OLGU SUNUMU CASE REPORT

DOI: 10.31609/jpmrs.2017-57576

Treatment Resistance in Hypoxic Brain Injury Caused by Cardiopulmonary Arrest Due to an Allergic Anaphylaxis: Spasticity, Myoclonus or Both?

Alerjik Anafilaksiye Bağlı Kardiyopulmoner Arrest Sonucu Oluşan Hipoksik Beyin Hasarında Tedavi Direnci: Spasitisite mi, Miyoklonus mu Yoksa Her İkisi de mi?

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Received: 07.08.2017
Accepted: 09.10.2017
Available online: 23.11.2018

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ABSTRACT Anaphylaxis is an acute and life threatening reaction and often results death. With medical developments, patients are more likely to survive after anaphylactic cardiopulmonary arrest. This leads to an increase in patients with hypoxic brain injury (HBI) who are in serious need of rehabilitation. Both hypoxic brain injury and cardiopulmonary arrest can cause many complications. Chronic post-hypoxic myoclonus, also known as Lance and Adams syndrome (LAS), is a rare condition that occurs after successful cardiopulmonary resuscitation. Here, we present a case with HBI that complicated by spasticity and LAS because of both it is a rare condition and effected to rehabilitation process.

Keywords: Anaphylaxis; hypoxic brain injury; spasticity; Lance and Adams syndrome

ÖZET Anafilaksi akut, yaşamı tehdit eden bir durumdur ve sıklıkla ölümle sonuçlanır. Tıbbi gelişmeler ile günümüzde anafilaksiye bağlı kardiorespiratuar arrest sonrasında hastalar daha fazla yaşatılabilmektedir. Bu durum beraberinde ciddi rehabilitasyon ihtiyacı olan hipoksik beyin hasarlı (HBH) hastaların artışını getirmektedir. Hipoksik beyin hasarının kendisi ve oluşan kardiyopulmoner arreste bağlı pek çok komplikasyon gelişebilmektedir. Lance ve Adams Sendromu (LAS) olarak da bilinen kronik post-hipoksik myoklonus, başarılı bir kardiyopulmoner resustasyondan sonra oluşan nadir bir durumdur. Burada spastisite ve LAS sendromu ile komplike olan HBH'lı hasta, nadir bir durum olması ve rehabilitasyonu etkilemesi nedeni ile sunulmuştur.

Anahtar Kelimeler: Anafilaksi; hipoksik beyin hasarı; spastisite; Lance ve Adams sendromu

A combination of beta-lactamase inhibitors including amoxicillin/clavulanate potassium are antibiotics with a broad spectrum of bactericidal activity against and have been used widely in the treatment of a broad range of clinical infections. The early allergic reactions are usually associated with the use of penicillin that is the most common cause of severe allergic drug reactions.[1,2] The repeated use of penicillin is estimated to produce an eight-time increase in the incidence of allergic reactions as compared to those for first-time users.[2,3]

Anaphylactic shock usually can lead to death but recent advances in healthcare have contributed to longer survival after cardiopulmonary arrest due to anaphylaxis. This results in increase in the incidence of hypoxic

brain injury, which is the classical example of condition after cardiopulmonary arrest. Hypoxic brain injury (HBI) can cause cognitive impairments and physical complications e.g. spasticity, contractures, loss of strength.[4] In addition, cardiopulmonary arrest can lead to rare complications such as post-hypoxic myoclonus, defined as Lance and Adams Syndrome (LAS) which can be confused with spasticity in patients with HBI during rehabilitation process.[5]

Here, we present a case of HBI developed after cardiopulmonary arrest due to anaphylaxis, and complicated by treatment resistant spasticity and LAS.

CASE REPORT

Forty-four year old man with the diagnosis of HBI complicated by tetraparesis and spasticity, was consulted and transferred to inpatient rehabilitation program.

Sixty-three days previously the patient had experienced a hypoxic insult caused by cardiopulmonary arrest due to an anaphylactic reaction to antibiotic therapy. He had suffered cardiopulmonary arrest in the emergency room, and he had been resuscitated and intubated. After 7 minutes of resuscitation, patient had been connected to the ventilator and transferred directly to an intensive care unit where he stayed for 42 days, after that, he was followed in neurology clinic for 21 days.

Upon admission to emergency department, his Glasgow Coma Scale score (GCS) was 3/15 which improved to 12/15 on 33rd day. He had been weaned from ventilator on 37th day, and on the 63rd day, consulted and transferred to rehabilitation clinic with GCS score of 12/15, for the rehabilitation of tetraparesis and spasticity due to HBI.

He had no history of endocrine/metabolic disease, alcohol use or smoking. He had no family history of the disease.

On examination, the patient was awake, alert, oriented to person and place but he had difficulty following complex commands and repeating short phrases. He was agitated, yelling at people, but making no sense. Cognitive function evaluated as confused and inappropriate behavior.

Musculoskeletal assessment revealed that he had no active movement of neck, upper or lower extremities and had no ability to sit independently. He had apparent spasms occurred on and off all day long, triggered with all activities such as moving, touching and speaking, affecting neck, trunk, all upper and lower extremities. He had flexion posture in upper and lower extremities bilaterally due to these spasms. The degree of spasticity was assessed by modified Ashworth scale (MAS); MAS scores were as follows; neck and back muscles 2/4, upper extremities 2-3/4, lower extremities 2-4/4 bilaterally. Deep tendon reflexes were increased bilaterally in upper and lower extremities.

Laboratory data were normal. Diffusion magnetic resonance imaging (MRI) revealed T2 weighted signal increase in basal ganglia and thalamus as well as in the white matter, of the lateral ventricle body and decreased blood flow in the gray and white matter junction adjacent to this area. Diffusion-weighted MRI series revealed chronic ischemic changes.

Agitation disrupting rehabilitation process was treated with propranolol hydrochloride 40 mg/day. Spasticity managed with oral baclofen 30 mg/day, which was started initially and increased up to 100 mg/day, and oral tizanidine 18 mg/day. Cold pack, transcutaneous electrical nerve stimulation and stretching exercises were also performed for spasticity therapy respectively.

He underwent spasticity therapy for fifteen days, without changes in his spastic pattern. He was treated with the injection of Botulinum toxin type A for the spasticity of neck and upper extremity muscles. Patients' lower extremity ranges of motions (ROM) were evaluated under anesthesia using midazolam infusion to differentiate spasticity and contractures. Then, the patient underwent capsular release surgery to the left knee and posterior tibial tendon release surgery to his right ankle for ankle and knee ROM limitations in orthopedic clinic. Twenty days after the day of surgery, he was admitted to our clinic again for post-operative rehabilitation. At the time of second admission, patient was still dependent at all levels of bed mobility and he had no neck control. Spasticity levels did not change.

Because of the patient had treatment-resistant-spasticity, he underwent bolus test dosing with 50, 75, or 100 micrograms of intrathecal baclofen at 24 hours intervals but he was refractory to intrathecal baclofen at a dose of 100 mg/d, with no side effect. Physical therapy modalities were again performed simultaneously as done during the first hospitalization. As there was no improvement in spasticity scores of patient after 45 days (25+20) of therapy, EMG and EEG were performed under guidance of neurology consultation. There was no result of any lower motor neuron involvement in EMG, and EEG was normal. Therefore these spasms were ascribed to bilateral generalized post-hypoxic myoclonus concomitant to HBI induced spasticity. He was started on clonazepam 4 mg/day, levetiracetam 1000 mg/day as well as quetiapine 200 mg/day. Spasms were improved partially, and spasms frequency decreased. Dosage of clonazepam and levetiracetam was not increased due to conspicuous muscle weakness occurred. He was discharged with a home exercise program at a wheelchair independent level.

DISCUSSION

Rare cases of hypoxic brain injury from allergic anaphylaxis, often resulting in death, have been reported.[6] Previous history of allergy, even skin rash as in our case, increases the death incidence up to 50-70%.[7,8]

Hypoxic brain injury (HBI) is associated with significant and persistent cognitive impairment depending on the severity of hypoxia and neuronal damage. Patients with severe HBI frequently exhibit agitated behavior, personality changes causing cognitive disability.[6] In accordance with the literature, our case also exhibited high levels of agitation that negatively affected rehabilitation process.

Several studies have been reported that the functional and cognitive disabilities are higher in patients with HBI compared to patients with TBI as to be related to mechanism of injury; TBI often causes a disruption of axonal integrity, as opposed to direct neuronal cell death in HBI that leads to early, rapid and severe spasticity, and contracture formation.[6-8] Our case had also severe and treatment-resistant spasticity leading to joint contractures in as early as two months, and although all known spasticity treatments were applied to him, he was still at bed level due to the spasms. Accordingly, our case with treatment-resistant spasticity along with the spasm triggers such as moving, touching and speaking, necessitate re-reviewing of patient, which led us to think of the diagnosis of LAS, and patient gained control of sitting independently after LAS treatment.

Lance and Adams Syndrome, also known as chronic post-hypoxic myoclonus, is a rarely diagnosed state that occurs days or weeks after successful cardiopulmonary resuscitation, and characterized with action myoclonus in awaked patients.[9] It was first described in the 1963, and approximately, 150 cases of this syndrome have been described worldwide. The emotional and sensorial stimulus can trigger intention or action myoclonus especially in the distal extremities, and even bilateral generalized multifocal myoclonus can be seen in LAS. During sleep and rest, spasm and cramps disappear. In our case, all joints had full ROM except knee and ankle joints, and spasms disappeared under anesthesia. In addition, there was a spasticity and complicated contractures in our case unlike those reported in literature.

The diagnosis of LAS is made by clinical examination. MRI and CT are not specific to it, but recent studies revealed that the LAS is seen more frequently after subcortical lesions including thalamus, as observed in our case.[6,10] EEG has frequently been reported as unremarkable in previous studies consistent with our case study.

Clonazepam and levetiracetam are the main drugs for the treatment of LAS, especially combination treatments have been reported to be effective for myoclonic jerks.[6] Although we have treated the patient with clonazepam and levetiracetam, which regulates neurochemical abnormalities, our case showed only a partial recovery, which can be explained by delayed diagnosis and treatment of LAS. In studies it has been shown that the drugs are the most effective and LAS has a better prognosis if treated early.[6] In addition, the occurrence of dose-limiting muscle weakness may have caused the partial recovery.

CONCLUSION

In conclusion, concomitant complications can rarely be present in cases with HBI caused by cardiopulmonary arrest. Especially, muscle spasms may be a component of LAS rather than spasticity in the presence of treatment-resistant spasticity, LAS may be overlooked, which may also affect the prognosis of patient.

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