Case Report
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Extrapyramidal Dysfunction as a Consequence of Hypoxic Brain Injury

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Objective: A case of parkinsonism after hypoxic ischemic encephalopathy is reported. **Methods and Results:** A 34-year-old woman presented parkinsonism due to hypoxic-anoxic encephalopathy. The manifestation of parkinsonian syndrome was more present axially, than in the limb. She has also experienced cognitive deficits. The brain Magnetic Resonance Imaging (MRI) studies have shown abnormalities in putamen, caudate nucleus, globus pallidus and cerebral cortex. **Conclusions:** Diversity of clinical features may be depend on different neuropathological damage and neurotransmitter disbalance within the basal ganglia after a hypoxic-ischemic arrest, as seen in our patient. Therefore, understanding of all factors that contribute hypoxic brain injury is important to determine the clinical outcome and further management of patients.

Keywords: Hypoxic Brain damage, Parkinsonism, Globus Pallidus

Introduction

Post anoxic or hypoxic encephalopathy is not a common clinical condition. Cerebral hypoxia can be caused by any event that affects the brain's ability to receive and process oxygen. Basal ganglia, especially the globus pallidus (GP), are highly vulnerable to generalized cerebral anoxia or hypoxia [1]. Cerebral hypoxia typically produces lesions of the globus pallidus and striatum, which may result not only in akinetic rigid syndrome but also in other movement disorders, including dystonia, chorea, tics, athetosis, tremor, and myoclonus. Although these conditions can develop soon after the hypoxic brain injury, they often develop months or years after the injury. Unfortunately, these conditions appear less responsive to pharmacologic treatment than primary

parkinsonism (i.e., Parkinson's disease) and idiopathic dystonia [2]. Herein, we report a case with a parkinsonism secondary to hypoxic-anoxic encephalopathy.

Case Report

A healthy, 34-year-old woman with secondary education (10 years) was admitted to the maternal hospital for childbirth, but she had severe hemorrhaging after delivery and underwent a hysterectomy. During the hysterectomy, she developed sudden cardio-respiratory arrest. She remained in coma for over 10 days, after which she gradually recovered consciousness over a period of several weeks. The first CT, performed 2 days after the insult, showed diffuse brain edema without focal abnormalities (Figure 1). On the 7th day of the coma, she had several epileptic seizures and was successfully treated with antiepileptic drugs. She remained in coma for over 10 days, after which she gradually recovered consciousness over a period of several weeks. The second CT, performed 3 weeks after the insult, showed more prominent small, bilateral, low density lesion in basal ganglia on the left side (Figure 2).

Our patient was treated at the maternity hospital for 50 days then transferred to the neurology department for continuous treatment. At admission to the neurology department, she could understand referred speech and perform simple tasks, but her speech was severely dysarthric and limited to performing simple neuropsychological test. On neurological examination, she had markedly noticeable bradykinesia and rigidity more on the left side. Axial bradykinesia was more prominent than in the extremities. She had severe resting, kinetic and intention tremor, which made her unable to perform any movement tasks, and she also had noted head tremor. She was unable to stand and walk, even with support, because of severe postural instability.

Brain MRI, performed 1.5 months after the insult, showed a bilateral hyper intense signal of the putamen, caudate nucleus, and globus pallidus with laminar necrosis in the superior frontal cortex and insula (Figure 3 and 4). Treatment with 2.0 mg/day of clonazepam, 4.0 mg/day of trihexyphenidyl and 250/25 mg/day of levodopa/carbidopa resulted in mild improvement of gait, hand tremor, and bradykinesia.

After discharged from the hospital, our patient took medication for one month and attended a long-term rehabilitation program. An examination two months later revealed that her speech, gait, and tremor had improved, but she still had severe postural instability due to axial bradykinesia. She still could not stand or walk without support and was unable to perform the simple activities of daily living. An examination five months later revealed that the action and rest tremor had almost disappeared, but the mild left hand rest tremor was still noticeable during contralateral hand movement. There was mild rigidity and bradykinesia on the left limbs. Her speech and gait had improved, but she still had postural instability. She could stand but could not walk without support. Her Mini-Mental State Examination (MMSE) score was 20 (normal range: 25-30). With regard to cognitive-specific domains, language and visuospatial deficits were detected on the MMSE, but orientation, memory,

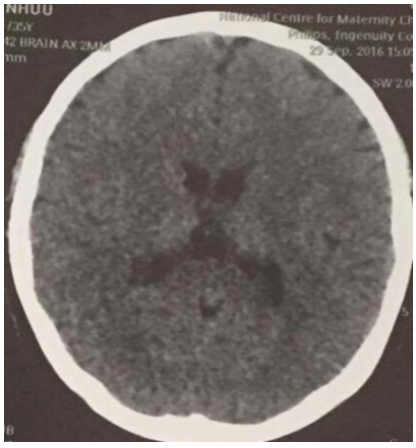

Figure 1. Initial CT scan, 2 days after the insult. Diffuse brain edema without focal abnormalities.

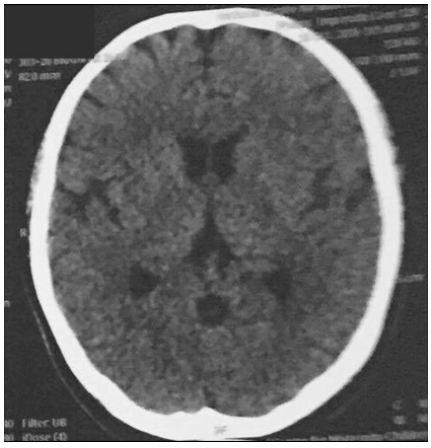

Figure 2. 2nd CT, 3 weeks after the insult. Small, bilateral, low density lesion in basal ganglia.

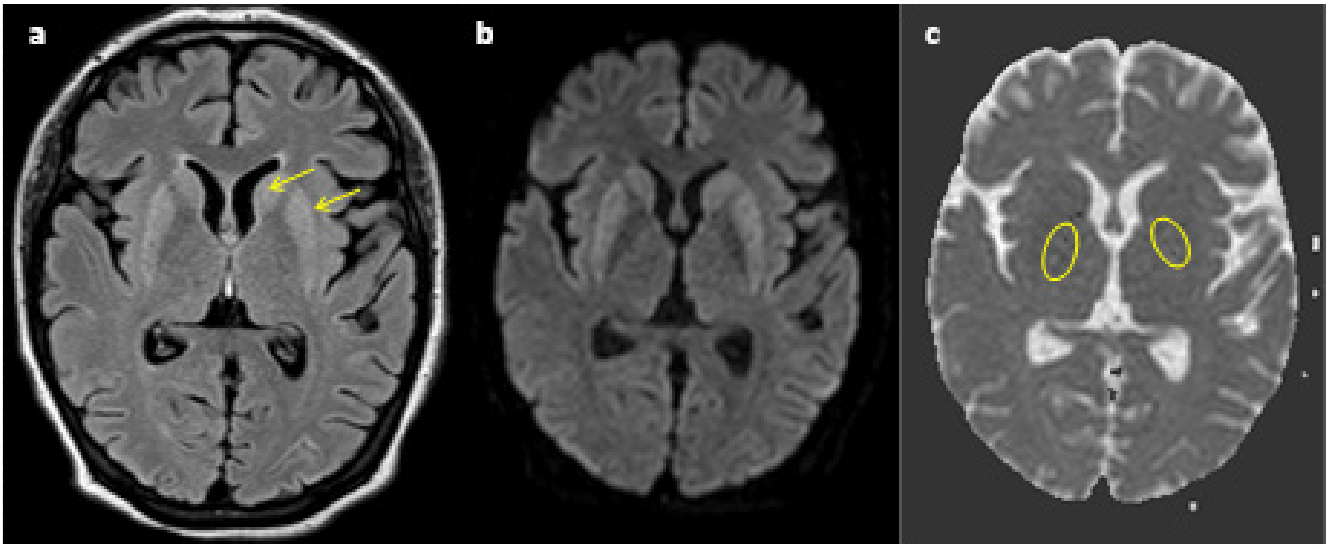

Figure 3. FLAIR (a) and DWI (b) images, 1.5 months after the insult, shows abnormal signal in the putamen and caudate nuclei; ADC map (c) shows elevated values in both globus pallidus, but greater on the right side, indicating greater involvement.

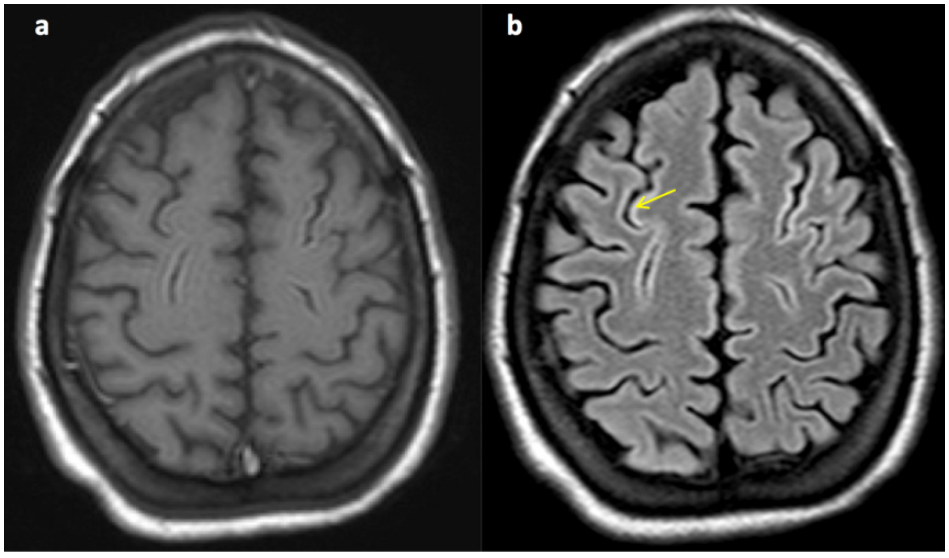

Figure 4. T1W (a) and FLAIR (b) images, 1.5 months after the insult, show laminar necrosis in the bilateral superior frontal gyri and insula.

attention, and executive function were relatively intact. Her language impairment included disability of reading (alexia), calculating (dyscalculia), and writing (agraphia).

Discussion

Herein, we report a case with Parkinsonism due to hypoxic-anoxic encephalopathy. Clinical sequelae of hypoxic anoxic injury of the brain, which can extend for over a period of weeks after the initial injury, are pyramidal, extrapyramidal dysfunction and cognitive deficits [3]. A history of the anoxic hypoxic event, physical and neurological exams, and brain CT or MRI scans should be used for diagnosis.

MRI studies of patients soon after hypoxic brain injury have shown abnormalities in the basal ganglia, the cortex, the hippocampus, and the cerebellum [4-6].

On neurological examination, our patient had severe akinetic rigidity and resting and kinetic tremor. In our patient, the manifestation of parkinsonian syndrome was more present axially, than in the limb, and she had severe postural

instability and walking disability. Her speech was monotonous and dysarthric. The brain MRI showed bilateral hyper intense signals in the putamen and caudate nucleus, and the ADC map showed elevated values in both globus pallidus, but greater on the right side. The symptoms of our patient were attributable to the damage of the motor system, including putamen and globus pallidus, as an adverse effect of the hypoxia. However, the sensory system of our patient was intact, which is consistent with previous reports that show that the predominant clinical residues after hypoxic injury of the brain are motor disorders [7-10].

Many studies have reported that the basal ganglia, especially the globus pallidus, are highly vulnerable to generalized cerebral anoxia/hypoxia. Feve AP and Fenelon G have also suggested that the globus pallidus plays an important role in controlling axial motion. In their study, four patients were examined due to axial motor impairment after hypoxic injury, and MRI scans found lesions of the globus pallidus in all four cases [8]. It is also generally accepted that parkinsonism or axial motor problems are commonly related to the pallidal lesions, and dystonia is related to lesions in the putamen [3].

Interestingly, in our case, abnormalities in the globus pallidus were less apparent on MRI scan, but our patient developed parkinsonian syndrome. We do not know the exact reason for this, but these clinical features may be depend on the nature of the hypoxic event and the pathological pattern of neuronal activity in the nervous system [3]. It has also been noted that developing dystonic or parkinsonian syndrome after a hypoxic/anoxic insult is determined by the patient's age at the time of the hypoxic/anoxic insult [13]. It has been known that the onset of dystonia may be delayed by months and years after the hypoxic insult [7, 13]. In our case, parkinsonian syndrome developed soon after the event and gradually showed improvement after treatment. However, we must continue clinical observation of our patient for any dynamic changes over time.

In addition, acute and chronic cognitive impairment is a common symptom in patients after a hypoxic anoxic event [11]. This is most likely related to the vulnerability of the CA1 area of the hippocampus and cortical layers 3, 5, and 6 to the adverse effects of hypoxia or ischemia [12]. In our case, our patient experienced cognitive deficits, as shown by the MMSE score of 20 and the MRI finding of laminar necrosis in the bilateral superior frontal gyri and insula.

Finally, the diversity of clinical features may be dependent on different neuropathological damage and neurotransmitter disbalance within the basal ganglia after a hypoxic ischemic insult, as seen in our patient. It has been suggested that a pathophysiology of the hypoxic ischemic state is associated with cellular energy failure, acidosis, glutamate release, increases in intracellular calcium, and generation of free radicals, which lead to neuronal death and irreversible brain injury [14]. Therefore, greater understanding about the pathogenesis of hypoxic brain injury will help to determine clinical outcomes and long term management of patients.

Conflict of Interest

The authors state no conflict of interest.

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