Neurological Sciences (2019) 40:627–629
https://doi.org/10.1007/s10072-018-3607-8

LETTER TO THE EDITOR

Coxsackievirus B1 resulting acute flaccid myelitis with entire spinal cord lesion: a case report

Keun Tae Kim[1] · Yong Won Cho[1]

Received: 17 April 2018 / Accepted: 11 October 2018 / Published online: 19 October 2018

Keywords Acute flaccid myelitis · Coxsackievirus B1 · Polio-like · Magnetic resonance image

Dear Editor,

Coxsackievirus is a ribonucleic acid virus belonging to the *Picornaviridae* family and the Enterovirus genus. It can be identified through a diagnostic real-time polymerase chain reaction. So far, more than 20 serotypes in group A and 6 serotypes in group B have been recognized. The majority of the patients in group B present minor symptoms such as fever, headache, and diarrhea [1]. Acute flaccid myelitis refers to a polio-like illness defined by acute flaccid limb weakness with a gray matter lesion of the spinal cord appearing in the spinal magnetic resonance image [2]. Hereby, we report a case of whole gray matter myelitis of the entire spinal cord resulting in acute flaccid myelitis with an identified coxsackievirus B1 infection.

Case report

A 60-year-old woman with neither medical history nor recent immunization came to the emergency department with high fever and confusion. She felt the same as usual until 10 days prior, when febrile sensations and diaphoresis developed. Upon initial examination, her body temperature was 38.7 °C. She had stiffness in her neck and a positive Kernig's sign, clear lungs, and intact oropharynx. There was no heart murmur, no lymphadenopathy, and no skin lesions, e.g., rash or crust. She was alert, but disoriented regarding time, place, and person, and could only perform one-step commands. There was no definite motor weakness, although a precise examination was unavailable due to the patient's confusion and poor cooperation. Upon initial laboratory test, C-reactive protein was slightly elevated: 0.71 mg/dL (< 0.5 mg/dL). The cerebrospinal fluid analysis revealed pleocytosis of 360/HPF, predominantly lymphocytes (95%); raised cerebrospinal fluid protein (138.4 mg/dL); and normal cerebrospinal fluid sugar (69 mg/dL). The electroencephalography showed background theta slowing. We assessed that the cerebrospinal fluid profile suggested infection. Considering viral encephalitis, we prescribed intravenous acyclovir. The initial cerebrospinal fluid and blood cultures were found to be negative. Also, Lyme disease, dengue fever, poliovirus, cytomegalovirus, adenovirus, herpesvirus, Epstein-Barr, and West-Nile virus tests were all confirmed as negative. On the eighth day of hospitalization, her orientation was gradually restored, but she complained of difficulty voiding, paraplegia, and painful sensations on her whole body. An indwelling Foley catheter was inserted to drain 800 mL of urine. Her muscle tone was reduced in all extremities, deep tendon reflexes were absent, and Babinski's sign was mute on both sides. Although we confirmed that she became aware of paraplegia on the eighth day and could lift her legs by herself until the day before, there was a lack of information to specify the exact onset time of paralysis due to the patient's inability to cooperate. As we cannot exclude progressive paralysis, the onset of paralysis may be earlier.

We did not consider Pleconaril as it is not available in South Korea. We prescribed intravenous immunoglobulin when the nerve conduction test suggested motor polyneuropathy. Spine magnetic resonance imaging showed an extensive whole gray matter lesion of the entire spinal cord (Fig. 1a–e). Diagnosis of acute flaccid myelitis was established—acute limb weakness and magnetic resonance imaging showing a spinal cord lesion of gray matter in multiple segments [2]. As paraplegia progressed to become quadriplegia on the fifteenth hospital day, we prescribed an intravenous steroid pulse for 5 days. One week later, she was

✉ Keun Tae Kim
6k5upa@gmail.com

[1] Department of Neurology, Keimyung University Dongsan Medical Center, 56 Dalseong-ro, Jung-gu, Daegu 41931, South Korea

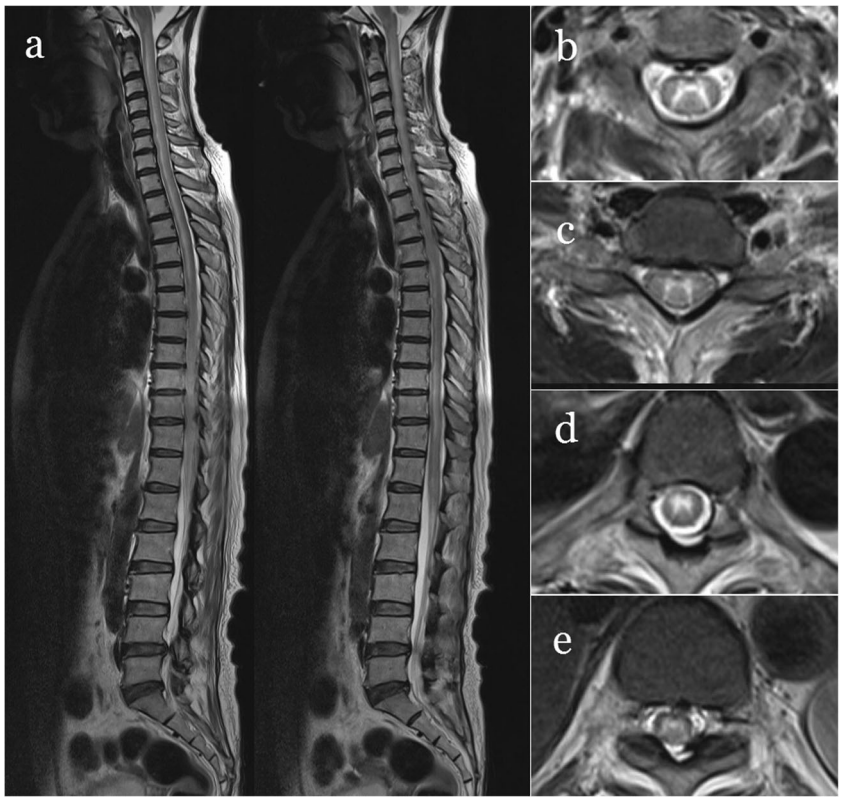

Fig. 1 Magnetic resonance imaging of spinal cord. Two consecutive T2 fast-spin echo sagittal images demonstrate diffuse longitudinal high signal intensity whole spinal cord from the cervicomedullary junction (**a**). Transverse images demonstrate high signal intensity of the whole gray matter of the C2 level (**b**); C7 level (**c**); T5 (**d**); and T12 level (**e**)

afebrile and motor ability of her upper extremities was restored. The second cerebrospinal fluid study performed on the eighth day of hospitalization revealed 16-folds increased coxsackievirus B1 through polymerase chain reaction. Although pain with dysesthesia below T5 dermatome and flaccid paraplegia remained, she could sit alone in bed and eat by herself upon discharge.

Coxsackievirus can cause diseases among a wide spectrum, such as meningitis, myocarditis, hepatitis, hand-foot-mouth disease, sepsis, severe neonatal illness, and in some cases death [1–4]. In previous reports, coxsackievirus A5 and 9 and coxsackievirus B2 to B5 were recognized as the cause of meningitis and acute transverse myelitis [1–5]. However, there is no report of coxsackievirus myelitis involving the entire spinal cord; moreover, there are few reports of significant infections caused by coxsackievirus in immunocompetent adults; almost all of the coxsackievirus B1 patients had been children or neonates [1, 3, 4]. In this case, the virology test through polymerase chain reaction revealed coxsackievirus B1, which was likely the cause of the whole gray matter lesion of the entire spinal cord.

Some authors suggest immunotherapy including steroids, intravenous immunoglobulin, and plasma exchange for acute flaccid myelitis [5]. However, as virology tests are often regarded as not cost-friendly, clinicians may take a passive approach rather than a more proactive one. In this case, the poor outcome of myelitis can be due to the late initiation of immunotherapy as the time of the first infusion of intravenous immunoglobulin was 8 days after admission, and acute flaccid myelitis was considered on the following day. Although there have been no verified treatments for acute flaccid myelitis thus far, early consideration of acute flaccid myelitis may lead to early initiation and active trial of immunotherapy, rather than using unnecessary treatments (e.g., acyclovir) and expect better prognosis.

Compliance with ethical standards

Ethical approval For this type of study, formal consent is not required.

Conflict of interest The authors declare that they have no conflict of interest.

References

1. Kim H, Kang B, Hwang S, Hong J, Chung J, Kim S, Jeong YS, Kim K, Cheon DS (2013) Molecular characteristics of human coxsackievirus B1 infection in Korea, 2008-2009. J Med Virol 85: 110–115. https://doi.org/10.1002/jmv.23359
2. Messacar K, Schreiner TL, Van Haren K, Yang M, Glaser CA, Tyler KL, Dominguez SR (2016) Acute flaccid myelitis: a clinical review of US cases 2012-2015. Ann Neurol 80:326–338. https://doi.org/10.1002/ana.24730
3. M. Angez, S. Shaukat, R. Zahra, M.M. Alam, S. Sharif, A. Khurshid, Y. Arshad, M. Suleman, G. Mujtaba, S.S.Z. Zaidi (2017)

Characterization of group B coxsackieviruses isolated from non-polio acute flaccid paralysis patients in Pakistan: vital assessment before polio eradication. Epidemiol Infect 145:2473–2481. https//doi.org/https://doi.org/10.1017/S0950268817001522

4. Moore M, Kaplan MH, McPhee J, Bregman DJ, Klein SW (1984) Epidemiologic, clinical, and laboratory features of Coxsackie B1-B5 infections in the United States, 1970-79. Public Health Rep 99:515–522
5. S.E. Hopkins (2017) Acute flaccid myelitis: etiologic challenges, diagnostic and management considerations. Curr Treat Options Neurol 19:48. https//doi.org/https://doi.org/10.1007/s11940-017-0480-3