists, the presence of West Nile Virus should be suspected. The CSF protein level rises slightly to moderately, and the presence of erythrocytes indicates hemorrhagic encephalitis. Giemsa staining may display pathogenic agents. In acute disseminated encephalomyelitis, which

is frequently observed in children, glycemic levels are normal, but increased levels of lymphocytic pleocytosis and protein are seen.^[2, 10, 11]

In cases where an LP cannot be performed (e.g., papilledema), imaging modalities may be helpful. In particular, MRI, which has a high level of sensitivity and specificity, may be useful.^[12] Hypointense edema and hemorrhagic areas can be seen in the temporal lobes on T1-weighted images in HSV encephalitis, the most common form. However, these manifestations are visible at an advanced stage of the disease. Before the onset of these signs, HSV positivity may be detected using a PCR test.^[13] EEG may illustrate cerebral dysfunction. In the early phase of HSV encephalitis with temporal lobe affinity, epileptic discharges may be detected.^[9, 11]

As in this case, HSV is the most frequent etiological agent of sporadic encephalitis, and type 1 is responsible for 95% of cases. It has a high mortality rate, and an annual incidence of 2 to 3 cases per million people.^[14] Following a prodromal period with fever and headache, clinical presentation may include neurological symptoms, such as seizures, behavioral changes, motor deficits, stupor, or coma.^[15] Currently, the gold standard diagnostic method is identification of HSV DNA using PCR.^[16] Acyclovir is the best therapeutic agent.^[17] In our case, *Burkholderia mallei* positivity required the addition of trimethoprim/sulfamethoxazole. Glanders is an infectious disease caused by the bacterium *Burkholderia mallei*. It primarily affects horses, but it can also affect mules, donkeys, and other animals, and, occasionally, humans. In human beings, it begins with skin and mucosal ulcers, and progresses with lymphangitis and sepsis. At present, it is rare in humans, and laboratory infections are the only source of the disease.^[18] The JC virus, a type of human polyomavirus, detected in the CSF culture of our patient was first isolated in patients with Hodgkin's lymphoma.^[19] The patient frequently presents with manifestations of progressive multifocal leukoencephalopathy (PML). The JC virus antibody is seen in 85% of cases with an asymptomatic primary infection. The latent infection becomes activated secondary to impaired cellular immunity. Currently, JC virus infection is also important because it impairs immunity secondary to HIV infection. In addition to hematological malignancies, organ transplantation, and immunomodulatory drugs used in the treatment of autoimmune diseases may lead to the emergence of PML secondary to JC virus infection.^[20] Despite the presence of antigen positivity in our patient, no immune deficit that would lead to activation of the JC virus was observed. No specific treatment is available today for the JC virus. However, addressing the underlying immune deficiency is important. Appropriate treatment of HIV and changing immunosuppressive drugs may be an applicable approach.^[21, 22]

Stroke is a frequent cause of mortality and morbidity. Hypertension, diabetes mellitus, dyslipidemia, and obesity are important risk factors, yet it should not be forgotten that while some viral infections may also induce stroke as a result of the development of vasculopathy. A review of the literature indicated that the varicella zoster virus (VZV), HIV, and cytomegalovirus have been associated with stroke.^[23] However, only shown VZV has thus far been clearly shown to induce vascular damage in the intracerebral arteries.^[24]

The objective of this case report was to focus attention on both the clinical manifestations of encephalitis and the rehabilitation process, in particular. Especially given the influx of immigrants to our country, which could increase the incidence observed, every physician should have adequate knowledge to be able to establish an appropriate diagnosis and provide suitable treatment and referral to other centers, as needed.

Disclosures

Informed consent: Written informed consent was obtained from the patient for the publication of the case report.

Peer-review: Externally peer-reviewed.

Conflict of Interest: None declared.

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